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**University of Southampton**

Faculty of Environmental and Life Sciences

School of Psychology

**Sex/Gender Differences in Autistic Restricted and Repetitive Behaviours and  
Interests**

by

**Hannah Molly Ceri Edwards**

Thesis for the degree of Doctorate in Educational Psychology

June 2022



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Autistic females are often diagnosed less frequently than autistic males, despite many displaying similar levels of autistic traits to males. Explanations for this include diagnostic assessment tools not being sensitive enough to the manifestation of autistic behaviours in females, as well as higher levels of camouflaging behaviours in autistic females resulting in some autistic traits being missed when using observational or parent/carer/teacher report measures. Consequently, autism research is relatively unrepresentative of autistic females, especially those who report high autistic traits but do not have a clinical diagnosis. Taking a critical realist approach, the aim of this thesis is to explore sex/gender differences in the subdomain of the autism diagnostic criteria; restricted and repetitive behaviours and interests (RRBIs). Research at the broad construct level of RRBIs indicates that autistic females present with fewer RRBIs than autistic males, however the subdomain is large, with a range of narrow constructs within it that are different to each other (e.g., stereotyped behaviours, insistence on sameness, passionate interests, and sensory experiences), warranting deeper exploration at a fine-grained level. Firstly, a systematic review and meta-analysis was conducted to explore sex/gender differences at the narrow construct level of RRBIs in autistic children, adolescents, and adults. This indicated that autistic males presented with significantly more stereotyped behaviours and passionate interests than females. There was also a trend towards autistic females presenting with more sensory experiences. There was no significant sex/gender differences for insistence on sameness. Autistic females also appeared to hold different types of passionate interests to males. It was deemed important to include participants who self-identify as autistic, without a clinical diagnosis (but who report high autistic traits), in the empirical research project, in an attempt to include a group who has previously been excluded from many studies. Likewise, a self-report measure was chosen so that the subjective experiences of autistic individuals could be captured. The empirical research project explored sex/gender differences in two narrow constructs of RRBIs, insistence on sameness (IS) and repetitive sensory motor behaviours (RSMB), in a sample ( $n = 84$ ) of autistic (diagnosed and self-identifying) and non-autistic 16-25 year olds. Results indicated that autistic females self-report significantly more IS behaviours compared to autistic males and similar levels of RSMB to autistic males. These findings emphasise the importance of exploring RRBIs at a fine-grained level and raises the importance of professionals involved in identification of autism, for example educational practitioners, being aware of sex/gender differences in RRBIs, particularly how these may present in autistic females.



# Table of Contents

Table of Contents .....	i
Table of Tables .....	v
Table of Figures .....	vii
Research Thesis: Declaration of Authorship.....	ix
Acknowledgements.....	xi
Definitions and Abbreviations.....	xiii
<b>Chapter 1 Sex/Gender Differences in Autistic Restricted and Repetitive Behaviours and Interests- An Introduction.....</b>	<b>1</b>
1.1 Motivation for this research .....	1
1.2 Research study .....	2
1.3 Ethical considerations.....	3
1.4 Epistemological stance .....	4
1.5 Autism, gender, and intersectionality .....	5
1.6 Implications for Educational Psychology .....	6
1.7 Dissemination plan .....	7
<b>Chapter 2 A Systematic Review and Meta-Analysis of Sex/Gender Differences in Restricted and Repetitive Behaviours and Interests in Autistic Children, Adolescents, and Adults.....</b>	<b>9</b>
Abstract.....	9
2.1 Introduction .....	10
2.1.1 Sex/Gender Bias in Autism Referral, Diagnostic Criteria and Assessment Tools 11	
2.1.2 Broad and Narrow Constructs in Autism .....	12
2.1.3 Current study.....	14
2.2 Method .....	14
2.2.1 Search Strategy.....	14
2.2.2 Eligibility Criteria.....	15
2.2.3 Search Results.....	16
2.2.4 Data Extraction .....	17

## Table of Contents

2.2.5	Characteristics of Included Studies .....	18
2.2.6	Study Quality Appraisal .....	18
2.2.7	Data Synthesis Strategy .....	39
2.3	Narrative Synthesis .....	39
2.3.1	Stereotyped or Repetitive Motor Movements, Use of Objects, or Speech ....	39
2.3.2	Insistence on Sameness, Inflexible Adherence to Routine, or Ritualised Patterns of Verbal or Nonverbal Behaviour.....	40
2.3.3	Highly Restricted, Fixated Interests that are Abnormal in Intensity or Focus.	41
2.3.4	Hyper- or Hypo-Reactivity to Sensory Input or Unusual Interests in Sensory Aspects of the Environment.....	42
2.4	Meta-Analysis .....	42
2.4.1	Publication Bias and Heterogeneity .....	42
2.4.2	Stereotyped or Repetitive Motor Movements, Use of Objects, or Speech ....	43
2.4.3	Insistence on Sameness, Inflexible Adherence to Routine, or Ritualised Patterns of Verbal or Nonverbal Behaviour.....	44
2.4.4	Highly Restricted, Fixated Interests that are Abnormal in Intensity or Focus.	45
2.4.5	Hyper- or Hypo-Reactivity to Sensory Input or Unusual Interests in Sensory Aspects of the Environment.....	47
2.5	Discussion .....	48
2.5.1	<b>Implications for Practice</b> .....	52
2.5.2	Limitations.....	53
2.5.3	Conclusion.....	54
<b>Chapter 3</b>	<b>Are there sex/gender differences in how autistic young people, with and without a diagnosis, engage in restricted and repetitive behaviours and interests? .....</b>	<b>57</b>
3.1	Introduction .....	57
3.2	Method .....	60
3.2.1	Participants and Procedure.....	60
3.2.2	Measures.....	61
3.2.2.1	Demographics .....	61



3.2.2.2	Autistic Traits .....	61
3.2.2.3	Restricted and Repetitive Behaviours and Interests.....	61
3.2.3	Analytic plan .....	62
3.2.4	Community involvement.....	62
3.3	Results.....	64
3.3.1	Restricted and Repetitive Behaviours and Interests .....	64
3.4	Discussion.....	66
3.4.1	Implications for Practice .....	68
3.4.2	Limitation and Future Research .....	69
3.5	Conclusion.....	71
	<b>Appendix A Full Search Terms.....</b>	<b>73</b>
	<b>Appendix B Newcastle-Ottawa Quality Assessment Scale with adaptations .....</b>	<b>74</b>
	<b>Appendix C Summary of studies in the systematic review and meta-analysis.....</b>	<b>77</b>
	<b>Appendix D Funnel plots .....</b>	<b>91</b>
	<b>Appendix E Questionnaires.....</b>	<b>95</b>
E.1	The Autism Symptom Self-Report for adolescents and adults (Posserud et al., 2013).....	95
E.2	The Adult Repetitive Behaviour Questionnaire-2A (RBQ-2A) .....	95
	<b>List of References .....</b>	<b>101</b>



## Table of Tables

Table 1.	<i>Examples of broad/narrow constructs and associated behavioural exemplars based upon DSM-5 diagnostic criteria. ....</i>	13
Table 2.	<i>Rationale for inclusion and exclusion criteria. ....</i>	15
Table 3.	<i>Study and sample characteristics of all included studies. ....</i>	21
Table 4.	<i>Adaptations for autistic participants participating in research based on recommendations by Gowen et al. (2020). ....</i>	63
Table 5.	<i>Analysis of variance for RSMB .....</i>	64
Table 6.	<i>Analysis of variance for IS .....</i>	65
Table 7.	<i>Mean (SD) and range for age, RBQ-2A RSMB subscale score, and RBQ-2A IS subscale score for male and female young adults in the autistic and non-autistic groups.....</i>	65
Table 8.	<i>Full Search Terms for Systematic Review .....</i>	73



## Table of Figures

Figure 1.	PRISMA Flowchart.....	17
Figure 2.	Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of stereotyped behaviours. ....	44
Figure 3.	Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of insistence on sameness. ....	45
Figure 4.	Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of passionate interests.....	46
Figure 5.	Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of passionate interests with RBS-R Endorsed total mean scores only.....	47
Figure 6.	Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of sensory experiences. ....	48
Figure 7.	Funnel plot for restricted interests meta-analysis.....	91
Figure 8	Funnel plot for stereotyped behaviour meta-analysis.....	92
Figure 9	Funnel plot for insistence on sameness meta-analysis.....	92
Figure 10	Funnel plot for sensory meta-analysis .....	93



## Research Thesis: Declaration of Authorship

Print name: Hannah Edwards

Title of thesis: Sex/Gender Differences in Autistic Restricted and Repetitive Behaviours and Interests

I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

1. This work was done wholly or mainly while in candidature for a research degree at this University;
2. Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
3. Where I have consulted the published work of others, this is always clearly attributed;
4. Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
5. I have acknowledged all main sources of help;
6. Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
7. None of this work has been published before submission;

Signature: .....

Date:





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This thesis is dedicated to my mum. Although you weren't able to join me on this journey, I hope that I have made you proud.



## Definitions and Abbreviations

AD.....	Autistic Disorder
AdAS.....	Adult Autism Subthreshold
ADI-R .....	Autism Diagnostic Interview-Revised
ADOS/ADOS-2 .....	Autism Diagnostic Observation Schedule- 2
ADOS-G.....	Autism Diagnostic Observation Schedule-Generic
ANOVA.....	Analysis of variance
ASD.....	Autistic spectrum disorder
ASDI.....	Asperger Syndrome and high functioning autism Diagnostic Interview
ASDS .....	Asperger Syndrome Diagnostic Scale
ASRS .....	Autism Spectrum Rating System
ASSERT.....	Autism Symptom Self-Report Questionnaire
AQ .....	Autism Spectrum Quotient
BDI-2.....	Battelle Developmental Inventory- Second Edition
BISCUIT-Part 1.....	Baby and Infant Screen for Children with Autism Traits- Part 1
CAMHS.....	Child and Adolescent Mental Health Services
CARS-TV.....	Childhood Autism Rating Scale-Tokyo Version
CAT-Q .....	Camouflaging of Autistic Traits Questionnaire
CCX .....	Caregiver-child interactions
CI .....	Confidence interval
DASH.....	Diagnostic Assessment for the Severely Handicapped-Second Edition
DIF .....	differential item functioning
DSM-5.....	Diagnostic Statistical Manual- Version 5
DSM-IV-TR .....	Diagnostic Statistical Manual- Version 4- Text Revised
DQ .....	Developmental quotient- a numerical indicator of an infant's development across a range of psychosocial competencies (e.g., behavioural, communication, and intellectual development)

## Definitions and Abbreviations

EPs	.....	Educational Psychologists
F	.....	F-statistic
HFA	.....	High functioning autism
$I^2$	.....	I-squared
ICD-10	.....	International Classification of Diseases (10th ed.)
IS	.....	Insistence on sameness
IQ	.....	Intelligent quotient
M-CHAT	.....	Modified Checklist for Autism in Toddlers
$N$	.....	Total population
$n$	.....	Sample population
$\eta^2P$	.....	partial-eta squared
N-OQAS	.....	Newcastle-Ottawa Quality Assessment Scale
$p$	.....	p-value
PCA	.....	Principal component analysis
PDD-NOS	.....	Pervasive developmental disorder, not otherwise specified
PICO	.....	Population Intervention Comparison Outcome
PRISMA	.....	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
RBS-R	.....	Repetitive Behaviour Scale-Revised
RBQ-II	.....	Repetitive Behaviours Questionnaire- Second Edition
RBQ-2A	.....	Adult Repetitive Behaviour Questionnaire- 2
RRB	.....	Restricted and repetitive behaviours
RSMB	.....	Repetitive sensory motor behaviours
RRBIs	.....	Restricted and repetitive behaviours and interests
SCQ	.....	Social Communication Questionnaire
SD	.....	standard deviation
SEM	.....	Standard error mean
SENCO	.....	Special Educational Needs Coordinator
SeSS	.....	Sensory Sensitivity Scales

SMD.....	Standard mean difference
SPQ.....	Sensory Perception Quotient
SRS-2.....	Social Responsiveness Scale-Second Edition
SSP.....	Short Sensory Profile
SSQ.....	Sensory Sensitivity Questionnaire
<i>t</i> .....	T statistic
Q.....	Cochran's Q test
3di .....	The developmental, dimensional and diagnostic interview



# Chapter 1 Sex/Gender Differences in Autistic Restricted and Repetitive Behaviours and Interests- An Introduction

## 1.1 Motivation for this research

My journey to researching the experiences of autistic people began long before I consciously chose this path. By the time I was born, my older brother had already received his diagnosis of autism. Without me realising, I spent my entire childhood learning about autism, or at least, learning what autism meant for my brother and my family. During and after my undergraduate studies, I had the privilege of working with a variety of autistic young people and adults and I learnt how every autistic person's experience is different and yet, so frequently, their experiences are generalised and misunderstood by non-autistic people. When I started working as an Assistant Psychologist in a Child and Adolescent Mental Health Service (CAMHS), I learnt about how autism is assessed and diagnosed in the UK and I learnt about how different autistic young people present in a clinical context. However, over time, I became aware that something was missing in what we knew about autism, particularly autism and girls. The girls I met were often older and many didn't seem to fit with the diagnostic criteria (The Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition then The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition) used for autism. We were becoming aware of the female autism phenotype and camouflaging (Hull et al., 2020) and how this could be contributing towards fewer girls being referred for an autism assessment and, subsequently, fewer being diagnosed (and those who were, were diagnosed much later into their adolescence or adulthood). Autistic girls appeared to display more 'typical' social skills and were self-reporting a variety of strategies that helped them to interact with others. Less was known, however, about sex/gender differences in the second domain of autism; restricted and repetitive behaviours and interests (RRBIs). In my clinic work, it seemed that autistic girls generally displayed fewer RRBIs compared to males and I wondered whether this was contributing towards a proportion of girls not meeting the diagnostic criteria. I also noticed a difference in the types of RRBIs reported by girls and their families. I met girls with passions for animals, make-up, TV soaps, and popstars, but the intensity of these interests were difficult to identify within observational assessments. Furthermore, these interests often were not highlighted by parents/carers or teachers during developmental histories or questionnaires, resulting in "no intense interests" being coded in many diagnostic assessments. Similarly, I spoke with young autistic women who described a variety of restricted and repetitive

behaviours and sensory needs that were just not captured by the assessment measures we were using. We were not asking the right questions about RRBIs, especially when assessing girls.

Nearly six years later, when I started to consider my area of research for my thesis, I knew I wanted to explore sex/gender differences in autism. There remained a paucity of research specifically focussing on RRBIs, with the majority of research exploring the social communication and interaction domain of autism (see Wood-Downie et al., 2020 for a review of sex/gender differences in social interaction). Therefore, I believed that a systematic literature review and meta-analysis exploring the current research about sex/gender differences in RRBIs would be timely.

### **1.2 Research study**

Based on the conclusions of my systematic review, I identified a need to explore autistic RRBIs at a fine-grained, narrow construct, level in order to explore the research question ‘are there sex/gender differences in self-reported RRBIs between young people diagnosed autistic, young people who self-identify as autistic and non-autistic young people?’ Firstly, I made the decision to utilise a self-report outcome measure of RRBIs to more accurately capture the lived experiences of autistic individuals. Many previous studies used outcome measures that rely on parent/carer or observer reports, which can be hindered by subjectivity, interpretation bias, and being influenced by cultural expectations (Gal, 2011). Secondly, I recognised that, to overcome the under-representation of autistic females in research, my research project needed to include self-identifying autistic participants (particularly females), as these individuals were likely to be the exact population being missed by current autism research and, subsequently, diagnostic instruments.

At the development stage of my research, I was greatly influenced by Gowan et al.’s (2020) recommendations on how to conduct research studies with the autistic community. I ensured that all information about the study was presented in a clear way, alongside a video for those who benefited from additional visual and auditory forms of communication, as well as providing information about myself as a researcher. Central to the design of the research was the feedback and guidance I received from an autism consultant who acted as an ‘expert-by-experience’; they supported me to ensure that both the online survey and the accompanying participant information and debriefing documents were accessible to autistic individuals. I strongly believe that autistic consultants should be compensated for their time and expertise, so I ensured that the autism consultant was financially reimbursed for the two hours they spent reviewing documents, paying them their rate of £12.50 per hour. I would have liked to recruit more than



one autistic individual to act as a consultant for the research; however, unfortunately, my research budget could not accommodate this. If I were to conduct my research again, I would definitely be seeking to work collaboratively alongside autistic researchers so as to ensure that autistic individuals are included at different stages of the research process and in all decision making.

Following development, the online survey was advertised via social media platforms as well as being directly sent alongside an information letter to all post-16 institutions, universities, and Autism charities/organisations in the UK, with the request for the link to be shared amongst students and service users. A financial incentive was offered whereby participants could enter a prize draw by providing their email addresses, which were collected separately from the data collected on the survey to maintain anonymity. Upon clicking the link, participants were taken to an online Microsoft Forms survey and they were provided with detailed information regarding the study, including information about confidentiality and their right to withdraw from the online survey at any time; however, they were made aware that their data thus far would still be collated. In line with guidelines for conducting research with autistic communities (Gowen et al., 2020), additional attention was given to ensure that participants understood the relevance of the research, what it would entail, and how they could find out more about the study and the results, before they began the survey.

It took participants, on average, around 20 minutes to complete the online survey which consisted of an 'about you' demographics section, The Autism Symptom Self-Report (ASSERT; Posserud et al., 2013), and the Adult Repetitive Behaviour Questionnaire-2A (RBQ-2A; Barrett et al., 2015).

### **1.3 Ethical considerations**

My study was approved by the University of Southampton's Ethics and Research Governance Online (ERGO), ID: 63592. An ethical consideration that arose during the development of my research study was that of the recruitment and experiences of participants. Due to budget constraints, I chose to offer an incentive in the form of entry into a prize draw to win a range of Amazon vouchers, however I had discussions with my supervisors (and indeed some people who responded to my recruitment advert) about the ethics of seeking an individual's time and feedback on their experiences without providing any direct financial compensation. This is a challenge for many researchers, especially post-graduate and doctoral researchers who have very limited research budgets, and is something that will need to be continuously discussed within

academia in order to ensure individuals are appropriately compensated for their time and sharing of their experiences.

## 1.4 Epistemological stance

I consider my epistemological stance to be that of a critical realist. Critical realism postulates that there is one reality, but that there will be multiple interpretations of that reality and that knowledge is historically, culturally and socially situated (Maxwell, 2012; Bhaskar, 2008; as cited in Botha., 2021). In relation to autism, critical realism addresses “the interplay between biology, environment, social and cultural values, and discourses, and further, how the autistic person interacts with each of those structures” (Botha, 2021; p.12). From my systematic literature review, I believe that I highlight how societal and cultural values influence what we view as ‘autistic behaviours’ in males and females and how this, in turn, influences the nosology of autism. However, taking a critical realist approach has also raised important questions and reflections regarding my research and for myself as a non-autistic researcher. Kourti (2021) argues that, in order to really understand autism, researchers must explore autism knowledge at a deeper level, something to which only autistic people themselves have access (whether they can communicate this knowledge or not) given their lived experience. I learnt about transphenomenality, which is the notion that “knowledge consists of more than appearances” (Kourti, 2021; p 5). This is incredibly important to consider given so much of society’s understanding of autism and how it is assessed, diagnosed and researched, comes from a standpoint whereby ‘surface-level’ observations of autism are possibly held at a higher explanatory value than they actually hold (Kourti, 2021). Kourti explains the limitations of such an approach and postulates that, as a result, all non-autistic researchers can research is the phenomena of autism, not the factors and mechanisms that cause the phenomena. In summary, Kourti (2021) argues that non-autistic researchers can only explore what autism looks like, not what it is. What non-autistic research lacks, including my own, is the exploration of embodied experience- what is it like to be autistic. For my research, this limitation, in part, stems from my choice of methodology. My initial view of what made a ‘good’ piece of research was most likely underpinned by positivist assumptions, including the high regard of objectivity in research and the importance of strictly adhering to a specific methodology (Botha., 2021). As a result, I chose a quantitative study design in order to negate some of the criticisms of qualitative designs being too subjective to draw conclusions regarding the experiences of a large and heterogeneous group, such as the autistic population. However, as a result, I think that I have fallen into the trap of many autism researchers before me, missing an opportunity to qualitatively collect rich and detailed lived experiences of autistic people, to help gain an understanding of what it is like to be

autistic. As a result, I advocate for future autism research to, at the very least, use a mixed method approach to capturing autistic individuals' experiences.

Reading papers by Botha (2021) and Kourti (2021) has also highlighted to me the importance of autistic researchers being part of autism research. This has made me reflect on my own position as a non-autistic researcher and, at times, I have questioned whether it was even right for me to identify a research topic without first discussing it with the autistic research community first. The James Lind Alliance Priority Setting Partnership top ten list of priority research areas, ranked by a sample of the UK autistic community (Warner et al., 2019), does not include research into sex/gender differences and, as a result, I accept that my research reflects a noticeable gap between the research priorities of academics and autistic people (Pellicano et al., 2014). However, I stand by my rationale for why this research is important, as the autistic community's call for improved autism services will, to some level, rely on there firstly being a better understanding of the differences in experience and presentation of autism across genders and how this may influence assessment and diagnosis.

## **1.5 Autism, gender, and intersectionality**

The complex way in which categories, such as sex, gender, and disability (including identifying as autistic) interact with each other, often resulting in marginalisation and discrimination, is referred to as intersectionality (Crenshaw 1989; Martino and Schormans., 2018). Within autism research, intersectionality relates to ensuring that individuals are not reduced to a singular identity (e.g., being autistic or being male or female) and acknowledging that the intersection of various categories will influence a person's experience and, indeed, access to referral, diagnosis, and research. As already stated, one of the key ways this occurs within autism research is the underrepresentation of autistic women as participants, which risks perpetuating the stereotypical male representation of autism which may hinder access to diagnosis and support for autistic females.

Addressing and acknowledging intersectionality in my research was a challenge for me. On the one hand, I felt that the focus of my work was to increase the representation of autistic women, for example by including self-identifying participants who may be missed in other studies requiring an official diagnosis for inclusion, and to identify the differences between autistic males and females that can go on to increase awareness and improve access to services. However, despite my attempts to ensure representation of individuals identifying outside of the gender binary (e.g., by asking participants to free-report their gender identity), data limitations meant that I was unable to use gender identity as the independent variable in analysis. Instead, I had to

use participants' biological sex, which I recognise perpetuates the underrepresentation of the non-binary population in research and, when reading my thesis, non-binary individuals may feel ignored and disrespected, which was not my intention. The inclusion of non-binary identities is particularly important in this area of research, given many autistic individuals identify with genders outside of the male-female binary (Cage and Troxell-Whitman, 2019; George and Stokes., 2017). Therefore I also recognise that my research findings may not be representative of autistic individuals who identify as non-binary. The limited number of participants self-reporting as non-binary (n=4) could reflect difficulties I had recruiting this specific population. In their review, Cascio, Weiss and Racine (2020) commented that recruitment materials may contribute towards the under-recruitment of non-binary persons, for example the use of gender terminology may put some individuals off taking part. This will be something future researchers will need to consider carefully. Collaborating with autistic individuals who identify as non-binary during the study development process would be advantageous.

## **1.6 Implications for Educational Psychology**

A greater understanding of sex/gender differences in RRBIs, and autism as a whole, will help education professionals to provide more appropriate support for autistic children and young people (Tierney et al., 2016). The fact that females continue to be under-diagnosed, or later diagnosed as autistic suggests that there may be many children and young people not receiving the tailored support they need, resulting in negative outcomes for their wellbeing and learning. Identifying young people as autistic in a timely manner is crucial if they are to receive early, needs-driven support.

For the majority, the assessment and diagnosis of autism is not something that Educational Psychologists (EPs) are directly part of. However, I would argue that EPs do play a pivotal role in the early identification of autism, due to their direct work with school staff and families. Often, the first query regarding autism comes from education staff, following observations of the child finding aspects of their social and learning environment difficult. School staff, such as Special Educational Needs Coordinators (SENCOs), may liaise with an EPs to explore why the child might be finding things challenging and to discuss ways to support them. As such, it is possible that, for some children, an EP is one of the first professionals they may encounter on their journey to an autism diagnosis. Early identification of autism within school, particularly for females, can be supported by EPs themselves being suitably knowledgeable about how autism manifests differently between sex/genders, as well as being aware of behavioural presentations, such as camouflaging, that might be particularly relevant to autistic females within the school context. EPs should ensure that they are particularly well-informed of the differences in RRBIs, as these are

behaviours that may be more difficult to capture in diagnostic instruments and via observations in school settings. This knowledge can then be disseminated to school staff via training and discussions in consultations, facilitated by EPs.

## **1.7 Dissemination plan**

I have written the systematic literature review and meta-analysis and the empirical research paper with a view to publishing in the *Journal of Child Psychology and Psychiatry (JCPP)* and *Autism*, respectively. I believe that my systematic literature review and meta-analysis would be a valuable addition to the JCPP as it would complement recent reviews the journal has published regarding sex/gender differences in autism (e.g., Wood-Downie et al., 2020). Similarly, *Autism* is known for publishing research exploring autistic experiences, with a focus on participatory research. I anticipate including a lay summary as part of my dissemination, to ensure accessibility for those outside of the research community, including the autistic community (as per recommendation by Gowen et al., 2020). I also plan to share the findings with the participants who completed the survey by sending an email (to those who provided one) with a lay summary and a link to the published journal article. Finally, my research will be shared with other professionals through my university's post-graduate conference and by sharing with my current Educational Psychology Service in a team meeting. I believe that dissemination of both my research papers is important to raise awareness of sex/gender differences in the presentation of autistic RRBIs as well as to promote future methodological decisions made in my research, such as including self-identifying autistic participants and using self-report outcome measures.



## **Chapter 2 A Systematic Review and Meta-Analysis of Sex/Gender Differences in Restricted and Repetitive Behaviours and Interests in Autistic Children, Adolescents, and Adults**

### **Abstract**

Evidence that autism manifests differently between males and females is growing, particularly in terms of social interaction and communication; however less is known about sex/gender differences in the 'restricted and repetitive behaviours and interests' (RRBIs) domain. At the broad construct level, previous reviews have identified autistic females as displaying fewer overall RRBIs than males. However, individual studies exploring sex/gender differences at the narrow construct level (i.e., stereotyped behaviours, restricted interests, insistence on sameness, and/or sensory experiences) have produced mixed results, and therefore require systematic synthesis. I conducted a systematic review and four random effects meta-analyses that investigated sex/gender differences in narrow construct measures of RRBIs in autistic children, adolescents, and adults. Forty-four studies were narratively synthesised and twenty-two of these were included in four random effects meta-analyses, which found that autistic males had significantly higher levels of stereotyped behaviours (SMD = 0.22,  $p < .001$ ) and restricted interests (SMD = 0.24,  $p < .01$ ) compared to autistic females. There was a trend towards females having higher levels of sensory experiences (SMD = -0.16,  $p = .09$ ). No significant sex/gender differences were identified for insistence on sameness (SMD = 0.02,  $p = .54$ ). The findings from the narrative synthesis were broadly consistent with those from the meta-analyses and also found qualitative differences in the types of passionate interests autistic females hold, which may not be captured by current diagnostic instruments. These findings highlight the importance of practitioners being aware of possible subtle sex/gender differences in RRBIs, which may contribute to the under-recognition of autism in females, and the need for future research to investigate autistic sex/gender differences at a fine-grained level.

## 2.1 Introduction

Autism is a neurodevelopmental condition characterised by differences in social communication and interaction and restricted and repetitive behaviours and interests (American Psychiatric Association, 2013). One in 57 children in England have a diagnosis of autism (Roman-Urrestarazu et al., 2021) and the sex/gender<sup>1</sup> ratio is around four males to every one female (Fombonne et al., 2009; Kreiser & White, 2014; Maenner et al., 2020). However, the ratio is greater (around 3:1) when using population-based studies that include participants screened for high autism traits (Loomes et al., 2017). This suggests that there is a proportion of women and girls reporting high autism traits that are not receiving a diagnosis despite meeting clinical criteria (Russell et al., 2011) and those who do, tend to present with lower cognitive ability and/or additional behavioural difficulties (Dworzynski et al., 2012) or receive their diagnosis much later in life (Begeer et al., 2013; Kirkovski et al., 2013; Rivet & Matson, 2011).

There are numerous hypotheses for the preponderance of male autism diagnoses. Some propose that there are genetic differences between males and females that influence the likelihood of autism, for example women having a genetic ‘protective factor’ (Robinson et al., 2013) whereby females need a larger genetic load for autistic behaviours to manifest (Skuse, 2007). The ‘extreme male brain theory’ and the ‘foetal testosterone hypothesis’ (see Baron-Cohen, 2002; Ingudomnukul et al., 2007) propose that autistic individuals<sup>2</sup> are more likely to have a ‘masculine brain type’ (difficulty with empathy and higher levels of systemising ability), hence the greater proportion of males diagnosed. However, these genetic theories have been criticised for neglecting the influence of gender socialisation and overly focussing on autistic individuals with average or above-average intelligence quotient (IQ), e.g., Aspergers (Buchen, 2011; Krahn & Fenton, 2012). Meanwhile, other researchers suggest that autism manifests differently in females, who may, for example, have more age and gender appropriate ‘restricted and repetitive’ interests compared to males, and females may use more behavioural strategies, such as camouflaging and masking (see the female autism phenotype; Hull et al., 2020; Kirkovski et al., 2013; van

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<sup>1</sup> The effects of biological sex and socially constructed gender are difficult to separate, therefore in this review the term ‘sex/gender’ will be used to reflect that most people’s identities are informed by both sex and gender (as commented by Lai, Lombardo, Auyeung, Chakrabarti & Baron-Cohen, 2015).

<sup>2</sup> In line with the identity-first movement endorsed by many within the autistic community (Gernsbacher, 2017; Milton, 2012; Kenny et al., 2016), in this review, identity-first language (‘autistic individual’, ‘autistic males’, ‘autistic females’) will be used throughout unless referring to quoted text from other authors.



Wijngaarden-Cremers et al., 2014 for reviews), that causes them to ‘fly under the radar’ with regards to early identification in school and, subsequently, receiving appropriate support. Female autism diagnosis may be hindered further by the current nosology and assessment of autism, which is criticised for being male-biased (Bargiela et al., 2016; Hiller et al., 2014; Mandy et al., 2012) and lacking sensitivity in identifying sex/gender differences (Lai & Baron-Cohen, 2015).

### **2.1.1 Sex/Gender Bias in Autism Referral, Diagnostic Criteria and Assessment Tools**

The nosology of autism, and subsequent development and validation of diagnostic tools, is influenced by research which has predominantly focussed on clinical samples (see reviews by Lai & Baron-Cohen, 2015; van Wijngaarden-Cremers et al., 2014) that are overly representative of males (Watkins et al., 2014) and ‘severe’ cases of autism (Hartung & Widiger, 1998). As a result, “most of what we believe we know about autism is actually about males with autism” (Thompson et al., 2003; p. 351). The notion of autism as a ‘male’ condition has reduced the opportunity for autistic females to be identified and referred for assessment, particularly during childhood, as autistic behaviours in girls are more likely to be overlooked (Hiller et al., 2014; Mandy et al., 2012) or misinterpreted by key adults, including educational practitioners (Aggarwal & Angus, 2015; Holtmann & Bölte, 2007). This is particularly pertinent given autism referral guidance stipulates that difficulties must be present across contexts, such as school and home (Attwood et al., 2006; Dworzynski et al., 2012; Mandy et al., 2012). Additionally, the potential presence of camouflaging in schools may further hinder early identification of autistic girls (Attwood et al., 2006; Dean et al., 2017).

Due to a lack of reliable genetic biomarkers for autism (Goldani et al., 2014), the assessment of autism relies on reported and observed behaviours, often using “gold standard” tools (Falkmer et al., 2013; Ozonoff et al., 2005) such as the Autism Diagnostic Interview Revised (ADI-R; Rutter et al., 2003) and a structured observation such as the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2012). However, such tools have been criticised for being developed and validated using predominantly male clinic samples (Bargiela et al., 2016; Lai & Baron-Cohen, 2015; McCrimmon & Rostad, 2014), and therefore may not be sensitive to how autism presents in females (Wood-Downie et al., 2021). The sensitivity of the ADOS at identifying autistic females can also been questioned by research findings that adult autistic females (diagnosed in childhood) were less likely to meet ADOS cut-off scores compared to males (Lai et al., 2011). Girls with the same levels of autistic traits to boys have also been found to be less likely to receive a diagnosis of autism (Russell et al., 2011), suggesting that females need to surpass a higher threshold of severity to meet diagnostic criteria (Dworzynski et al., 2012; Kreiser & White, 2014). This might explain why females with higher IQ, less extreme stereotypies, and/or fewer

behavioural difficulties are often missed by autism diagnostic tools (Begeer et al., 2013; Dworzynski et al., 2012) or may be diagnosed much later in adulthood (Begeer et al., 2013; Kirkovski et al., 2013; Rivet & Matson, 2011).

### 2.1.2 Broad and Narrow Constructs in Autism

According to the Diagnostic Statistical Manual Version 5 (DSM-5; American Psychiatric Association, 2013, p. 53), individuals who demonstrate both “persistent deficits in social communication and social interaction across multiple contexts” and “restricted, repetitive patterns of behaviour, interests, or activities” meet the clinical diagnostic criteria for autism. These observable/reported behaviours can be categorised at both the ‘broad’ and ‘narrow’ construct level (Lai et al., 2015). Broad constructs refer to the more abstract definitions of autism symptomatology, whilst narrow constructs refer to the subdomains within the broad construct, all of which will have a variety of behavioural exemplars. Restricted, repetitive patterns of behaviours, interests, or activities (RRBIs) are a core component of the autism presentation (see Table 1 for detailed example of RRBIs at the broad, narrow, and behavioural exemplar level).

Reviews and large-scale studies into sex/gender differences in autistic RRBIs at the broad construct consistently find that autistic females display fewer RRBIs than males (Frazier et al., 2014; Supekar & Menon, 2015; Szatmari et al., 2012 ;see Lai et al., 2015; and Wijngaarden-Cremers et al., 2014 for reviews). For example, a meta-analysis of 22 studies identified that females had, on average, less RRBIs (based on ADI-R and ADOS overall scores) compared to males, but there were no sex/gender differences prior to the age of six years old (van Wijngaarden-Cremers et al., 2014), suggesting less sex/gender difference in RRBIs in young children. A criticism of research focussing only on the broad construct of RRBIs (e.g., using the RRB domain on the ADOS or ADI-R) is that potential subtle differences at the narrow construct level may be missed, which has previously been demonstrated in the social interaction and communication domain (Wood-Downie et al., 2021).

Consistent with this proposition, studies that have explored sex/gender differences in narrow constructs of RRBIs produce more mixed results, for example no sex/gender difference on ADI-R items relating to behavioural exemplars such as stereotyped language, unusual sensory interests, and resistance to change (McLennan et al., 1993). Lai et al., (2011) also reported autistic females as having more ‘lifetime sensory issues’ compared to autistic boys. These studies suggest that there may be sex/gender differences in specific domains of RRBIs however, there is yet to be a systematic literature review and meta-analysis of narrow constructs and behavioural exemplars of ‘restricted, repetitive patterns of behaviour, interests, or activities’ (RRBIs) based upon DSM-5

criteria (see Table 1), which provides the most recent diagnostic classification of autism. The current DSM-5 also includes sensory symptoms as a narrow construct within RRBIs, meaning sensory experiences may not have been captured in studies prior to 2013 using the DSM-IV criteria (American Psychiatric Association, 1994). Research interest in autistic sensory experiences has expanded in the last decade (see meta-analysis by Ben-Sasson et al., 2009, 2019), but there has yet to be a systematic literature review and meta-analysis specifically exploring sex/gender differences in this narrow construct of RRBIs.

A more detailed understanding of the similarities and differences between autistic male and female RRBIs could contribute towards the growing awareness researchers, professionals and the wider public have of the various presentations of autism, moving away from the male-centric stereotype. A more nuanced understanding of sex/gender differences in RRBIs could also inform the development of assessment instruments that are sex/gender dependent and are more sensitive to autistic women and girls (Lai et al., 2015).

Table 1. *Examples of broad/narrow constructs and associated behavioural exemplars based upon DSM-5 diagnostic criteria.*

Broad Construct	Narrow Construct	Behavioural Exemplars
Restricted and repetitive behaviours and interests	Stereotyped or repetitive motor movements, use of objects, or speech	Lining up toys Flipping objects Echolalia Idiosyncratic phrases
	Insistence on sameness, inflexible adherence to routines, or ritualized patterns of verbal or nonverbal behaviour	Distress at small changes Difficulties with transitions Rigid greeting rituals Need to take same route or the same eat food every day
	Highly restricted, fixated interests that are abnormal in intensity or focus	Strong attachment to or preoccupation with unusual object Excessively circumscribed or perseverative interests
	Hyper- or hypo reactivity to sensory input or unusual	Apparent indifference to pain/temperature

interest in sensory aspects of the environment	Adverse response to specific sounds or textures
	Excessive smelling or touching of objects
	Visual fascination with lights or movement

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### 2.1.3 Current study

The current systematic literature review and meta-analysis explores sex/gender differences in narrow constructs of restricted and repetitive behaviours and interests (RRBIs). Data was collected from studies that featured measures of narrow constructs of restricted and repetitive behaviours and interests based on the DSM-5 symptom subdomains (see Table 1), including hyper- or hypo reactivity to sensory input or unusual interest in sensory aspects of the environment, for both autistic (or high autistic trait) males and females. The review aims to explore the question:

How do different restricted and repetitive behaviours and interests manifest between autistic males and females?

## 2.2 Method

The systematic literature review, including search terms, inclusion and exclusion criteria, and PICO chart, was prospectively registered on Prospero (registration number: CRD42021254221). The review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA; Moher et al., 2009).

### 2.2.1 Search Strategy

A search of databases APA PsychInfo, Medline, ERIC, Science Direct, PsycArticles, and CINAHL Plus with Full Text on 25th May 2021, based upon the DSM-5 autism symptom subdomains of restricted, repetitive patterns of behaviour, interests, or activities, including the population terms including as 'autism spectrum disorder' and 'autism spectrum condition'; comparator terms 'sex' and 'gender'; and outcome terms including 'repetitive behavi?r\*', 'restricted interest', 'insistence on sameness', 'sensory', and 'circumscribed interest\*' (Details of can be found in Full Search Terms in appendix A). An English language restriction was applied. In

addition, reference lists of included studies were hand-searched to detect any pertinent study possibly missed with the electronic search.

### 2.2.2 Eligibility Criteria

Cross-sectional, peer-reviewed, articles including autistic males and females, of any age, and including an outcome measure of subdomains of restricted and repetitive behaviours and interests (RRBIs) based upon the DSM-5 autism diagnostic criteria, were included (see Table 1). For studies that only report overall RRBIs scores (e.g., overall Stereotyped Behaviours and Restricted Interests score from ADOS-2), corresponding authors were contacted to request subscale scores. Likewise, authors of studies that did not report RRBIs data for males and females separately, were contacted to request this information. In acknowledgement to camouflaging theories of autism and the documented limitations of current autism diagnostic methods, studies including participants with high autistic traits and/or participants self-identifying as autistic were also included. Studies featuring a very small number (six or less) of autistic female participants were excluded, to allow for appropriate statistical comparisons. Of note, all studies that were excluded due to small number of autistic females, also met other exclusion criteria and/or did not provide data for males and females separately. In response to the latter, authors were contacted to request data, so that a post-hoc sensitivity analysis could be conducted, however none responded. Studies that did not include a measure reporting the subdomain of RRBIs according to the DSM-5 diagnostic criteria were excluded. Studies that reported data pertaining to the domain of 'Repetitive Sensory Motor Behaviours' (RSMB) were excluded due to the inability to separate the two subdomains encapsulated by this definition (e.g., stereotyped behaviours and sensory experiences) from each other. Studies that did not use a quantitative cross-sectional design, for example qualitative, intervention, review papers, and single-case designs were excluded (see Table 2).

Table 2. *Rationale for inclusion and exclusion criteria.*

Inclusion/exclusion criteria	Rationale
Quantitative cross-sectional studies only	The review is exploring whether there are quantifiable differences in RRBIs between male and female autistic individuals at a given time point.

Studies including a measure of the subdomain of the DSM-5 autism diagnostic criteria of restricted, repetitive patterns of behaviour, interests, or activities (RRBI)	Previous sex/gender reviews (van Wijngaarden-Cremers et al., 2014; Lai et al., 2015) have typically focused on broad constructs of autism symptomology, for example, restricted, repetitive patterns of behaviour, interests, or activities (RRBI), not at the narrow construct level. However, recent studies have used narrow construct levels to explore sex differences in other domains of autism, such as social interaction (Wood-Downie et al., 2020).
Studies featuring a very small sample of autistic female participants (six or less) were excluded.	To allow for appropriate statistical comparison, a sufficient number of participants in each group is required.

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### 2.2.3 Search Results

As can be seen from Figure 1, a total of 1370 studies were identified through database searching and an additional one study was identified through other sources, such as hand searching relevant reference lists. After duplication removal and title and abstract screening, the full-text of 126 studies were assessed for eligibility, of which 44 were included within the narrative synthesis and 22 of these were included in the meta-analyses. I was unable to include the remaining 22 studies in the meta-analyses as the either the authors did not respond to my requests for data or, when they did, the data needed was not available (e.g., data for males and females separately).

This process was first conducted by the first author (HE). To check for reliability and minimise bias, a second author (HWD) independently completed abstract screening of 25% of studies. Cohen's Kappa test indicated a substantial agreement between both reviewers (0.68). Following inter-rater checks, it was established that there was a particular discrepancy between the two author's inclusion of studies with outcomes measures capturing sensory sensitivities. As a result, HE re-reviewed the remaining 75% of studies to double check for sensory sensitivity outcomes measure studies that could be included. HWD also assessed the full-texts of 25% of studies for eligibility and again Cohen's Kappa test indicated a substantial agreement between both reviewers (0.74). Discrepancies were resolved by the two reviewers through discussion.

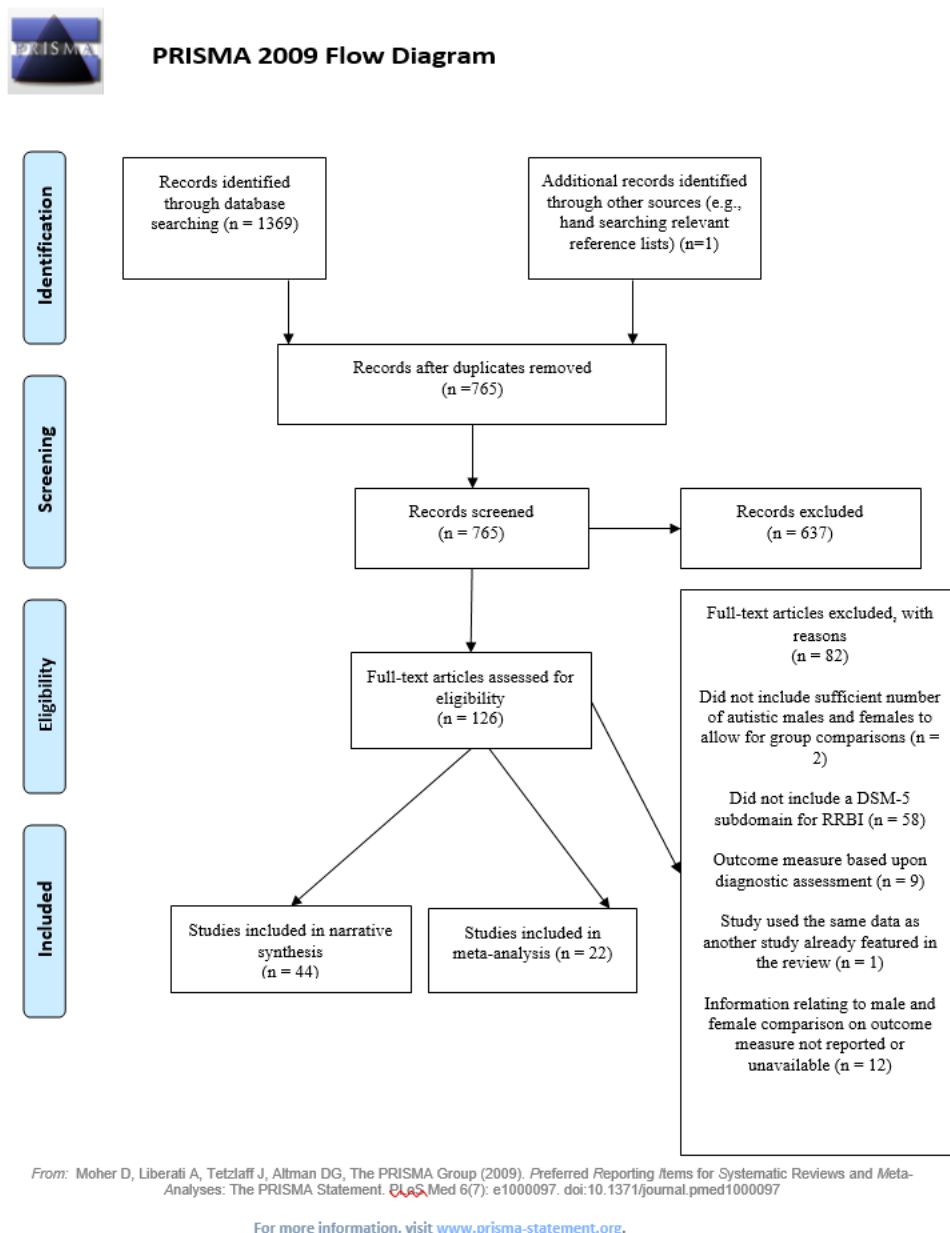


Figure 1. PRISMA Flowchart

### 2.2.4 Data Extraction

Data on sample characteristics (e.g., sex, age, diagnostic criteria used, IQ data (where available) and outcomes related to narrow constructs of restricted, repetitive patterns of behaviour, interests, or activities) were extracted from included studies and independently entered by the first author. A second author (HWD) also extracted means, standard deviations, and participant numbers for 25% of studies included in the meta-analysis and agreement was perfect (100%).

A factor analysis by Lam and Aman (2007) indicated five factors that accounted for 47.5% of the variance amongst the 43 items on the Repetitive Behaviour Scale- Revised (RBS-R: Bodfish et al., 2000), rather than the originally proposed six. Subscales 'ritualistic behaviour' and 'sameness behaviour' were combined due to significant overlap and similarity across items. In this review, I chose to maintain the five-factor model, excluding data pertaining to the 'ritualistic' subscale in favour of the 'sameness' subscale so that I could map this on to the narrow construct of 'insistence on sameness', in studies where the original six-factor structure was used.

### **2.2.5 Characteristics of Included Studies**

Stereotyped or repetitive motor movements, use of objects, or speech<sup>3</sup> (N=27) and highly restricted, fixated interests that are abnormal in intensity or focus<sup>4</sup> (N=26) were the most frequently studied outcome measures featured in the review, followed by insistence on sameness, inflexible adherence to routines, or ritualised patterns of verbal/non-verbal behaviour (N=23) and hyper- or hypo reactivity to sensory input or unusual interest in sensory aspects of the environment<sup>5</sup> (N=22). For the purpose of this review, studies reporting data pertaining to 'total sensory' scores or 'sensory sensitivity' were included in the meta-analysis, as this reflected the most predominant element of sensory experiences explored in the available research. Numerous studies explored multiple narrow constructs and therefore were categorised into multiple descriptors. Characteristics of all included studies are presented in Table 3.

The majority of studies included children (n =34) with 10 studies exploring RRBI outcomes exclusively in adults (those over 18 years; Aita et al., 2019; Aykan et al., 2020; Barrett et al., 2018; Caldwell-Harris & Jordan, 2014; Dell'Osso et al., 2017; English et al., 2021; Grove et al., 2018; Hattier et al., 2011; Lever & Geurts, 2018; Weiland et al., 2020). Diagnosis of autism was confirmed in 22 studies, often via clinical assessment and/or ADOS assessment (n = 13) or autism trait screening tools or review of diagnostic reports (N=9). Nearly all studies used questionnaire data as outcome measures, apart from Harrop et al., (2015; 2016) who used video observations of caregiver-child interactions (CCX) and eye-tracking technology, respectively.

### **2.2.6 Study Quality Appraisal**

The included studies were evaluated for quality using the Newcastle-Ottawa Quality Assessment Scale (N-OQAS, see appendix B), adapted for cross-sectional studies (Herzog et al.,

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<sup>3</sup> Hereafter referred to as 'stereotyped behaviours' unless other terminology used in specific studies.

<sup>4</sup> Hereafter referred to as 'passionate interests' unless other terminology used in specific studies.

<sup>5</sup> Hereafter referred to as 'sensory experiences' unless other terminology used in specific studies.



2013). The adapted N-OQAS was used to assess representativeness and size of the sample, response rate, validity of measurement tools, comparability, outcome (including provision of confidence intervals and power calculation), and appropriateness of statistical analysis. Language was adapted to fit with the population and outcome measure being studied. Studies could be awarded a maximum score of 11 points. Studies with scores of 10-11 points were considered 'very good', scores of 6-9 points were considered 'good', studies with scores of 4-5 points were considered 'satisfactory', whilst a score of 0-3 points were considered 'unsatisfactory'. A total of six studies received a score of 'good', 16 'satisfactory' and 22 'unsatisfactory'. No studies received a score of 'very good'. Of the 20 'unsatisfactory' studies, the majority scored poorly due to unrepresentative samples (e.g., participants were all selected from one group/location based on convenience; N= 18), no justification of sample size (N=20), not reporting data on non-responders (N=19), not describing how autism diagnosis was confirmed or not using a validated measurement tool (N=18), not controlling for factors such as age or IQ (N=19), and not reporting p-values (N=9). Ten studies did report p-values, however they did not report effect sizes or confidence intervals.



Table 3. *Study and sample characteristics of all included studies.*

Study characteristics									
Authors (date)	Narrow construct/behavioural exemplar assessed	Measure used for Meta-Analysis	Diagnoses at the time of study	Diagnostic criteria used	How diagnosis was confirmed	Males ( <i>n</i> )	Females ( <i>n</i> )	Mean age (years)	Quality assessment score
Aita et al., (2019)	Stereotyped behaviours Insistence on sameness Restricted interests	Mean of RBS-R (Japanese translation) subscales	ASD	DSM-4 TR	Childhood Autism Rating Scale-Tokyo Version (CARS-TV; Kurita et al., 1989)	39	15	36.69	4 (Satisfactory)
Antezana et al., (2019)	Stereotyped behaviours Insistence on sameness Restricted interests Sensory	RBS-R item-level scores	ASD	DSM-4 TR	Not confirmed	507	108	10.26	3 (Unsatisfactory)

## Chapter 2

Anthony et al., (2015)	Restricted interests	Percentage of interests endorsements and intensity of interests (Interests Scale; Bodfish, 2004).	Autism, Asperger's Syndrome, PDD-NOS	DSM-4 TR	ADI/ADI-R and/or ADOS	93	16	12.70 years	2 (Unsatisfactory)
Aykan (2020)	Sensory experiences	Mean auditory and visual sensitivity scores from the Sensory Sensitivity Scales (SeSS; Aykan et al., 2020).	No clinical diagnosis of autism.	N/A	Autistic traits established using Autism Spectrum Quotient (AQ; Baron-Cohen et al., 2001; Kose et al., 2010)	36	39	23.01 years	3 (Unsatisfactory)
Barrett et al., (2018)	Insistence on sameness	Mean score on insistence on sameness subscale of the Repetitive Behaviours Questionnaire-2 (RBQ-2A; Barrett et al., 2018).	Participants self-reported Asperger's syndrome, ASD, HFA, or other (e.g., PDD-NOS)	Not commented.	Participant self-reported.	100	171	36.56	4 (Satisfactory)

Bitsika, Sharpley & Mills (2018)	Sensory experiences	Mean of Sensory processing scores:  Low registration  Sensation seeking  Sensory sensitivity	ASD	'Relevant DSM criteria'	ADOS-2 by research team	51	51	10.2 years	8 (Good)
Boyd (2020)	Stereotyped behaviours	Results from differential item functioning analysis for items on the ADOS-2 restrictive, repetitive behaviour subscale.	ASD	Not commented	ADOS-2	5848	1599	6.87	4 (Satisfactory)
Brierley (2020)	Stereotyped behaviours  Insistence on sameness	Mean of RBS-R subscales (stereotyped behaviours and insistence on sameness subscales reported only).	ASD	Not commented	ADOS and ADI-R	496	138	10.2	5 (Satisfactory)
Caldwell-Harris & Jordan (2014)	Restricted interests	Reports from The Cambridge University Obsessions Questionnaire (Baron-Cohen & Wheelwright, 1999), modified to add	Self-reported ASD, Asperger's syndrome, autistic spectrum	Not commented	Not commented	39	30	29.2	0 (Unsatisfactory)

Chapter 2

categories of nature,  
history and culture.

disorder,  
PDD-NOS.

Self-report rate of  
intensity from 1 (casual)-  
3 (intense).

Dell’Osso et al., (2017)	Sensory experience  Inflexible adherence to routine  Restricted interest and rumination	Mean domain scores from Adult Autism Subthreshold Spectrum (AdAS Spectrum; Dell’Osso et al., 2017).	ASD diagnosed or participants endorsing at least one criterion symptom for ASD.	DSM-5	Not commented	66	36	24.29	2 (Unsatisfactory)
English et al., (2021)	Sensory sensitivity  Repetitive behaviour <sup>a</sup>  Cognitive rigidity <sup>b</sup>	Mean of Comprehensive Autism Trait Inventory (CATI ; English et al., 2021) sensory sensitivity, restricted behaviour, and cognitive rigidity scores.	Diagnosed ASD or self-identifying ASD	Not commented	Not commented	557	522	37.41	3 (Unsatisfactory)

Evans (2018)	Stereotyped behaviour  Insistence on sameness  Restricted interests	Mean of RBS-R subscale scores.	ASD	Not commented	Not commented	143	32	3.5	2 (Unsatisfactory)
Feldman et al., (2020)	Sensory seeking Hyper-responsiveness Hypo-responsiveness	Mean scores of Sensory Profile (Dunn., 2012) subscales; low registration, sensation seeking, sensory sensitivity, sensation avoiding.	Autism	DSM-5	ADOS-2 and clinical judgement by research team	37	13	13.1	2 (Unsatisfactory)
Fetta et al., (2021)	Sensory Stereotyped behaviour  Insistence on sameness  Restricted interests	Mean of Short Sensory Profile 'under-responsive/seeks sensation' subscale.   Mean of RBS-R subscales.	ASD	ICD-10/DSM-5	ADOS-2	39	11	3-15 range	4 (Satisfactory)

## Chapter 2

Fulceri et al., (2016)	<p>Stereotyped Behaviours</p> <p>Insistence on sameness</p> <p>Restricted Interests</p>	Mean of RBS-R (Italian translation) subscale scores.	ASD	DSM-4 TR	ADOS-G	64	15	4.3	7 (Good)
Grove et al., (2018)	Restricted interests	Participant responses to; listing special interests, number of days per week/hours per day spent engaging in special interests, ratings on what extent they feel their special interest has a positive impact on their life, and how much it impacts with their daily functioning.	Autism Spectrum Disorder	DSM-4 or DSM-5	Not commented	222	185	42.4	1 (Unsatisfactory)



Harrop (2015)	Stereotyped behaviour (grouping, repetitive use, or manipulation of objects; whole body movements; complex and unusual mannerisms; verbal atypicality)	Frequency of lower order RRBs, based on scheme by Harrop et al (2014), observed via videotaped caregiver-child interactions (CCX).	ASD	Not commented	ADOS-2	29	29	girls (3.2) boys (2.9)	6 (Good)
	Sensory experiences (sensory seeking, sensory aversion, sensory visual)								
Harrop (2018)	Restricted interests	Eye-tracking quantifying attention to male/female/neutral images: exploration (number of images viewed), perseveration (length of time each image was explored), and detail orientation (the amount of detail each image was inspected).	ASD	'a previous DSM diagnosis of ASD'	Verified via phone screen and parent/carer completion of Social Communication Questionnaire (SCQ; Rutter et al., 2003).	25	26	boys (9.4) girls (8.5)	4 (Satisfactory)

## Chapter 2

Hattier et al., (2011)	Stereotypies	Mean of Stereotypies subscale of Diagnostic Assessment for the Severely Handicapped-Second Edition (DASH- II; Matson, 1998)	ASD and severe to profound ID	Not commented	Not commented	77	63	49.28	5 (Satisfactory)
Hiller, Young & Weber (2014)	Restricted interests Stereotyped behaviours	Information relating to restricted interests as reported on diagnostic assessments and reports	High-functioning ASD	DSM-4-TR and DSM-5 for 114 participants.	Not commented	69	69	8.06 (girls)	4 (Satisfactory)
								8.76 (boys)	
Hiller, Young & Weber (2016)	Restricted interests Stereotypies	Predictive odds ratios for RRBs and diagnosis of ASD.	ASD or Asperger's syndrome or PDD-NOS	Not commented on	Not commented on	92	60	10.94	3 (Unsatisfactory)
Hus et al., (2007)	Insistence on sameness	Sum of parent responses to items relating to insistence on sameness on the ADI-R (LeCouteur et al., 2003).	ASD/Asperger's syndrome.	Not commented	Not commented	812	171	7.75	1 (Unsatisfactory)

Knutsen (2019)	<p>Stereotyped behaviour (stereotyped/idiosyncratic use of words or phrases; hand and finger or other complex mannerisms)</p> <p>Sensory experiences</p> <p>Repetitive interests (ADOS-2 modules 1 and 2)</p> <p>Restricted interests (Excessive interest or reference to unusual or highly specific topics or objects or repetitive behaviours (ADOS-2 module 3)</p>	Odds ratios based on scores on the RRB subscale of ADOS-2.	Autistic disorder, Asperger syndrome, PDD-NOS, ASD.	DSM-4 or DSM-5	Not commented	512	512	2-12 range	5 (Satisfactory)
Lane (2014)	Sensory	Mean scores on the Short Sensory Profile (SSP; McIntosh et al., 1999).	ASD	Not commented	Not commented	203	25	5.01	3 (Unsatisfactory)
Lam (2004)	Stereotyped behaviours	Mean of RBS-R subscale scores.	ASD	Not commented	Not commented	253	53	15.34	4 (Satisfactory)

## Chapter 2

Insistence on sameness

Restricted interests

Lawrence (2017)	Stereotypies Sensory	Mean score for sensory processing and stereotypies on the Autism Spectrum Rating System (ASRS).	Autism	DSM-5	Not commented	114	31	16	2 (Unsatisfactory)
Lee (2009)	Sensory experiences	Mean score low registration, sensory seeking, sensory sensitivity, and sensation avoiding subscales on the Adolescent/Adult Sensory Profile (Brown & Dunn, 2002).	Asperger syndrome	DSM-4	Confirmed using parental completion of the Asperger Syndrome Diagnostic Scale (ASDS; Myles, Bock, & Simpson, 2001)	116	33	12-18 range	4 (Satisfactory)
Lever (2018)	Sensory sensitivities	Total score on Sensory Sensitivity Questionnaire (SSQ; Minshew and Hobson 2008).	ASD	DSM-4	Not commented	116	56	19-79 range	4 (Satisfactory)
Mandy et al., (2012)	Repetitive and stereotyped behaviours Auditory sensitivity	Frequency of reported repetitive and stereotyped behaviours from the activities and interest subscale of the	Asperger's syndrome, Autistic Disorder, PDD-NOS.	Szatmari (2000) guidelines used to differentiate	3Di and ADOS (where available) alongside structured reports from school/nursery.	273	52	10.2 (boys) 9.7 (girls)	8 (Good)

		Children’s Communication Checklist (Bishop, 1998).			e between Asperger’s syndrome and AD diagnosis. PDD-NOS was diagnosed using DSM-4 TR.				
		Mean score for auditory sensitivity on the 3Di (Skuse et al, .2004).							
May, Cornish & Rinehart (2016)	Repetitive motor movements Rigidity/adherence to routine Autistic preoccupations Sensory sensitivities	Mean scores for repetitive motor movements, rigidity/adherence to routine, autistic preoccupations, and sensory sensitivities subscales on the Repetitive Behaviours Questionnaire–Second Edition (RBQ-II; Leekam et al., 2007).	ASD or Asperger’s syndrome	DSM-4	Symptom checklist based on DSM-4 criteria.	32	32	7-12 range	3 (Unsatisfactory)
McFayden et al., (2019)	Stereotyped behaviour Insistence on sameness Restricted interests	Mean scores from RBS-R subscales.  Frequency of reported types of interests by males and females.	ASD	DSM-5	ADOS and ADI-R	55	20	12.33	8 (Good)

Chapter 2

Nicholas et al., (2008)	<p>Stereotyped behaviour (preoccupation with objects, stereotyped mannerisms, repetitive language)</p> <p>Insistence on sameness (routines and rituals)</p> <p>Restricted interests</p>	<p>Frequency of reported diagnostic codes pertaining to behaviours associated with autism (e.g., repetitive language, restricted interests, routines and rituals, stereotyped mannerisms, preoccupation with parts of objects) in records obtained from clinical and educational sources.</p>	ASD	DSM-IV-TR	<p>A child was included as an 'ASD case' if they displayed behaviours as described in the DSM-IV-TR based on a comprehensive evaluation by a qualified professional (such as psychologist or developmental paediatrician).</p>	224	71	All aged 8 years	<p>1</p> <p>(Unsatisfactory)</p>
Sipes (2011)	<p>Repetitive motor movements</p> <p>Prefers foods of a certain texture or smell</p> <p>Reactions to normal, everyday sounds</p> <p>Reaction to normal, everyday lights</p> <p>Restricted interests and activities</p> <p>Routines and rituals</p>	<p>Percent endorsements for items on Baby and Infant Screen for Children with aUtism Traits-Part 1 (BISCUIT-Part 1; Matson, Boisjoli, &amp; Wilkins, 2007).</p>	ASD (Autistic disorder, PDD-NOS)	DSM-IV-TR	<p>Diagnoses were made by a licensed psychologist and information for diagnoses was obtained from scores on the Battelle Developmental Inventory, Second Edition (BDI-2; Newborg, 2005) and the Modified Checklist for Autism in Toddlers (M-CHAT; Charman et</p>	294	96	2.4	4 (Satisfactory)

Preoccupation with parts of an object

Interest in a highly restricted set of activities

Abnormal fascination with the movement of spinning objects

Repetitive speech

Upset with change in routine

Needs reassurance if events don't go as planned

Limited interests

Repetitive hand or arm movements

Repetitive motor movements including entire body

al., 2001; Robins, Fein, Barton, & Green, 2001), criteria from the DSM-IV-TR (APA, 2000), and clinical judgment.

Siracusa et al., (2021)	Stereotyped behaviour Insistence on sameness	Mean scores for RBS-R (Italian translation) subscales.	ASD	DSM-5	Diagnosis confirmed via a multidisciplinary assessment	154	65	9.1	5 (Satisfactory)
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## Chapter 2

### Restricted interests

Smerbeck (2019)	<p>Stereotyped behaviour</p> <p>Insistence on sameness</p> <p>Restricted interests (atypicality)</p>	<p>Mean scores of RBS-R subscales.</p> <p>Mean scores from Survey of Favorite Interests and Activities (Atypicality subscale)</p>	High Functioning ASD (HF ASD)	Not commented	Not commented	121	49	11.81	2 (Unsatisfactory)
Solomon et al., (2012)	<p>Stereotyped behaviour</p> <p>Insistence on sameness</p> <p>Restricted interests.</p>	Mean scores from RBS-R subscales.	ASD (including HFA, AS, and PDD-NOS)	DSM-4	Confirmed using ADOS-G and SCQ	20	20	12-18 range	2 (Unsatisfactory)
Stephenson, Norris & Butter (2021)	<p>Stereotypy</p> <p>Behavioural rigidity</p> <p>Sensory sensitivity</p>	Mean scores from unusual behaviours subscale; (stereotypy, behavioural rigidity, sensory sensitivity, atypical language) from the Autism Spectrum Rating Scale (ASRS).	ASD	Not Commented	Not commented	381	100	3.41	7 (Good)



Sutherland et al., (2017)	Restricted interests Sensory	Parent responses to questions regarding their children's restricted interests (open questions).  Responses on researcher-developed multiple-choice questionnaire exploring sensory sensitivities and seeking behaviours across the senses (vision, hearing, smell, taste, touch, proprioception, and vestibular).	ASD	Not commented on	Not verified (parent/carer report only)	163	171	5-18 range	2 (Unsatisfactory)
Tang et al., (2021)	Motor Stereotypies Echolalia Repeated/learned phrases repetitive use of objects distress at routine change	Frequency of clinical features of ASD (as described by DSM-5) collected via reading and interpretation of Autism Diagnostic reports.	ASD	DSM-5	ADOS-2	145	50	8.3 (female)  7.21 (male)	5 (Satisfactory)

## Chapter 2

difficulties with transition

ritualistic behaviour

fascination with spinning objects, lights and mirrors

auditory manifestations

olfactory manifestations

mouthing or licking objects

tactile manifestations

movement manifestations

proprioception and vestibular issues

fears reflecting sensory avoidance

Uljarevic et al., (2021)	Stereotyped behaviour Insistence on sameness	Mean scores from the RBS-R subscales.	ASD	Not commented	Not validated but SCQ scored collected and participants had to meet the cut-off.	14186	3395	8.24	3 (Unsatisfactory)
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	Restricted interests.								
Uljarevic et al., (2020)	Repetitive motor behaviours	Clinician rated severity of each DSM-4 items related to RRB domain.	ASD	Not commented on	Not commented on	3007	640	6.6	3 (Unsatisfactory)
	Insistence on sameness								
	Restricted interests								
Wanzek (2014)	Insistence on sameness Restricted interests.	Mean scores from RBS-R subscales.	Autistic Disorder, Asperger's Disorder, PDD-NOS	DSM-4 or ICD-10	Not commented	21	21	19 years, 6 months (females)  18 years 5 months (males)	3 (Unsatisfactory)
Weiland et al., (2020)	Sensory	Mean scores from Sensory Perception Quotient (SPQ)- Short (Weiland et al., 2020).	ASD	DSM-5	Participant self-reported diagnosis.	316	340	43.2	3 (Unsatisfactory)

Williams (2019)	Sensory	Mean Sensory subscale score on Social Responsiveness Scale – Second Edition (SRS-2; Constantino and Gruber., 2012)	ASD	Not commented	ADOS-2 and ADI-R (where applicable)	62	21	16.77 (males)	5 (Satisfactory)
								18.53 (females)	

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*Note.* <sup>a</sup> Repetitive behaviours' was classified as being within the stereotyped behaviour narrow construct. <sup>b</sup> Cognitive rigidity' was classified as being within the insistence on sameness narrow construct.

### **2.2.7 Data Synthesis Strategy**

In the narrative synthesis, findings from included studies are described, which have been categorised into narrow constructs corresponding to the four RRBI subdomains detailed in the DSM-5. A summary of findings for each narrow construct are featured below (full details of studies can be found in appendix C). The narrative synthesis details findings to the narrow construct level, however studies that comment of specific behavioural exemplars encompassed are also reported, where possible.

Random-effect meta-analyses were performed using Comprehensive Meta-Analysis (Borenstein et al., 2013) for the four narrow construct measures of restricted and repetitive behaviours and interests, including sensory experiences, as per the DSM-5 criteria. Standardised mean differences (SMD) were calculated for autistic males and autistic females, converting from other effect size metrics (e.g., odds ratio) if needed. Where more than one measure was used in a study, the measure most closely reflecting the narrow construct being explored was used, based on the DSM-5 criteria.

Due to the wide range of participant ages featuring in many of the included studies, it was not possible to conduct moderator analyses using different age subgroups. Only three studies (Aita et al., 2019; Hattier et al., 2011; Lawrence, 2017) featured participants with identified IQ <70, as all participants within these studies had a co-occurring diagnosis of intellectual disability, and therefore I was not able to investigate cognitive functioning as a possible moderating variable. There was also not sufficient studies for each behavioural exemplar to investigate this as a potential moderating variable.

Publication bias was assessed through Egger's test and visual inspection of funnel plots. Heterogeneity was assessed using chi-squared tests and interpretation of the  $I^2$  statistic (see below for further details).

## **2.3 Narrative Synthesis**

### **2.3.1 Stereotyped or Repetitive Motor Movements, Use of Objects, or Speech**

Eleven studies that explored sex/gender differences in 'stereotyped or repetitive motor movements, use of objects, or speech' found non-significant results (Aita et al., 2019; Brierley et al., 2021; English et al., 2021; Evans, 2018; Fetta et al., 2021; Fulceri et al., 2016; Lam, 2004; McFayden et al., 2019; Siracusano et al., 2021; Smerbeck, 2019; Solomon et al., 2012), though

eight of these found higher levels in males, despite being non-significant (Aita et al., 2019; Fetta et al., 2021; Fulceri et al., 2016; Lam, 2004; McFayden et al., 2019; Siracusano et al., 2021; Smerbeck, 2019; Solomon et al., 2012). Six studies concluded significantly higher endorsement of stereotyped behaviours in autistic males compared to females (Hattier et al., 2011; Lawrence, 2017; Mandy et al., 2012; May et al., 2016; Stephenson et al., 2021; Uljarević et al., 2020). Uljarevic et al., (2021) also reported male sex as a significant predictor of 'stereotypy'.

Albeit unable to be included in the meta-analysis (due to the data needed not being reported or provided), multiple studies also explored behavioural exemplars of stereotyped behaviours, reporting interesting findings. Autistic males have been identified as displaying significantly more preoccupations with part of objects (Antezana et al., 2019; Nicholas et al., 2008). Autistic girls were also significantly more likely to be reported as having 'little or no interest' in parts of mechanical objects compared to boys (Hiller et al., 2016). Harrop et al. (2015) and Hiller et al. (2014) also reported autistic boys displaying more stereotyped object use (e.g., arranging objects, repetitive or non-functional use, and object manipulation), though not to a level of significance. Autistic males have also been reported to display significantly more stereotyped mannerisms, such as hand and finger mannerisms (Antezana et al., 2019; Nicholas et al., 2008), than females. Sipes et al (2011) identified greater endorsement of certain repetitive motor behaviours (e.g., repetitive hand or arm movements and whole body movements) for autistic boys of average developmental quotient (DQ) compared to autistic females with average DQ, though significance levels were not reported. In contrast, however, some studies exploring the same behavioural exemplars report non-significant findings (Knutsen et al., 2019; Tang et al., 2021). Finally, non-significant sex/gender differences have also been reported for echolalia and/or idiosyncratic words or phrases (Boyd, 2020; Harrop et al., 2015; Knutsen et al., 2019; Nicholas et al., 2008), although some studies showed a trend towards a greater prevalence in males (Sipes et al., 2011; Tang et al., 2021).

### **2.3.2 Insistence on Sameness, Inflexible Adherence to Routine, or Ritualised Patterns of Verbal or Nonverbal Behaviour**

Research into to insistence on sameness (IS) appears to consistently conclude no significant sex/gender differences between autistic males and females (Aita et al., 2019; Barrett et al., 2018; Brierley et al., 2021; Dell'Osso et al., 2017; English et al., 2021; Evans, 2018; Fetta et al., 2021; Fulceri et al., 2016; Hus et al., 2007; Lam, 2004; May et al., 2016; McFayden et al., 2019; Sipes et al., 2011; Siracusano et al., 2021; Smerbeck, 2019; Solomon et al., 2012; Stephenson et al., 2021; Tang et al., 2021; Uljarević et al., 2020; Uljarevic et al., 2021; Wanzek, 2014).

At the behavioural exemplar level, Nicholas et al., (2008) reported autistic girls as being less likely to present with behaviours in the 'inflexible adherence to specific non-functional routines or rituals' subdomain (according to the DSM-IV criteria), compared to boys. However, Antezena et al (2019) reported higher endorsement of 'distress at small changes' in autistic females compared to males.

### **2.3.3 Highly Restricted, Fixated Interests that are Abnormal in Intensity or Focus**

A significant sex/gender difference in relation to passionate interests was reported in five out of 18 studies, with autistic males endorsing passionate interests more than autistic females (Antezana et al., 2019; Fetta et al., 2021; Grove et al., 2018; Knutsen et al., 2019; Solomon et al., 2012). Uljarevic et al (2021) also reported male sex as a significant predictor of 'restricted interests'. However, non-significant results have also been reported in adult (Dell-Osso et al., 2017) and child populations (Aita et al., 2019; Evans, 2018; Fulceri et al., 2016; Lam, 2004; May et al., 2016; McFayden et al., 2019; Nicholas et al., 2008; Siracusano et al., 2021; Uljarević et al., 2020; Wanzek, 2014). Smerbeck (2019) reported autistic females as displaying significantly higher 'atypicality' in their interests (e.g., interests being unusual for their age or sex or being an interest more commonly associated with the opposite gender/sex) compared to autistic males, after controlling for autism severity. However, in this meta-analysis, when autism severity was not controlled for, the results became non-significant.

In terms of the specific passionate interests reported, autistic males reported greater interests in object-related constructs and topics such as technology, mechanics and construction toys, transport, and science, whilst autistic females reported greater interests that hold more of a social quality to them and/or are related to living constructs such as autism, nature, psychology, animals, arts and crafts (Anthony et al., 2013; Caldwell-Harris & Jordan, 2014; Grove et al., 2018; Hiller et al., 2014; McFayden et al., 2019; Sutherland et al., 2017; Tang et al., 2021). One study also found that autistic infants paid more attention to different gendered interests (e.g., building toys and games consoles for males and dolls and dress-up toys for females) during eye tracking (Harrop et al., 2018), in line with the gender differences observed in non-autistic children and adults. There is also some suggestion that autistic females may be more likely to display a passionate interest in relation to the collection of "seemingly random" items such as rocks, pens and stickers compared to males (Hiller et al., 2014, 2016). Of note, however, there seems to be no sex/gender difference in terms of the intensity of passionate interests (Grove et al., 2018). This suggests that, whilst the content or the way the interest is expressed might be different, the intensity may not be.

### **2.3.4 Hyper- or Hypo-Reactivity to Sensory Input or Unusual Interests in Sensory Aspects of the Environment**

Most studies reported no significant sex/gender difference in sensory experiences (Bitsika et al., 2018; Feldman et al., 2020; Lane et al., 2014; Lawrence, 2017; Lee, 2008; Mandy et al., 2012; May et al., 2016; Stephenson et al., 2021; Williams et al., 2019). However, some indicated significantly more (Dell’Osso et al., 2017; English et al., 2021; Lever & Geurts, 2018; Weiland et al., 2020), or a trend towards significantly more (Fetta et al., 2021) sensory experiences reported by autistic females.

At the behavioural exemplar level, and not included in the meta-analysis due to the data needed not being reported or provided, Sutherland et al., (2017) identified autistic females as reporting significantly higher rates of sensory sensitivity in relation to the specific element of taste. Visual sensitivity has also been correlated with autistic traits in females whereas auditory sensitivity was correlated with autistic traits in males (Aykan et al., 2020).

Some studies reported on specific aspects of sensory experiences (such as hypo-sensitivity, sensory seeking, and unusual sensory interests), however these were not included in the meta-analysis due to being different to the definition of ‘sensory sensitivities’ and/or insufficient data available for analysis. Two studies found non-significant sex/gender differences for hypo-sensitivity and sensory seeking behaviours (Lane et al., 2014; Sutherland et al., 2017). Finally, non-significant sex/gender differences have been reported for ‘unusual sensory interests’ (Antezana et al., 2019; Harrop et al., 2015; Knutsen et al., 2019; Sipes et al., 2011; Tang et al., 2021). However, a trend towards autistic boys displaying more behaviours associated with unusual interests in smell, texture, and/or sound (although defined as ‘sensory seeking’ in the study) and visual interests (e.g., visual inspection, looking at objects from certain angles, bringing objects close to eyes, visual stimulatory behaviours) during recorded caregiver-child interaction (CCX) has been reported by Harrop et al., (2015).

## **2.4 Meta-Analysis**

### **2.4.1 Publication Bias and Heterogeneity**

Studies with statistically significant results are more likely to be published (Dickersin et al., 1987) which can create a publication bias (Borenstein et al., 2009) within systematic literature reviews and meta-analyses. Although this review included non-published studies, following a grey literature search, evidence of publication bias was assessed using funnel plots. On the X axis, the effect size (SMD) is reported and, on the Y axis, the standard error mean (SEM). Studies with



larger samples will plot towards the top of the graph and cluster around the mean, whilst smaller studies will plot towards the bottom of the graph and will have a wider spread of values (indicating sampling error). If there is no publication bias, the graph should resemble a funnel with studies being symmetrically distributed around the mean effect size. Asymmetry indicates publication bias where studies reporting non-significant findings may not have been published.

One plot (passionate interests) indicated asymmetry with Eggers test being significant ( $p < .001$ ) and therefore indicating publication bias (see Figure 7, in appendix D). Three plots (stereotyped behaviour, insistence on sameness, and sensory experiences) indicated symmetry, with most studies clustered around the overall SMD, suggesting no publication bias (see Figures 8, 9, and 10 in appendix D). Consistently, Eggers test was non-significant for these three analyses (for stereotyped behaviour,  $p = .19$ ; for insistence on sameness,  $p = .36$ ; and sensory,  $p = .31$ )

Heterogeneity relates to the variation in effect sizes across studies. Large heterogeneity suggests that the differences found could be due to moderating variables and not solely that of the variables being studied. Heterogeneity was assessed using chi-squared tests, with a significant p-value of  $< .05$  indicating heterogeneity, and interpretation of the  $I^2$  statistic using conventions outlined by Cochrane (Higgins et al., 2003).

#### **2.4.2 Stereotyped or Repetitive Motor Movements, Use of Objects, or Speech**

A random-effects meta-analysis found significant differences between autistic males and females, SMD = .22, 95% confidence interval (CI) [0.09, 0.34],  $p < .001$ , indicating that autistic males had significantly higher rates of stereotyped behaviour than autistic females (see Figure 2), with heterogeneity tests being significant,  $Q = 28.12$ ,  $p < .05$ ,  $I^2$  indicated moderate heterogeneity ( $I^2 = 45.66$ ). As Wanzek (2014) reported very different results to all other studies, a sensitivity analysis was conducted by re-running the analysis with the removal of this data point. However, the results remained the same, and therefore I have reported results with the inclusion of this study.

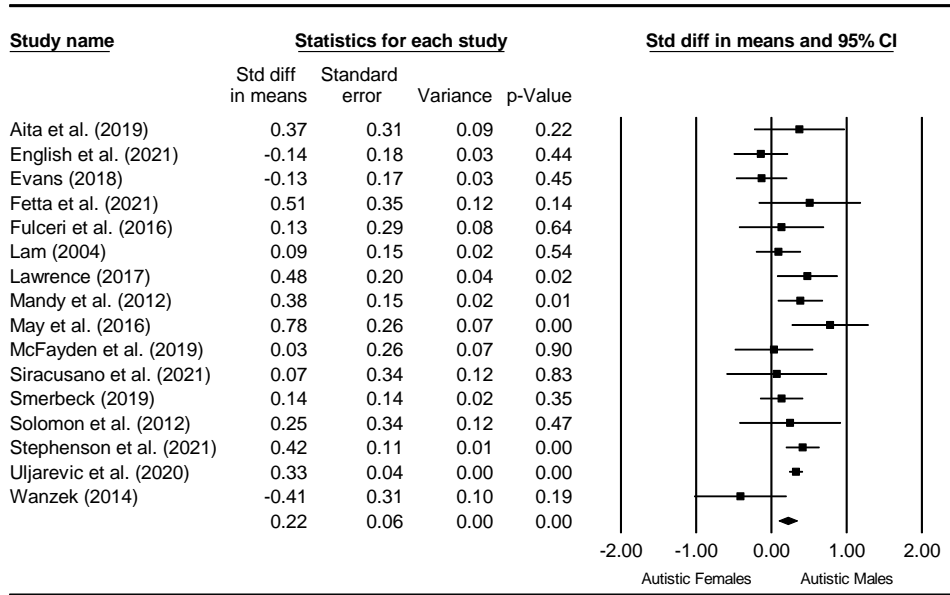


Figure 2. Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of stereotyped behaviours.

*Note.* <sup>a</sup> the data from McFayden et al (2019) used in this meta-analysis is not the final data set used and reported in their published paper. <sup>b</sup> McFayden et al (2019) data used in meta-analysis refers to RBS-R Endorsed mean subscale scores whereas in their published paper McFayden et al (2019) refer to RBS-R Total scores. <sup>c</sup> the data from English et al (2021) refers to autistic (diagnosed and self-identifying) only.

### 2.4.3 Insistence on Sameness, Inflexible Adherence to Routine, or Ritualised Patterns of Verbal or Nonverbal Behaviour

A random-effects meta-analysis found no significant difference between autistic males and females, SMD = .02, 95% CI [-0.05, 0.09], p=.54), indicating that autistic males and females had similar rates of insistence on sameness (see Figure 3). Heterogeneity tests were not significant, Q=10.56, p=.72, I<sup>2</sup> indicated minimal heterogeneity (I<sup>2</sup>= .00).

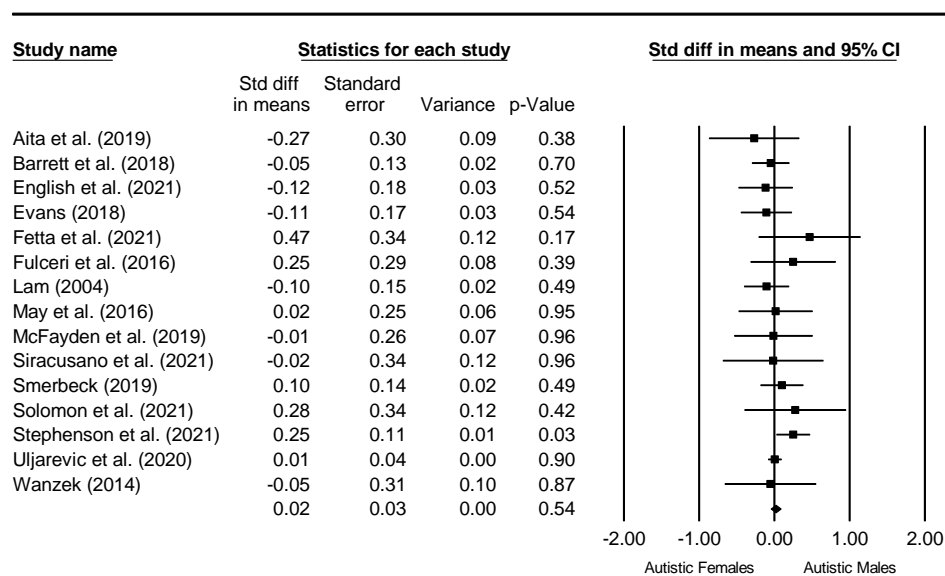


Figure 3. Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of insistence on sameness.

*Note.* <sup>a</sup> the data from McFayden et al (2019) used in this meta-analysis is not the final data set used and reported in their published paper. <sup>b</sup> McFayden et al (2019) data used in meta-analysis refers to RBS-R Endorsed mean subscale scores whereas McFayden et al (2019) refer to RBS-R Total scores. <sup>c</sup> the data from English et al (2021) refers to autistic (diagnosed and self-identifying) only. <sup>d</sup> the p-value for Stephenson et al (2021) is different to the p-value reported in the published paper due to different statistical tests being used (e.g., Welch's Two-Sample T test used in the published paper).

#### 2.4.4 Highly Restricted, Fixated Interests that are Abnormal in Intensity or Focus

A random-effects meta-analysis found significant differences between autistic males and females, SMD = .24, 95% CI [0.07, 0.41],  $p < .01$ , indicating that autistic males had significantly higher rates of passionate interests than autistic females (see Figure 4). Heterogeneity tests were significant,  $Q = 21.78$ ,  $p = .03$ ,  $I^2$  indicated a moderate amount of heterogeneity ( $I^2 = 49.49$ ), indicating significant variation in effect sizes that cannot be accounted for by sex/gender alone.

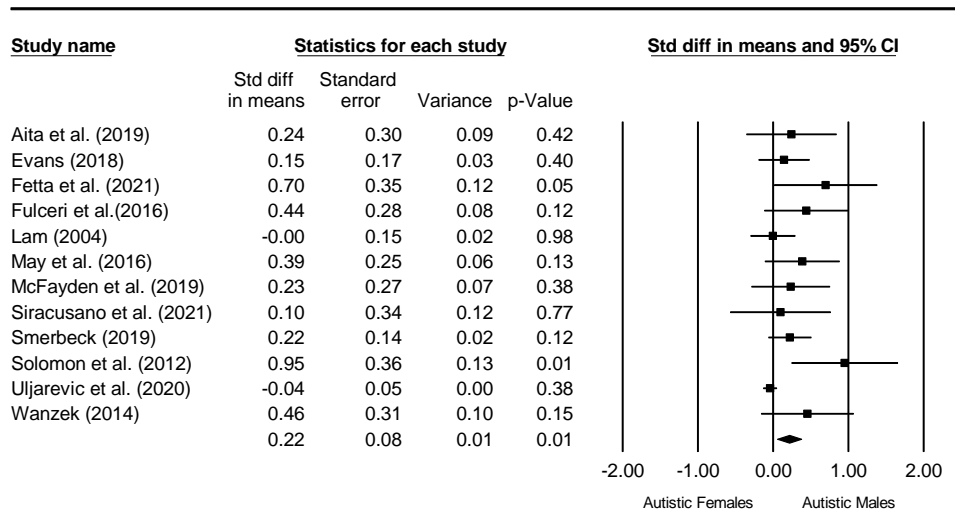


Figure 4. Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of passionate interests.

*Note.* <sup>a</sup> the data from McFayden et al (2019) used in this meta-analysis is not the final data set used and reported in their published paper. Likewise, data in this meta-analysis refers to RBS-R Endorsed mean subscale scores whereas McFayden et al (2019) refer to RBS-R Total scores. <sup>b</sup> Data presented for Smerbeck (2019) is prior to controlling for autism severity.

Given the frequent reporting of ‘RBS-R Total’ scores (which includes parent-reported perception of the severity of the behaviour) in studies using the RBS-R, a post-hoc random-effects meta-analysis was also conducted using only studies reporting RBS-R Endorsed scores (referencing to prevalence of a behaviour rather than parent-reported endorsement *and* perceived severity) and studies using other measures for each narrow construct. For ‘highly restricted, fixated interests that are abnormal in intensity or focus’, this produced a non-significant result, SMD = .16, 95% CI [-0.06, 0.38], p=.16) (see Figure 5).

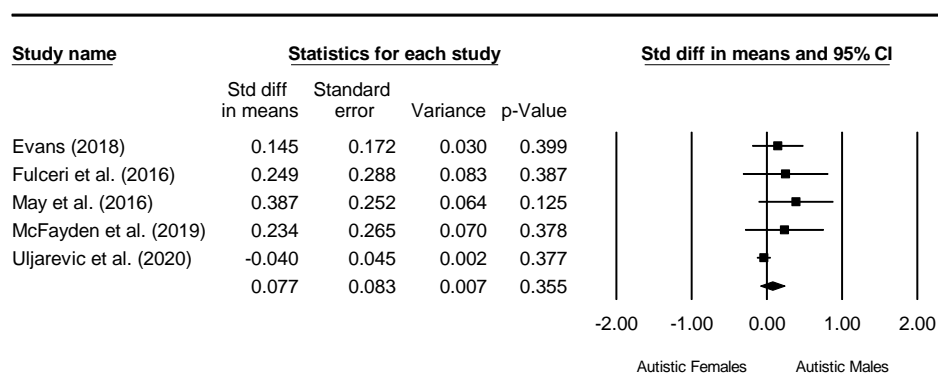


Figure 5. Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of passionate interests with RBS-R Endorsed total mean scores only.

Note. <sup>a</sup> the data from McFayden et al (2019) used in this meta-analysis is not the final data set used and reported in their published paper. Likewise, data in this meta-analysis refers to RBS-R Endorsed mean subscale scores whereas McFayden et al (2019) refer to RBS-R Total scores.

#### 2.4.5 Hyper- or Hypo-Reactivity to Sensory Input or Unusual Interests in Sensory Aspects of the Environment

A random-effects meta-analysis found a trend towards autistic females have higher rates of sensory experiences,  $SMD = -.16$ ,  $95\% CI [-0.35, 0.03]$ ,  $p=.09$ . (see Figure 6). Heterogeneity tests were significant,  $Q=28.95$ ,  $p=.001$ ,  $I^2$  indicated a substantial amount of heterogeneity ( $I^2= 65.46$ ), indicating significant variation in effect sizes that cannot be accounted for by sex/gender alone.

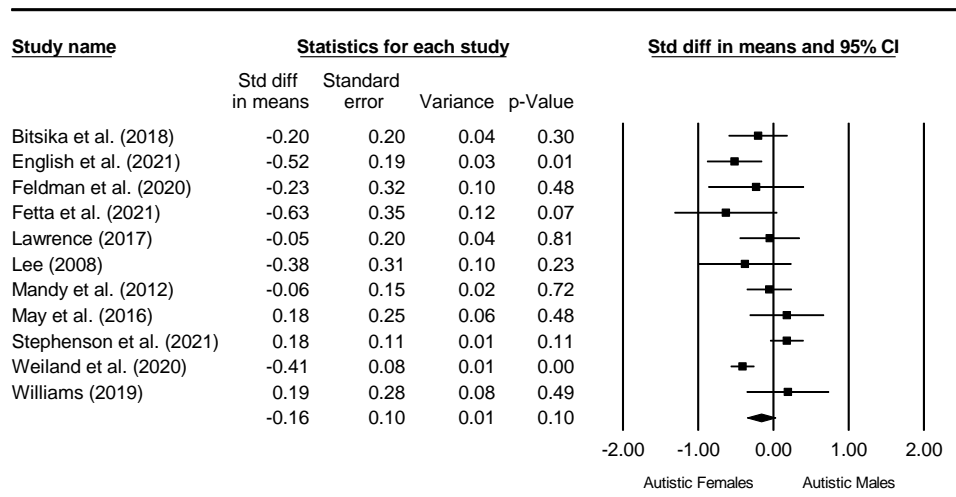


Figure 6. Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of sensory experiences.

*Note.* Six studies reported data pertaining to ‘sensory sensitivity’ scores (Bitsika et al., 2018; Feldman et al., 2020; Lee, 2008; Mandy et al., 2012; May et al., 2016; Stephenson et al., 2021) meanwhile the remaining five reported data pertaining to ‘total sensory’ scores (English et al., 2021; Fetta et al., 2021; Lawrence, 2017; Weiland et al., 2020; Williams et al., 2019).<sup>a</sup> the data from McFayden et al (2019) used in this meta-analysis is not the final data set used and reported in their published paper. Likewise, data in this meta-analysis refers to RBS-R Endorsed mean subscale scores whereas McFayden et al (2019) refer to RBS-R Total scores.<sup>b</sup> the data from English et al (2021) refers to autistic (diagnosed and self-identifying) only.

## 2.5 Discussion

This systematic review and meta-analysis explored sex/gender differences in the narrow constructs and behavioural exemplars of autistic restricted and repetitive behaviours and interests (RRBIs). Research findings were narratively synthesised from 44 studies and data from 22 studies were included in four random-effects meta-analyses, in line with the subdomains of RRBIs outlined by the DSM-5. The meta-analytic findings indicate that there are significant differences between autistic males and females and the presentation of stereotyped behaviours and passionate interests, with autistic males presenting with higher levels of these behaviours than autistic females. A trend towards females having higher levels of sensory experiences was also found. No significant sex/gender differences were identified for insistence on sameness. The narrative synthesis findings were broadly consistent with those from the meta-analyses and also

revealed interesting findings regarding qualitatively different types of passionate interests autistic females hold in comparison to males, discussed below.

The sex/gender differences found for passionate interests is in line with previous studies indicating fewer passionate interests in autistic females compared to males (Uljarević et al., 2021; Uljarević et al., 2021). The fact that autistic females' passionate interests appear to be more socially appropriate and developmentally normative than males' interests (McFayden et al., 2019; Sutherland et al., 2017), for example interests in animals, could contribute towards autistic females being missed by practitioners and, subsequently, less likely to be referred for an autism assessment. Consistently, Whitlock et al., (2020) found that, when educational staff were presented with vignettes of the female autism phenotype (which included social or relation restricted interests), they reported that they were "unlikely" to seek support from an external professional. The type of passionate interests displayed by autistic females may also be difficult to identify using diagnostic tools, such as the ADOS and ADI-R, as there are few items pertaining to these, reflecting the criticism of such tools being male-biased (Bargiela et al., 2016; Hiller et al., 2014; Mandy et al., 2012), which could further lead to under-recognition of autism for females.

As a result, it is important for practitioners at both the early identification and diagnostic stage to be aware of how autistic passionate interests may present in females. For example, it will be important for educational staff, who will often be the first professionals to raise the possibility that a young person may be on the autism spectrum, to receive training on these differences. Similarly, clinicians completing diagnostic assessments should ensure that they directly ask parents/carers and teachers about passionate interests that may seem 'typical' and/or not adhere to the male autism stereotype (e.g., trains, number plates), or ideally ask the child or young person themselves. Clinicians should also consider these differences as part of observations, being mindful that autistic females may score lower on items pertaining to passionate interests on diagnostic measures. Given some professionals report feeling unconfident diagnosing autism in females (Tromans et al., 2019), it will be important for appropriate training to be provided to diagnosing clinicians, specifically on how autism manifests in females.

This review has also highlighted the importance of establishing what RRBI outcome measures are actually measuring, for example the RBS-R Total score refers to both prevalence and perceived severity of passionate interests by caregivers, whereas the RBS-R Endorsed score refers to just prevalence. This is an important distinction as, when RBS-R Total scores were substituted with RBS-R Endorsed scores (or excluded if substitution was not possible) for passionate interests, the analysis as a whole was no longer significant. This suggests that a significant driver behind the higher prevalence of passionate interests on the RBS-R for males is the *perceived* severity of the

interest rather than the prevalence. Given this review has identified that autistic females display more socially and developmentally normative interests, it is plausible that practitioners and parents/carers deem these less 'severe' and subsequently autistic females may again be missed.

Although the findings suggest that passionate interests are lower in autistic females, it is important to acknowledge that this could reflect the insensitivity of RRBI outcome measures at capturing autistic female interests. For example, the RBS-R has been criticised for only providing examples of male-centric interests (e.g., trains; Fulceri et al., 2016) on passionate interest items and is lacking a free-report option, which could prevent autistic female interests that are qualitatively different from being captured (Antezana et al., 2019). However, one study featuring autistic adults (Grove et al., 2018) did find that autistic males free-reported higher levels of passionate interests than females, suggesting they may have increased levels of such interests even when not constrained by measurement tools. However, the intensity of interests were similar between sexes/genders, suggesting that the nature of autistic interests being 'narrow' and 'deep' is consistent across genders but that the frequency of these interests may differ. Consequently, practitioners should consider the intensity as well as type of interest held. For example, females may hold very intense interests but, again, due to these being considered gender- and developmentally-normative, may not be recognised by practitioners.

Future research exploring autistic passionate interests will need to ascertain the respective levels of passionate interests in males and females in a way, as far as possible, that is free from gender bias and constraints of current instruments. This could include using self-report (without constraint on items) measures, tools and observations that include both male and female (or gender-neutral) passionate interests (Frazier, Ratliff, et al., 2014; Solomon et al., 2012; Sutherland et al., 2017), which will require the adaptation of current instruments or development of new instruments that can be used in both research and practice.

The significant sex/gender differences identified for stereotyped behaviours is also in line with previous research, with autistic males displaying more stereotyped behaviours than females (Beggiato et al., 2017; Hartley & Sikora, 2009; Kaat et al., 2021; Tsirgiotis et al., 2021). As with passionate interests, the lower level of stereotyped behaviours in autistic females could contribute towards the under-recognition of autistic females, particularly as these behaviours are more externally observable and perhaps more in accordance with the male-stereotyped nosology of autism. Certain stereotyped behaviours might also be easier for observers to identify in autistic boys because boys are more likely to have access to male-gendered toys (e.g., cars and construction toys) that provide greater opportunity for the repetitive use or interest in parts typically associated with autism (e.g., spinning wheels, moving mechanisms). In comparison,



stereotypically female toys (e.g., dolls or dress-up) are associated more with imaginative and social-oriented play, meaning that subtle stereotyped behaviours with these items may be missed or misinterpreted.

Gal (2011) argued that culture-oriented judgements could also influence how stereotyped and repetitive behaviours are interpreted, particularly given the emphasis of such behaviours being deemed 'odd' or 'unusual'. This may be especially pertinent for the identification of autistic females as some stereotyped behaviours could be deemed less unusual in females compared to their male counterparts, for example lining up toys being interpreted as atypical in boys whilst a sign of neatness or organisation when displayed by girls. There is also emerging research that suggests that autistic females might be motivated to mask autistic features, known as camouflaging (Hull et al., 2020), which could be contributing towards the lower levels of stereotyped behaviours observed by others. The majority of research into camouflaging has focussed on the social interaction and communication domain (e.g., Cook et al., 2021; Dean et al., 2017; Wood-Downie., et al., 2020 for a review), however autistic adults have described suppressing stimming behaviours as a camouflaging strategy (Hull et al., 2017; Kreiser & White, 2014). The motivation to mask stereotyped behaviours may be particularly stronger in autistic females with average or higher-than-average IQ, due to greater social awareness, which could explain the reduced presentation of repetitive motor movements in studies exploring these behaviours in autistic individuals of varying cognitive ability (e.g., Sipes et al., 2011). Future research needs to understand how camouflaging could be impacting the sex/gender differences in stereotyped behaviours, and RRBIs as a whole, and how this may change across the lifespan. Exploration as to how stereotyped behaviours are observed and interpreted by those key to early recognition of autism (e.g., educational practitioners) is also required so that we can better understand how this influences the under-recognition of autism females, particularly during childhood where observation reports are more heavily relied upon.

The non-significant results for insistence on sameness is different to previous research that identifies autistic females as displaying lower levels of overall RRBIs compared to males, both in large-scale studies (e.g., Kaat et al., 2021) and in systematic reviews and meta-analyses (e.g., van Wijngaarden-Cremers et al., 2014). This illustrates the importance of future research exploring RRBIs at a fine-grained level, in order to identify specific differences and similarities between males and females, particularly as sex/gender differences can be identified at the behavioural exemplar level (Antezana et al., 2019). Similarly, practitioners and clinicians will also need to be aware of the narrow constructs within RRBIs and consider whether these are all currently being accurately and routinely captured by the referral and diagnostic tools used.

To my knowledge, this is the first time that sex/gender differences in autistic sensory experiences have been systematically synthesised. The findings from the meta-analysis suggest that autistic females report or display higher rates of sensory experiences than males, though this finding did not reach statistical significance. Consistently, some of the findings detailed in the qualitative synthesis indicate higher levels of sensory experiences for females compared to males (Dell’Osso et al., 2017; English et al., 2021; Lawrence, 2017; Lever & Geurts, 2018; Sutherland et al., 2017; Tang et al., 2021; Weiland et al., 2020).

In comparison to other domains (e.g., stereotyped behaviours, passionate interests), sensory experiences may not be immediately apparent to others as they may not be so externally observable via behaviour. This again could contribute to under-recognition of autistic females, particularly for children and young people for whom measures rely on reports from others. Furthermore, diagnostic tools, such as the ADOS-2 and ADI-R, only capture ‘unusual sensory interests’ and ‘undue sensitivity to noise’ (e.g., hyposensitivity), omitting other elements of sensory experiences, such as hypo-sensitivity, and experiences relating to the other four senses (touch, taste, smell, and sight). Arguably, the function behind sensory experiences may also be subject to interpretation bias when reported by others, for example a sensory-seeking behaviour could fall either within hyposensitivity (seeking to receive sensory input), unusual interest in a sensory element (seeking to experience pleasure), or a category in its own right. Considering the majority of studies for this narrow construct in this review were based on teacher, clinician or parent report, the rates of sensory experiences in females may be under-estimated, and future research needs to utilise self-report, along with more objective measures of sensory response (e.g., neurophysiological measures) relating to hyper- and hypo-sensitivity and unusual sensory interests (for all key senses).

### **2.5.1 Implications for Practice**

One of the key implications for practice is to improve professionals’ awareness of how autism may manifest in females. Professionals involved in the early identification and referral process (e.g., education staff) need to be aware that passionate interests may appear more gender- and/or socially-normative in autistic females compared to males and thus may not be as easily identifiable in the school environment. Furthermore, practitioners would benefit from being aware of the potential role of camouflaging and how some autistic individuals may seek to hide or mask some of their stereotyped behaviours in order to fit in. Additional training for educational staff on the topic of autistic sex/gender differences, through a neurodiversity lens that does not pathologize autistic behaviours and interests, would be required to support this.

Similarly, professionals involved in the diagnostic process will also need to be aware of the role of camouflaging and some of the unique differences in autistic female interests and actively explore these by asking parents/carers, and indeed children and young people themselves, about camouflaging behaviours and interests that may seem 'typical' (although intense in nature). This will be particularly important in light of the identified lack of sensitivity some current diagnostic instruments have in capturing the experiences of autistic females, resulting in lower diagnostic rates and the frequent miss-diagnosis of autistic females with other mental health needs.

A second key implication is the need to adapt current diagnostic instruments or to develop new instruments that hold a greater emphasis on self-report and provide more opportunities for a range of gendered (or gender-neutral) passionate interests to be captured, so that a wider range of autistic individuals' experiences can be identified. The use of self-report within the exploration of narrow constructs of RRBIs will be especially important given the influence gender, culture and societal expectations can have on observer interpretation of autistic behaviours. Individuals being able to self-report their experiences of more internalised narrow constructs (e.g., insistence on sameness and sensory experiences) would be valuable both in terms of diagnostic assessment and research that informs the nosology of autism. Practitioners (e.g., Educational Psychologists) may need to adapt their practice (e.g., utilising person centred approaches) so that autistic individuals are able to self-report on their RRBIs.

### **2.5.2 Limitations**

A limitation of this review is the limited number of studies that could be included in the meta-analyses, due to unavailability of data or researchers not responding to my requests for data, though findings from the narrative synthesis and meta-analyses were consistent. Furthermore, a large proportion of studies were rated as 'unsatisfactory' in quality analysis, primary due to unrepresentative samples, and/or lack of autism diagnosis confirmation using validated measures, which may have influenced the findings, such as not being generalisable to all individuals on the autism spectrum (e.g., those with co-occurring learning disabilities). In future, researchers should focus on building the representativeness of autistic samples using wide scale recruitment (e.g., population-based studies), rather than relying on convenience sampling, and incorporating diagnosis confirmation by using diagnostic measures or autistic trait measures.

A significant variation in effect size across studies was identified for passionate interests, stereotyped behaviours, and sensory experiences, suggesting heterogeneity that cannot be accounted for by sex/gender alone. Previous research suggests that IQ and age may moderate sex/gender difference in autism generally (Jiujiyas et al., 2017; Stratis & Lecavalier, 2013; Wood-

Downie et al., 2021) therefore, it is possible that IQ and age account for some of the heterogeneity within the current analysis. Unfortunately, due to under-representation of individuals with lower IQ, I was unable to conduct analyses to see whether IQ was a moderator. Similarly, due to studies including very wide age range of participants, I was unable to see whether this was a moderating variable as well. Additionally, due to the small amount of studies for each behavioural exemplar, I was unable to investigate this as moderating variable, which may be accounting for some of the heterogeneity. Finally, the type of outcome measure used may have affected results, such as in the sensory experiences analysis, in which a wide range of outcome measures were used which could explain some of the variation in results. As such, it will be important for future research to include individuals with intellectual disabilities, narrower age ranges in samples, as well as using a wider range of behavioural exemplars as outcome measures, so that future analyses can investigate whether these moderate sex/gender differences.

There are also some wider factors that should be considered with regards to future autism research and theory. Firstly, research into autistic sex/gender differences needs to be less dichotomous both in terms of gender (e.g., including non-binary individuals) and diagnostic status (e.g., including self-identified autistic individuals). The latter is particularly important given issues of intersectionality the identified male-bias in assessment and research that may be particularly excluding autistic females. Secondly, it is important that theories regarding sex/gender differences recognise that the female autism phenotype (e.g., camouflaging; differences in behavioural presentation) is not unique to females and can present across genders (Cook et al., 2021; Lai & Szatmari, 2020). For example, although the current review has identified some average group level differences between males and females, these findings will not apply to all males and females. Nonetheless, I believe the current findings have important implications for both research and practice, as have been discussed.

### **2.5.3 Conclusion**

In conclusion, this review identified significant sex/gender differences in two narrow constructs of RRBIs (as defined by the DSM-5), stereotyped behaviours and passionate interests, with autistic males reporting higher rates compared to autistic females, as well as a trend towards females reporting more sensory experiences. Whereas no sex/gender differences were concluded for insistence on sameness. This highlights the importance of fine-grained analysis into the narrow constructs of RRBIs rather than merely exploring at the broad construct level. These results could, in part, explain the late and under-diagnosis of autism for females and highlights the importance of developing assessment tools that are sensitive to females or that are less-impacted by gender

bias. More research is needed to explore potential moderating variables of IQ, age, and specific behavioural exemplars.



## **Chapter 3 Are there sex/gender differences in how autistic young people, with and without a diagnosis, engage in restricted and repetitive behaviours and interests?**

Females are routinely under- or late-diagnosed as autistic compared to males. This under-representation has been attributed to factors including the male-biased nature of autism research and subsequent biases in referral and assessment, as well as the higher prevalence of camouflaging behaviours in the female autistic population, resulting in behaviours and presentations being missed. Research into the second domain of the autism diagnostic criteria, restricted and repetitive behaviours and interests (RRBIs), highlights that autistic females present with fewer RRBIs to males. However there is less research into how these differences present at the narrow construct level, for example insistence on sameness (IS) and repetitive sensory motor behaviours (RSMB). In addition, few studies use self-report measures and include self-identifying autistic participants, which is particularly important given the possibility that that are group of autistic individuals who may appear to have lower levels of RRBIs to others than they actually do, due to masking or camouflaging such behaviours, which could impact on their access to appropriate support and services..

The present research explores sex/gender differences in the self-reported RRBIs of young people using the Repetitive Behaviours Questionnaire (RBQ-2A), completed by autistic (including both diagnosed and self-identifying) and non-autistic 16-25 year olds. The results indicate that overall, autistic individuals have higher levels of RSMB and IS scores on the RBQ-2A compared to non-autistic individuals. Autistic females also self-report significantly more IS behaviours compared to autistic males and similar levels of RSMB to autistic males. Diagnosed autistic and self-identifying autistic participants also self-reported similar high levels of autistic traits, supporting the inclusion of self-identifying autistic participants in autism research. The findings in relation to previous literature and implications for practice are discussed, as well as future recommendations for autism sex/gender differences research.

### **3.1 Introduction**

Autism is defined as differences in social communication and interaction and restricted and repetitive behaviours and interests (American Psychiatric Association, 2013). Autistic individuals have also self-reported a number of strengths including attention to detail, specialist knowledge of areas of interest, creativity, honesty, and loyalty (Russell et al., 2019). The estimated prevalence of autism in the UK ranges between 1% to 1.7% (Baron-Cohen et al., 2009; Russell et al., 2014), with around 1 in 57 children in England diagnosed autistic (Roman-Urrestarazu et al.,

2021), and the sex/gender<sup>6</sup> ratio for diagnosed autism is around four males to every one female (Fombonne et al., 2009; Kreiser & White, 2014). However, the ratio is around 3:1 when using population-based studies, including participants screened for high autism traits rather than just those with a clinical diagnosis (Loomes et al., 2017). This suggests that there is a proportion of females who are not receiving a diagnosis, despite meeting clinical criteria (Russell et al., 2011), especially during childhood (Begeer et al., 2013; Kirkovski et al., 2013; Rivet & Matson, 2011).

Hypotheses regarding the higher male autism diagnostic rates relate to a variety of biological, psychological and social factors. Whilst some propose biological differences (i.e., the extreme male brain theory; see Baron-Cohen, 2002; Ingudomnukul et al., 2007), others highlight behavioural differences in how autistic males and females present, including the use of camouflaging and masking (whereby an individual employs strategies or behaviours to hide their autistic traits in order to fit in with the social world), social differences, and more age and gender appropriate 'restricted and repetitive' interests (see the female autism phenotype; Hull et al., 2020; Kirkovski et al., 2013; Wood-Downie et al., 2020). Autism assessment tools have been predominantly developed from samples of males, which therefore may not capture phenotypic differences between males and females, suggesting they may be male-biased (Bargiela et al., 2016; Hiller et al., 2014; Mandy et al., 2012) and lacking sensitivity in identifying sex/gender differences (Lai et al., 2015; Lai & Baron-Cohen, 2015; Wood-Downie et al., 2021).

Sex/gender differences in autistic restricted and repetitive behaviours and interests (RRBIs) have been identified, with females consistently being reported to display fewer RRBIs than males (see Frazier et al., 2014; Supekar & Menon, 2015; Szatmari et al., 2012 and see reviews by Lai et al., 2015; van Wijngaarden-Cremers et al., 2014). However, these studies predominantly only explored RRBIs at the broad construct level (i.e., the more general and abstract definition of autism symptomatology; see Lai et al., 2015), referring to overall RRBI scores on outcome measures, and therefore could have missed potential subtle differences at the narrow construct level (e.g., subdomains within the broad construct such as stereotyped behaviours, insistence on sameness, passionate interests, and sensory experiences).

In contrast to the aforementioned studies, research into sex/gender differences at the narrow constructs of RRBIs have produced mixed results. Autistic males are reported to display significantly more stereotyped behaviours (e.g., Mandy et al., 2012; May et al., 2016; Uljarević et al., 2020) and passionate interests (e.g., Fetta et al., 2021; Solomon et al., 2012) than females,

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<sup>6</sup> The effects of biological sex and socially constructed gender are difficult to separate, therefore in this review the term 'sex/gender' will be used to reflect that most people's identities are informed by both sex and gender (as commented by Lai, Lombardo, Auyeung, Chakrabarti & Baron-Cohen, 2015).



however there are also studies reporting non-significant results for these narrow constructs (e.g., McFayden et al., 2019; Siracusano et al., 2021, respectively). Similarly, the results are mixed for the sensory experiences narrow construct, with some studies reporting no significant sex/gender differences (e.g., Feldman et al., 2020; May et al., 2016) whilst others suggest that autistic females report more sensory experiences than autistic males (Dell’Osso et al., 2017; English et al., 2021; Weiland et al., 2020). Interestingly, no significant sex/gender differences are routinely reported for insistence on sameness (May et al., 2016; McFayden et al., 2019; Uljarević et al., 2020; Uljarevic et al., 2021). These mixed results are incongruent with findings at the broad construct level suggesting that autistic males display or report more RRBIs than females. This highlights the importance of exploring RRBIs at the narrow construct level to identify possible specific differences in autistic males and females, which may not be apparent when focusing on overall levels of RRBIs.

Females tend to be under-represented in research (Watkins et al., 2014), and those that are included tend to have a clinical diagnosis of autism. Previous research suggests that there may be a group of females who are not receiving a diagnosis, despite meeting clinical criteria, suggesting biases in referral and assessment processes (Loomes et al., 2017), and that there may be differences between females on the autism spectrum who do and do not receive a diagnosis (Belcher et al., 2022). This would bias findings of research that only includes diagnosed autistic females, as they may present in a way that is more aligned to the male-stereotyped nosology of autism. Therefore, it is important for research to include females who may have high autistic traits, despite having not receive a clinical diagnosis.

The aim of this study was to explore sex/gender differences in self-reported RRBIs at the narrow construct level, focussing on the two main subcategories of ‘repetitive sensory-motor behaviours’ (RSMB) and ‘insistence on sameness’ (IS). These two subcategories have been routinely identified in factor-analyses of RRBIs in both autistic (Georgiades et al., 2010; Mooney et al., 2009; Richler et al., 2010) and non-autistic populations (Evans et al., 2017; Uljarevic et al., 2017). A unique element to this study was the inclusion of self-identifying autistic participants which, to the researchers’ knowledge, has only been utilised in one other study exploring sex/gender differences of RRBIs (see English et al., 2021). When comparing ‘cognitive rigidity’ (aligned with the IS subdomain), ‘repetitive behaviour’, and ‘sensory sensitivity’ scores on the Comprehensive Autistic Trait Inventory (CATI) between autistic and non-autistic men and women, English et al., (2021) found significantly higher scores in males for repetitive behaviour and significantly higher scores in females for sensory sensitivities. They concluded no significant sex/gender difference for cognitive rigidity. However, English et al., (2021) did not report on sex/gender differences for these subdomains solely in the autistic group. Given the identified

under-representation of autistic females in research, particularly in RRBI research using measures that rely on parent/carer or other reports, it was also considered pertinent to use a self-report measure in order to more accurately capture the experiences of RRBI for autistic people themselves, particularly as females may camouflage/suppress RRBI.

Due to the mixed findings of previous research and the novelty of the current study, it was not possible for us to form specific hypotheses.

### 3.2 Method

#### 3.2.1 Participants and Procedure

A sample of 16-25 year olds was initially systematically recruited, followed by use of opportunistic and convenience sampling strategies, to complete an online survey by emailing head teachers, SENCOs and/or heads of pastoral in post-16 colleges, student support services at universities, and autism organisations, support agencies and charities in the UK. The study was also advertised on social media. Participants were eligible if they lived in the United Kingdom and if they self-reported having a minimum reading age equivalent to GCSE level. The latter was required in order to ensure that participants could read, understand and complete the online survey independently. Due to the online nature of this study, it was not possible to independently verify reading age and/or capability. Participants self-reported whether they were diagnosed autistic, self-identified as autistic, or non-autistic. Diagnosed autistic ( $n=22$ ) and self-identifying autistic ( $n=11$ ) participants were combined to form one 'autistic' group. Both groups scored above threshold ( $>8$ ) on a measure of autistic traits (see measures section below), with the diagnosed autistic group scoring 9 and the self-identifying group scoring 8.9. An independent samples t-test indicated no significant difference on levels of autistic traits ( $p = .92$ ) between these two groups, nor the two main outcome measures of IS ( $p = .14$ ) and RSMB ( $p = .31$ ), suggesting it was appropriate to include them in one group.

The final sample comprised 84 participants (15 males reporting a diagnosis ( $n=10$ ) of autism or self-identifying ( $n=5$ ) as autistic, 18 non-autistic males, 18 females reporting a diagnosis ( $n=12$ ) of autism or self-identifying ( $n=6$ ) as autistic, and 33 non-autistic females) aged between 16- 25 years (see Table 7. for sample characteristics).

### 3.2.2 Measures

#### 3.2.2.1 Demographics

Demographic questions (e.g., participant's age, gender identity, ethnic background, biological sex, diagnosis status (non-autistic, diagnosed autistic, or self-identifying autistic), and the nature of this diagnosis (if applicable) such as age of diagnosis, who made this diagnosis (e.g., psychiatrist, paediatrician, psychologist), and how many times the participant was assessed before receiving the diagnosis) were collected.

#### 3.2.2.2 Autistic Traits

Autistic traits were established using the Autism Symptom SElf-RepOrT (ASSERT; Posserud et al., 2013), which is a self-report measure adapted from the Asperger syndrome and high functioning autism diagnostic interview (ASDI; Gillberg et al., 2001). The measure comprises of four items exploring social understanding (e.g., 'do you have difficulties perceiving social cues?') and three exploring restricted and repetitive behaviours and interests (e.g., 'do you or do other people feel that you impose your routines or interests on others?') that are answered on a three-point Likert scale (*not true, somewhat true, certainly true*), leading to a total score range of 0-14 (see appendix D). A cut-off of >8 is considered indicative of the possibility of an individual being on the autism spectrum. A large, bi-factor analysis and validation of the measure with a sample of 10,220 adolescents (aged 16-19 years) shows that the measure has promising validity (Posserud et al., 2013). Within the current study sample, the ASSERT was found to have good internal consistency (7 items;  $\alpha = .82$ ).

#### 3.2.2.3 Restricted and Repetitive Behaviours and Interests

To explore narrow constructs of RRBI (e.g., repetitive sensory motor behaviours and insistence on sameness), the adult Repetitive Behaviour Questionnaire-2 (RBQ-2A; Barrett et al., 2015) was used (see appendix D). The RBQ-2A is a self-report version of the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007) adapted for use with adult populations. The 20-item measure features items relating to two subtypes; insistence on sameness (IS; including routines and circumscribed interests) and repetitive sensory motor behaviours (RSMB; such as hand-flapping and rocking and special interests in the smell of people or objects). Responders are asked to rate the repetitive behaviours they have shown over the last month and rate the most usual way that they display the behaviour, for example 'make repetitive hand and/or finger movements' (RSMB) and 'insist that aspects of daily routine must remain the same' (IS). Items are answered using three or four-point Likert scales (e.g., '*never or rarely, one or more times daily, 15 or more times daily, 30 or more times daily*' or '*never or rarely, mild or occasional, or marked or*

*notable* respectively), however the fourth option is collapsed into option three when scoring. Item 20 (activities) is not included in scoring due to being qualitatively different to other items (Leekam et al., 2007; Lidstone et al., 2014). The RBQ-2A is scored in terms of total mean score (ranging from 1 to 3) with a higher score indicating higher levels. There is no clinical threshold cut-off.

Good reliability and construct validity has been reported for the RBQ-2A, with internal consistency (Cronbach's alpha) of the whole scale rated as acceptable ( $\alpha = 0.83$ ) when used with samples of non-autistic adults (Barrett et al., 2015) and autistic 13-20 year olds ( $\alpha = 0.89$ ; Joyce et al., 2017). Within the current study sample, the RBQ-2A was found to have excellent internal consistency (20 items;  $\alpha = .92$ ). Studies have also reported adequate internal consistency for the two subscales of IS and RSMB both in non-autistic adult samples (Jia et al., 2019) and autistic adults (Barrett et al., 2018) and adolescents (Uljarevic et al., 2017). As such, it was deemed appropriate to use the RBQ-2A in the sample of 16-25 year old autistic and non-autistic participants. One of the primary rationales for selecting this measure was that it is, to the researchers' knowledge, one of the only self-report outcome measure that specifically explores RRBIs currently available (the importance of which has been described within the introduction).

### **3.2.3 Analytic plan**

A 2X2 between-subjects ANOVA, with the factors of Sex/Gender (females; males) and Group (non-autistic; autistic) was used to explore group and sex/gender differences in repetitive behaviours for (1) RBQ-2A RSMB subscale scores, (2) RBQ-2A IS subscale scores (see Table 7. for summary of results). Independent t-tests were also conducted to compare autistic and non-autistic males and females, for, RBQ-2A RSMB and RBQ2-A IS subscale scores.

### **3.2.4 Community involvement**

The study was developed in line with guidelines for conducting research with autistic communities (Gowen et al., 2020) for example, additional attention was given to ensure that participants understood the relevance of the research, what it would entail, and how they could find out more about the study and the results, before they gave informed consent and began the survey (see Table 4 for more details). Participants were also provided with detailed information regarding confidentiality and their right to withdraw from the online survey at any time; however, they were made aware that their data thus far would still be collated. An expert-by-experience (autistic young adult) was also consulted, as part of the study development process, whereby they reviewed the participant information and debrief documents and the online survey to ensure that

the information was accessible to the autistic community. This provided valuable feedback prompting some changes in language, for example co-constructing a clear definition of RRBIs, drawing upon in-context examples. Additional accessibility measures were also recommended, such as the inclusion of an ‘immersive reader’ function for the online survey, which allowed participants to hear an audio description of questionnaire items on the screen.

Table 4. *Adaptations for autistic participants participating in research based on recommendations by Gowen et al. (2020).*

Adaptations	
Participant information sheet	<p>Exact details of the online survey (including an example of how the questions will be presented on screen).</p> <p>Clear language and accessible fonts used throughout.</p> <p>A brief bio of the main researcher, along with a photograph, her role at the time (Trainee Educational Psychologist) and a brief explanation about why she has chosen to focus on autism as her research area was provided.</p> <p>Videos detailing the same information as the written participant information embedded into the survey, for those who find a combination of auditory and visual information more accessible.</p>
Participant debriefing statement	<p>Information on what the research was about, using simple language, provided.</p> <p>Provided information on what will happen to the data participants have provided and researcher anticipations to publish the research and findings in an open-access journal and to enable open access for the data collected.</p> <p>Participants were informed that, following completion of the study or publication, an email could be sent to them with advice on where the research can be located. Within this email, a lay summary of findings was included for the recruitment sources to disseminate in whatever way they see fit. In order to receive this, participants were advised to provide a contact email, which was separated from data in order to maintain anonymity.</p>

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Videos detailing the same information as the written participant debriefing embedded into the survey, for those who find a combination of auditory and visual information more accessible.

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The study was approved by the University of Southampton's Ethics and Research Governance Online (ERGO) (ID: 63592) on March 25, 2021. An online consent form was included for participants to complete prior to beginning the online survey. Participants were asked to confirm that they had read/understood the participant information, they understood their right to withdraw at any time (but the data provided up to that point may still be included), they gave permission for their data to be deposited to the University of Southampton Data Repository for future research use, that they met the inclusion criteria, and that they consented to take part in the survey, by selecting 'yes' sequentially for each question. If participants selected 'No' for any question, they were taken to the end of the survey and no data was collected.

The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this study.

### 3.3 Results

#### 3.3.1 Restricted and Repetitive Behaviours and Interests

A 2X2 between subjects ANOVA was conducted for RBQ-2A RSMB scores for 2 sex/gender (male, female) and 2 group (autistic, non-autistic). This analysis showed a significant main effect of group ( $F(1,80) = 16.73$ ,  $p < .001$ ,  $\eta^2P = .173$ ), reflecting higher total RSMB scores in the autistic group compared to the non-autistic group. A second 2X2 between subjects ANOVA was conducted for RBQ-2A IS scores for 2 sex/gender (male, female) and 2 group (autistic, non-autistic). This analysis showed a significant main effect of group ( $F(1,80) = 90.71$ ,  $p < .001$ ,  $\eta^2P = .531$ ), reflecting higher total RBQ-2A IS scores in the autistic group compared to the non-autistic group.

Table 5.

Analysis of Variance for RSMB

Source	df	MS	F	p	$\eta^2P$
Sex	1	.10	.42	.520	.005
Diagnosis	1	3.99	16.73	<.001†	.173
Sex X Diagnosis	1	.29	1.21	.275	.015

Error	80	.24
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Note.—MS = Mean squares, effect size =  $\eta^2$  or partial  $\eta^2$ . \* $p < .05$ . † $p < .01$ . ‡ $p < .001$ .

Table 6.

*Analysis of Variance for IS*

Source	<i>df</i>	MS	<i>F</i>	<i>p</i>	Effect Size
Sex	1	.20	1.57	.214	.019
Diagnosis	1	11.59	90.71	<.001‡	.531
Sex X Diagnosis	1	.26	2.02	.159	.025
Error	80	.13			

Note.—MS = Mean squares, effect size =  $\eta^2$  or partial  $\eta^2$ . \* $p < .05$ . † $p < .01$ . ‡ $p < .001$ .

Independent t-tests showed a significant difference on RBQ-2A IS subdomain scores between autistic males and females, with autistic females having higher scores for IS than males,  $t(31)=-1.83$ ,  $p<.05$ . A non-significant difference between sex/gender was found for the narrow construct of RSMB,  $t(31)=-1.23$ ,  $p=.12$ . There was also a significant difference on RBQ-2A total scores between autistic males and females, with autistic females having higher total scores than males,  $t(31)=-1.87$ ,  $p<.05$ .

For the non-autistic group, independent t-tests showed a non-significant difference on RBQ-2A IS subdomain scores between non-autistic males and females,  $t(49)=.128$ ,  $p=.45$ , and RBQ-2A RSMB subdomain scores,  $t(49)=.338$ ,  $p=.37$ . There was also no significant difference on RBQ-2A total scores between non-autistic males and females,  $t(49)=.091$ ,  $p=.46$ .

Table 7. *Mean (SD) and range for age, RBQ-2A RSMB subscale score, and RBQ-2A IS subscale score for male and female young adults in the autistic and non-autistic groups.*

Autistic ( <i>n</i> =33)		Non-autistic ( <i>n</i> =51)					
Male ( <i>n</i> =15)		Female ( <i>n</i> =18)		Male ( <i>n</i> =18)		Female ( <i>n</i> =33)	
Mean (SD)	Range	Mean (SD)	Range	Mean (SD)	Range	Mean (SD)	Range

Age	17.8 (2.59)	16-24	19.22 (2.67)	16-24	18.89 (2.5)	16-25	18.94 (2.73)	16-25
RBQ-2A	1.94 (0.40)	1.16-2.5	2.14 (0.49)	1-3	1.61 (0.47)	1-2.5	1.56 (0.53)	1-3
RSMB								
RBQ-2A IS	2.17 (0.34)	1.55-2.82	2.39 (0.34)	1.91-2.91	1.51 (0.35)	1.09-2.55	1.50 (0.38)	1-2.55

*Note.* 73.81% of the sample identified as English/Welsh/Scottish/Northern Irish/British. Specific data on socioeconomic status were not recorded.

### 3.4 Discussion

The aim of this study was to investigate sex/gender differences in restricted and repetitive behaviours and interests (RRBIs), at the narrow construct level of repetitive sensory motor behaviours (RSMB) and insistence on sameness (IS), in young adults using the RBQ-2A. The findings indicate that autistic females self-report higher levels of IS and similar levels of RSMB compared to males. This is incongruent with previous literature which has routinely reported no sex/gender difference for the narrow construct of IS in autistic children/adolescents (e.g., McFayden et al., 2019; Uljarevic et al., 2021; Siracusano et al., 2021) and adults (e.g., Dell’Osso et al., 2017; English et al., 2021; Barrett et al., 2018) however, the findings are supported by some research exploring IS to the behavioural exemplar level, such as autistic girls endorsing higher levels of the item ‘distress at small changes’ on the RBS-R (Antezana et al., 2019). One explanation for these contrasting findings could be that I utilised a self-report measure that allows autistic individuals to report their own experiences of RRBIs, rather than relying on the observation and interpretation of others, such as parent/carers, teachers, and clinicians. Studies reporting non-significant sex/gender differences in IS have predominantly used child/adolescent samples and outcome measures, such as the RBS-R, that are parent/carer report (e.g., McFayden et al., 2019; Uljarevic et al., 2021; Siracusano et al., 2021).

Research that reports non-significant results using adult samples also use a range of different outcome measures such as the Adult Autism Subthreshold Spectrum (AdAS; Dell’Osso et al., 2017) and the Comprehensive Autism Trait Inventory (CATI; English et al., 2021) which are not solely measures of RRBIs and therefore have fewer items pertaining to the narrow construct of IS compared to other measures, such as the RBQ-2A. Of note, Barrett et al (2018) reported no significant sex/gender difference in IS for autistic adults using the RBQ-2A. The discrepancy



between my results and Barrett et al.'s could relate to the wider age range used in Barrett et al. (18-66 years) compared to my smaller range of 16-25 year olds. Howlin et al., (2013) commented that autistic characteristics become less severe over time and it is possible that autistic young adults report different levels of IS during early adulthood, reflecting the significant period of change that occurs for many in their early twenties. It may be that autistic females are more aware of this and are more accurate in self-reporting this than autistic males. Self-reporting of autistic experiences is important, particularly for IS, as there may be fewer observable behavioural exemplars relating to this narrow construct, particularly in relation to the distress or unease felt in response to change, as this is a somewhat internalised experience for many. This could result in IS being under-reported by others, both in research and in referral and diagnostic assessment, and may be particularly pertinent for autistic females who may display less outward preference for IS (or distress in response to change) or may use camouflaging/masking strategies to hide their differences in this domain more so than autistic males (Hull et al., 2017).

To my knowledge, this study is also one of few to include self-identifying autistic participants (also see English et al., 2021). Autism research has been criticised for recruiting exclusively clinically diagnosed participants (see reviews by Lai et al., 2015; van Wijngaarden-Cremers et al., 2014) which resulted in many samples being overly representative of autistic males (Watkins et al., 2014) because autistic females, even those with reporting high autistic traits (Russell et al., 2011), are less frequently diagnosed. As certain behaviours relating to IS are less externally observable (see above), it is possible that individuals experiencing higher levels of IS (such as autistic females, as suggested by this study) are being missed at the referral and diagnostic stage, culminating in females with higher IS levels being excluded from the previous studies reporting non-significant sex/gender differences in IS.

Our study also found no significant difference in RSMB levels between autistic males and females. This supports some of the previous literature reporting non-significant results for this narrow construct (e.g., Fulceri et al., 2016; McFayden et al., 2019; Siracusano et al., 2021) but not others, for example significantly higher endorsement of stereotyped behaviours has been reported for autistic males compared to females (e.g., Mandy et al., 2012; May et al., 2016; Uljarevic et al., 2020). Again, these conflicting findings could relate to the type of outcome measure used, for example Mandy et al. (2012) referred to behaviours identified via the developmental, dimensional and diagnostic interview (3Di; Skuse et al., 2004) and the ADOS (Lord et al., 2012) and Uljarevic et al., (2020) predicted odds ratios of stereotyped behaviours by reviewing clinician-rated severity of items relating to the RRB domain, according to the DSM-IV (American Psychiatric Association, 1994). Both of these approaches particularly rely on the observation of behaviours by others in clinical settings (e.g., ADOS), which could be biased

towards males due to male-stereotyped nosology of autism and the differing culture-oriented judgements held for how 'atypical' a stereotyped behaviour is when displayed by males compared to females (Gal, 2011). Studies reporting non-significant sex/gender differences that use parent/carer report measures, such as the RBS-R (e.g., Fulceri et al., 2016; McFayden et al., 2019; Siracusano et al., 2021), may be less vulnerable to male-bias than clinical observations, due to the wider range of opportunities autistic females have to displace RSMB in non-clinical settings over a longer period of time. The finding that autistic females self-report similar levels of RSMB to autistic males (compared with previous research that has found lower levels compared with autistic males) could potentially result from camouflaging within autism, particularly in relation to the masking of RRBIs (Hull et al., 2017; Kreiser & White, 2014), as it is suggested that, whilst observers might report fewer RSMB in autistic females, autistic females themselves are reporting equal amounts to males.

Finally, a more detailed measure of RRBIs was used in this study, compared to previous studies that have utilised total RRBI domain scores from measures such as the ADI-R (Frazier et al., 2014; Supekar & Menon, 2015; Szatmari et al., 2012). Only 12 questions relate to passionate interests and stereotyped behaviours on the ADI-R, which could hinder opportunities to capture a wide range of RRBIs compared to other measures dedicated to exploring RRBIs at a deeper level. Furthermore, some of the items on the ADI-R can be criticised for being male-biased (e.g., referring to stereotypically male passionate interests, such as trains or cars; Bargiela et al., 2016; Hiller et al., 2014; Mandy et al., 2012) which could cause autistic females' interests and behaviours to go undetected. The same could be said for studies using the RRBI domain on the ADOS, which may not be sensitive enough to capture autistic female RRBIs in observational settings, due to female camouflaging (e.g., females masking their RRBIs; Hull et al., 2017; Kreiser and White, 2014). This highlights the potential for autistic females to be missed in research and especially in diagnostic assessment where tools such as the ADOS and ADI-R are frequently used, when an in-depth exploration of RRBIs doesn't occur.

### **3.4.1 Implications for Practice**

A key implication drawn from this research is the importance of asking autistic individuals themselves about their experiences of RRBIs, particularly less observable RRBIs such as IS. It will also be important, where possible, to directly ask children and young people about their experience of RRBIs, as most childhood measures rely on parent/carer, education staff, or clinician observation and reports. This will not only be relevant to future research into sex/gender differences in RRBIs but also in terms of identification of autistic individuals via referral and subsequent diagnostic assessment. Often the first professionals to identify autistic behaviours, it

will be vital for education staff to have an awareness of autistic RRBI, particularly how these may manifest in autistic females, so that they can refer for assessment and seek appropriate, needs-based, support much earlier. This is especially important given a previous study has shown that educational staff reported that they were “unlikely” to seek support from an external professional, when presented with a vignette of the female autism phenotype (Whitlock et al., 2020). Professionals, such as Educational Psychologists (EPs), can support this awareness development by disseminating the findings from autism sex/gender research to practitioners, such as school staff, via training and promoting support (particularly for autistic girls) at both the individual and whole-school level (see Morewood et al., 2019).

In terms of diagnostic assessment, this research highlights that a proportion of autistic girls might be being missed by current diagnostic tools, due to the reliance of parent/carer report and clinician observation. In future, it will be important for diagnostic clinicians to consider RRBI that might be more internalised (e.g., distress at change) and less externally observed, especially in individuals who may be masking or camouflaging this distress in order to fit in. Likewise, the results regarding no significant sex/gender differences in RSMB also highlights the importance of asking questions about RSMB, particularly with females, as it is possible that current measures that rely on observer-reports are underestimating levels of RSMB in females. As part of this, it will be valuable to ask individuals about any camouflaging/masking behaviours they may use to hide their RRBI. It has been proposed that the diagnostic pathway for autism for children and young people could be enhanced by the involvement of EPs, who are well-placed to collect contextual evidence that could support diagnostic teams (O’Hagan & Bond, 2019) via means of observation in structured and unstructured school environments, views gathering with young people themselves using creative approaches, and discussions and consultations with school staff.

### **3.4.2 Limitation and Future Research**

A strength of this study is that participants who self-identify as autistic (and also have high autistic traits) were included in the autistic sample, which enabled the researchers to explore differences that might be being missed in research using only clinically diagnosed autistic samples. Analysis of both groups indicated that diagnosed and self-identifying autistic participants had similar levels of autistic traits and RRBI scores, evidencing that self-identification is a legitimate way to identify as neurodiverse and that including self-identifying individuals in autistic samples is valid and should be encouraged, in order to reduce the barriers to research participation experienced by this group. Previous research suggests there could also be differences (e.g., in terms of social functioning) as well as similarities (e.g., in terms of social motivation) between autistic diagnosed females and females with high levels of autistic traits without a diagnosis

(Belcher et al., 2022), though there is very little research which directly compares these individuals. Therefore, future research needs to compare females on the autism spectrum who do and do not have a diagnosis on a wider range of outcome variables, so that a more nuanced understanding of their possible similarities and differences can be developed, though the minimal differences found indicate it was appropriate to include them in one group in the present study.

A second strength is the use of a self-report outcome measure that explores a wider and more nuanced range of RRBIs in comparison to previous measures, such as the ADI-R and ADOS. The findings suggest that, by using self-report measures, research is able to establish subtle differences in autistic RRBIs that are not identifiable when relying on observer-report alone and, therefore, should be used more regularly in research exploring autistic behaviours and experiences. Future research needs to further explore the relationship between the specific way in which RRBIs are measured, including directly comparing self- and clinician-report, as well as how these relate to individuals self-reported use of camouflaging strategies.

However, as with any study, ours is not without limitations. Firstly, the sample size was relatively small, meaning replications are needed, particularly as I was unable to form a priori hypotheses. Secondly, it was not fully possible to independently verify that all participants had the required reading skills (to GCSE level) to complete the survey. This could have influenced results if some did not have the required skills, despite self-reporting, to complete the survey independently. Additionally, the requirement of a specific reading age will have contributed towards the exclusion of individuals with intellectual disabilities or more severe experiences of autism, meaning that the study was unable to capture the experiences of these groups. Previous research has indicated that IQ could serve as a moderating factor in the differences in RRBIs between males and females (Jiujiyas et al., 2017; Stratis & Lecavalier, 2013; Wood-Downie et al., 2021). Unfortunately, due to the nature of this study using an online platform, it was not possible to accurately capture the IQ of participants and subsequently use this as a covariate in analysis. Future research would benefit from including a measure of IQ in order to establish this relationship with RRBIs. Further work is also required to consider how to make research using self-report measures accessible to a wider range of autistic individuals (including those with intellectual disabilities).

Thirdly, by using the recommended two-factor component on the RBQ-2A (RSMB and IS), I was unable to separate data into the four specific narrow constructs of RRBIs referenced in the DSM-5 (e.g., stereotyped behaviour, insistence on sameness, passionate interests, and sensory experiences; American Psychiatric Association, 2013), for example the IS subscale on the RBQ-2A features items relating to IS and circumscribed interests. Separating these narrow constructs

further will be important, given studies have reported autistic males as displaying significantly higher rates of circumscribed interests than females (e.g., Antezana et al., 2019; Fetta et al., 2021; Solomon et al., 2012) which is inconsistent with the higher rates of IS (of which circumscribed interests is included) in autistic females reported in this study. Likewise, the developers of the RBQ-2A recognise that the measure may have a weakness in capturing sensory elements (Barrett et al., 2018). However, despite these challenges, the decision to remain using the RBQ-2A was made due to, to my knowledge, there being no current outcome measures available that separately explore the individual narrow constructs of RRBIs that are validated for the 16-25 years age group. As such, a focus for future research should be to develop more comprehensive measures for each of the narrow constructs so that sex/gender differences can be investigated further.

Finally, it is acknowledged that, although data was collected regarding participant gender identity, it was not possible to include this in analysis due to few participants (n=4) reporting an identity outside of the male/female binary. As a result, participant biological sex was used in analysis which does perpetuate the underrepresentation of non-binary groups in research, particularly for the autistic community. It will be important to consider intersectionality in autism research in future and researchers should to take steps to explore differences in terms of gender, rather than biological sex, in order to include individuals who do and do not identify their gender as the sex they were assigned at birth. This is especially important for autistic individuals at this intersection, for which context and societal pressure may influence how they present, both in terms of gender and autistic traits (Kourti & MacLeod, 2019) and how they access diagnosis and support (Cascio, Weiss & Racine., 2020).

### **3.5 Conclusion**

This study has shown the value of using self-report measures and including self-identifying autistic participants in autism research. Autistic females reported experiencing higher levels of IS compared to autistic males, but similar levels of RSMB. This is incongruent to some previous research, suggesting that the experiences of RRBIs for autistic females may not have been accurately captured, particularly in studies using parent/carer report or observations. This could be due to measures being developed based on a male-stereotyped nosology of autism or due to a lack of exploration of how camouflaging/masking might influence how autistic females display RRBIs. Furthermore, given the inclusion of self-identifying autistic participants, the findings could suggest that individuals experiencing higher levels of IS (such as autistic females) may be being missed at the referral and diagnostic stage, contributing towards the under-diagnosis of autism in females, though additional research, including replications, are needed to substantiate these

## Chapter 3

hypotheses. This has implications for future practice, particularly how autistic females are identified in terms of RRBIs. Educational Psychologists (EPs) can play an important role in the training of education staff to recognise the manifestation of autistic traits, such as RRBIs, in females, leading to more timely identification and referral of females for diagnostic assessment and needs-driven support. EPs can also inform diagnostic assessments by providing a range of contextual-based information drawn upon their time observing and directly working with children and young people, their families and school staff.

## Appendix A Full Search Terms

Table 8. Full Search Terms for Systematic Review

Population	Comparator	Outcomes
autis* asd asc Asperger* ‘autism spectrum disorder’ ‘autism spectrum condition’ ‘pervasive developmental disorder- not otherwise specified’ ‘PDD-NOS’	Sex Gender	‘restricted behavi?r*’ ‘repetitive behavi?r*’ ‘restricted interest*’ ‘repetitive interest*’ ‘RRBI*’ ‘repetitive movement*’ ‘stereotyped movement*’ ‘repetitive use of object*’ ‘repetitive speech’; ‘echolalia’ ‘insistence on sameness’ ‘inflexible adherence to routine*’ Ritualised Sensory ‘fixated interest*’ ‘intense interest*’ ‘Circumscribed interest*’ ‘perseverative interest*’, ‘special interest*’ ‘inflex*’

## Appendix B Newcastle-Ottawa Quality Assessment Scale with adaptations

**Total available score = 6**

**Original:**

1. Representativeness of the sample:

1. Truly representative of the average in the target population. \*\* (all subjects or random sampling)
2. Somewhat representative of the average in the target group. \* (non-random sampling)
3. Selected group of users/convenience sample.
4. No description of the derivation of the included subjects.

**Adaptations:**

- A. A sample truly representative of the target population would be an adequate mix of sex/gender, IQ, SES, ethnicity, geographical area etc = 2 star
- B. A somewhat representative sample would show some attempts to achieve representativeness = 1 star
- C. If participants were all selected from one group/location based on convenience e.g., if recruited from the same ASD clinic or geographical area = 0 star
- D. No description of how sample was recruited or sample demographics = 0 star

**Original:**

2. Sample size:

- a. Justified (including sample size calculation). \*
- b. Not justified.
- c. No information provided

**Adaptations:**

- A. Sample size calculation detailed and reported to be satisfactory = 1 star
- B. Sample size given but no justification = 0 star
- C. No comment at all on sample size = 0 star

**Original:**

3. Non-respondents:

- a. Proportion of target sample recruited attains pre-specified target or basic summary of non-respondent characteristics in sampling frame recorded. \*
- b. Unsatisfactory recruitment rate, no summary data on non-respondents.
- c. No information provided

**Adaptations:**

- A. Details given about the number of people approached and how many responded (e.g., 'non responders' OR a target response rate aim given = 1 star
- B. Unsatisfactory recruitment rate or no data on non responders = 0 star
- C. No information = 0 star

**Original:**

4. Ascertainment of the exposure (risk factor):

- a. Validated measurement tool. \*\*
- b. Non-validated measurement tool, but the tool is available or described in the article. \*
- c. No description of the measurement tool

**Adaptations:**



- A. A validated measure of Autism is used (e.g., ADOS, ADI) to confirm diagnosis or high-trait autism = 2 stars
- B. A non-validated tool is used to confirm Autism diagnosis but the tool is described or available (e.g., if using a newly-created measure) = 1 star
- C. No description of Autism confirmation measurement tool = 0 stars

**Comparability:****Total available score = 2****Original:**

1. Comparability of subjects in different outcome groups on the basis of design or analysis. Confounding factors controlled.
  1. Data/ results adjusted for relevant predictors/risk factors/confounders e.g. age, sex, time since vaccination, etc. \*\*
  2. Data/results not adjusted for all relevant confounders/risk factors/information not provided.

**Adaptations:**

- A. The researchers have looked for differences in age, IQ and controlled for this if there is a difference (e.g., by using ANCOVAs) = 2 stars
- B. No exploration in differences in age or IQ and thus no controlling for this = 0 star

**Outcome:****Total available score = 3****Original:**

1. Assessment of outcome:
  1. Independent blind assessment using objective validated laboratory methods. \*\*
  2. Unblinded assessment using objective validated laboratory methods. \*\*
  3. Used non-standard or non-validated laboratory methods with gold standard. \*
  4. No description/non-standard laboratory methods used.

**Adaptations:**

- A. The dependent variable is being measured using a validated tool = 2 stars
- B. The dependent variable is being measured using a tool that has limited information regarding validity = 1 star
- C. A non-validated measurement tool is used = 0 star
- D. No information on measurement tool given = 0 star

**Original:**

2. Statistical test:
  - a. Statistical test used to analyse the data clearly described, appropriate and measures of association presented including confidence intervals and probability level (p value). \*
  - b. Statistical test not appropriate, not described or incomplete.

**Adaptations:**

- A. An appropriate statistical test has been used (e.g., ANOVA) and the specific p value, effect size and confidence intervals are provided = 2 stars
- B. An appropriate statistical test has been used BUT they have only reported specific p value (no confidence intervals or effect size)
- C. No report of specific p value, confidence intervals and effect size.

Cross-sectional Studies:

Very Good Studies: 10-11 points

## Appendix B

Good Studies: 6-9 points

Satisfactory Studies: 4-5 points

Unsatisfactory Studies: 0 to 3 points

This scale has been adapted from the Newcastle-Ottawa Quality Assessment Scale for cohort studies to provide quality assessment of cross sectional studies.

## Appendix C Summary of studies in the systematic review and meta-analysis

**Aita et al., (2019)** used the Japanese translation version of the RBS-R with 54 hospitalised autistic adults (39 male, 15 female; mean age 36.69 years) with intellectual disabilities as part of their study exploring the association between oxytocin concentrations and ASD symptoms. Participant diagnosis of ASD was confirmed using the CARS-TV. With regards to sex/gender differences in RRBIs, they concluded no significant differences in any of the RBS-R subscales (stereotyped behaviours, restricted interests, and sameness).

**Antezana et al., (2019)** explored sex/gender differences to the item-level on the RBS-R with a sample of autistic boys ( $n=507$ ) and girls ( $n=108$ ) with a mean age of 10.26 years. Overall, they found no sex/gender differences for the RBS-R Total score ( $p=.67$ ). However, at the item-level, they did identify significant differences between males and females. They found that autistic boys had significantly higher ratings of repetitive hand/finger mannerisms compared to autistic girls ( $p<.01$ ), as well as higher rates of repetitive use of objects, including preoccupation with parts of objects, ( $p<.05$ ). In contrast, behavioural exemplars relating to insistence on sameness (e.g., 'sitting in the same place') was significantly higher in autistic females ( $p<.05$ ). There was also an observed trend towards females displaying higher rates of dislike in change and ritualistic behaviours relating to 'self-care: bathroom/dressing' (both  $p<.10$ ). Whilst these findings indicate that autistic females may present with more behaviours relating to IS compared to males, it is important to acknowledge that the exemplars used in this study are open to interpretation. 'Sitting in the same place' may indeed represent a preference for sameness, however it could also reflect a sensory preference, depending on the context. A greater prevalence of 'fascination with movement' was also observed in boys ( $p<.01$ ). A step-wise discriminant function analysis also revealed that 'fascination with one subject/activity', along with seven other items, best-predicted sex/gender difference (SCFC= -0.48). The results suggests that, whilst there was no reported sex/gender difference for total RBS-R symptom severity, when exploring at the item level, there is some variability in terms of significant differences at the behavioural exemplar level. However, limited access to IQ data meant cognitive ability could not be considered as a moderator, which might explain some of the variability in significant differences at the item level. Likewise, the fact that the RBS-R is a parent-report may make this study further susceptible to bias as responses are based on parent interpretation rather than the autistic individual themselves.

**Anthony et al., (2015)** explored the endorsement and intensity of restricted interests in 109 autistic children and young adults using the Interests Scale (Bodfish., 2004), compared to an age

and IQ-matched sample of non-autistic participants. They found that autistic females ( $n=16$ ) mostly reported or displayed interests in numbers and maths (18%) and music (12.5%) meanwhile autistic boys ( $n=93$ ) reported or displayed interests in video games (21.5%), Lego (7.5%) and playing games along (4.3%). Whilst the exploration of the content of restricted interests in this study is unique and useful, the small sample size (particularly for females) is notably small ( $n=16$ ).

**Aykan et al., (2020)** used the Sensory Sensitivity Scales (SeSS) with a small sample of adults (36 male, 39 female; mean age 23 years) who, although not formally diagnosed autistic, scored high for autistic traits on the Autism Quotient, as part of their study investigating sensory processing differences (auditory and visual) in autistic traits. They found that visual sensitivity was correlated with autistic traits in females ( $p<.05$ ) whereas auditory sensitivity was correlated with autistic traits for males ( $p=.05$ ). They concluded that auditory processing differences were particularly related to autistic traits in non-autistic male individuals.

**Barrett et al., (2018)** asked 271 adults (100 male, 171 female, mean age 36.56 years) who self-reported a diagnosis of either Asperger's Syndrome, ASD, or high functioning autism or 'other' (including atypical autism; autistic disorder; childhood autism; high-functioning autism/Asperger's syndrome; pathological demand avoidance; Pervasive Developmental Disorder–Not Otherwise Specified), to complete the self-report RBQ-2A questionnaire and compared rates of repetitive sensory motor behaviours (RSMB) and insistence on sameness (IS) between sex/genders. They concluded no significant sex/gender difference in terms of RSMB ( $p=.64$ ) or IS ( $p=.72$ ). However, RSMB was significantly negatively correlated with age ( $p<.001$ ) suggesting that RSMBs in both sexes/genders decreased with age.

**Bitsika et al., (2018)** investigated sex-based differences in sensory features in a sample of age and IQ-matched autistic children and adolescents. They reported non-significant sex/gender differences between age and IQ-matched autistic males ( $n=51$ ) and females ( $n=51$ ) (mean age; 10.2 years) on all subscales of the Sensory Profile including; low registration, sensation seeking, sensory sensitivity, sensory avoiding, auditory processing, visual processing, vestibular processing, touch processing, oral sensory processing, and multisensory processing.

**Boyd (2020)** Drawing from the ADOS-2 data records of 7,477 racially-diverse autistic children, Boyd (2020) completed a differential item functioning (DIF) exploration to see if any particular items on the ADOS-2 were sensitive to demographic characteristics, including gender. There was no significant difference between boys and girls for any items on the restrictive, repetitive behaviour subscale in ADOS-2 modules one or two, which are used for children who are not fluent in verbal language. However, a potential DIF was identified for item A4 (stereotyped/idiosyncratic use of words or phrases) in ADOS-2 module 3 ( $p<.05$ ) with autistic boys

having lower scores on this item compared to autistic females. This suggests that verbally fluent autistic girls display more stereotyped and/or idiosyncratic use of words or phrases than autistic boys. However, the effect size was small (DIF contrast = .34) and less than the recommended meaningful effect size of DIF contrast  $>.64$  (Boone et al., 2014) which reduces how much can be concluded from this finding.

**Brierly et al., (2020)** used the RBS-R in their study exploring the measure's factor structure in a large sample of children and young adults diagnosed with ASD or ADHD ( $n= 1182$ ; mean age 10.2 years). In addition, they investigated the relationship between subdomains on the RBS-R and sex and cognitive ability, and whether these differed between ASD and ADHD. They found no significant difference in scores on the stereotypy and ritualistic/sameness subscales of the RBS-R between autistic males and females ( $n=643$ ), indicating no clinically meaningful difference in RRBI variation. The wide age range of participants in this sample (ranging from 1.5 years to 21.9 years) is a limitation and unfortunately the study was not powered enough to run factor analysis for stratified age ranges.

**Caldwell-Harris and Jordan (2014)** used a modified version of the Cambridge University Obsessions Questionnaire (including categories of nature, history, and culture) with 69 self-reported autistic participants (mean age 29.2 years) to rate the level of intensity they experience for a range of circumscribed interests. They concluded that autistic males ( $n=39$ ) had stronger interests in machines and technology, with vehicles being of the highest intensity. Autistic males also reported having stronger interests (albeit not to a level of significance) in 'collecting' compared to autistic females ( $n=30$ ), however the interest in collecting items such as rocks were higher for autistic females than autistic males.

**Dell-Osso et al., (2017)** developed the Adult Autism Subthreshold Spectrum (AdSS) questionnaire and explored sex/gender differences for the domains of sensory, IS (inflexible adherence to routine), and restricted interests (restricted interest and rumination) in a sample of 102 adults (66 male; 36 female; mean age 24.29 years) who either had a diagnosis of ASD or who endorsed at least one criterion symptom for ASD based on the DSM-5 diagnostic criteria. They found a significant difference for hyper-hypo reactivity to sensory input, with females reporting higher levels ( $p<.05$ ). No significant difference between males and females was reported for 'inflexibility and adherence to routine ( $p=.20$ ). Finally, no significant sex/gender difference was reported for restricted interests and rumination ( $p=.06$ ).

**English et al., (2021)** explored self-report of sensory sensitivity, restricted/repetitive behaviour (classified in this review as stereotyped behaviour), and cognitive rigidity (classified as insistence on sameness in this review) using the Comprehensive Autistic Trait Inventory in a

## Appendix C

sample of 1079 (557 male, 522 female) diagnosed and self-identifying autistic adults (mean age; 37.41) and found that males reported significantly more restricted/repetitive behaviours than females ( $d=.23$ ) whilst females reported significantly more sensory sensitivity ( $d=.24$ ). There was no significant sex/gender difference for cognitive rigidity ( $d=.01$ ).

**Evans (2018)** analysed parent/carer reports of stereotyped behaviours, insistence on sameness, and restricted interests using the RBS-R for 175 infants (143 male, 32 female), of an average age of 42 months, and concluded that girls demonstrated higher RBS-R overall total scores than boys (mean; 41.65 and 34.96 respectively). At the subscale level, there were differences between sex/genders with girls demonstrating more stereotyped behaviours than boys (mean; 4.68 and 4.18 respectively) and more sameness behaviours than boys (mean; 9.97 and 8.32 respectively) meanwhile boys demonstrated more restricted interests than girls (mean; 5.01 and 4.71 respectively).

**Feldman et al., (2020)** used the Sensory Profile to examine differences in sensory responsiveness between autistic children ( $n=50$ ) and age and gender-matched non-autistic children ( $n=50$ ). The autistic sample consisted of 37 males and 13 females, with an average age of 13 years. All participants in the group had a diagnosis of Autism, which was confirmed using the ADOS-2 and clinical judgement by the research team. The study did not report of sex/gender differences, however the research provided data to be used in the current meta-analysis.

**Fetta et al., (2021)** used the RBS-R to explore sex/gender differences in autistic children (39 male, 11 female) with an age range of three to 15 years. On the RBS-R subscales, no significant sex/gender differences were identified for any of the subscales, apart from restricted interests where autistic males displays higher levels of this behaviour ( $p<.05$ ) compared to females. The researchers also examined parent/carer-reports on the Short Sensory Profile and found that autistic males displayed higher levels of movement sensitivity ( $p<.01$ ) and under-responsive/seeks sensation ( $p<.05$ ) than females. All other subscales on the Short Sensory Profile (e.g., tactile sensitivity, taste/smell sensitivity, auditory filtering, low energy/weak, visual/auditory sensitivity) were non-significant.

**Fulceri et al., (2016)** explored the accuracy of the RBS-R as well as the relationship between restricted and repetitive behaviours and sex, age, non-verbal IQ, autism severity. They reported no significant sex/gender differences on any of the subscales of the RBS-R (Italian translation) apart from the Stereotypies subscale, where parents/carers of autistic boys ( $n=64$ ; mean age of 51.8 months) endorsed significantly more stereotyped behaviours than parents/carers of autistic females ( $n=15$ ) ( $p=.033$ ).

**Grove et al., (2018)** asked participants ( $n= 407$ , mean age 42.4 years) to list their special interests and how much time they spend engaging in them each day/week. Participants also completed the Special Interests Motivation Scale and rated how much their special interest impacts their lives positively and on their daily function. Overall, autistic males ( $n=222$ ) were significantly more likely to endorse having a specialist interest than autistic females ( $n=185$ ) ( $p<.01$ ). There was no sex/gender difference in the frequency or intensity of special interests or in the positive or negative impact of special interests between sexes/genders. In terms of content, female special interests were significantly more likely to include autism, nature/gardening, the human body/psychology, animals, arts and crafts (all  $p<.05$ ) whereas males were more likely to endorse special interests in computer/gaming, music, science, politics/history, countries/language/travel, maps/calendars/dates, maths/numbers, construction toys, vehicles, collecting, anime, and timetables/schedules ( $p<.05$ ), which could be argued are rather stereotypically associated with autism.

**Harrop et al., (2015)** used videotaped interactions between 29 autistic boys (mean age; 35.83 months) and 29 autistic girls (mean age; 38.81 months) and their caregivers to explore a range of restricted and repetitive behaviours coded based on a scheme developed by Harrop et al (2014). They concluded that there was no significant difference in the frequency of verbal behaviours described as ‘echolalia, scripted language, atypical vocalisations and atypical rhythm/intonation’ between autistic boys and girls. Albeit not to a level of significance, they also coded more repetitive use of objects by autistic boys than girls ( $p=.21$ ), suggesting a trend towards higher prevalence of this type of stereotyped behaviour in autistic boys, however the reported effect size was small ( $\eta^2 = .02$ ) and could instead reflect the study lacking sufficient power, due to small sample size. The coded ‘sensory aversion’ category, referring to a sensitivity to sounds and touch, yielded a non-significant difference between girls and boys. However, this is a broad category and the behavioural examples used in the coding manual (e.g., covering ears in response to sounds or displaying a clear reaction to caregivers rolling a toy car on the child’s leg) may be vulnerable to subjective interpretations or may lack evidence of a specific sensory response. The researchers also found that boys demonstrated a trend towards higher frequencies of ‘sensory-visual’ behaviours ( $p=.05$ ) and more ‘sensory seeking’ behaviours ( $p=.19$ ) compared to girls.

**Harrop (2018)** used eye-tracking technology to quantify the attention male ( $n= 25$ , mean age 113.12 months) and female ( $n=26$ , mean age 102 months) autistic infants paid towards different images during a visual exploration task. Images were categorised as ‘male’ (e.g., building toys, games consoles, ‘star wars’ characters, and Lego), ‘female’ (e.g., dress up toys, make up, dolls, and tea sets), or ‘neutral’ (e.g., ‘mickey mouse’) based on previous literature reviewed by

## Appendix C

the researchers and confirmed for representativeness via a small online survey with parents of non-autistic children. The number of images viewed (exploration), the length of time each image was explored (exploration), and the amount of detail each image was inspected in (detail orientation) were all explored. Results concluded that autistic girls spent longer exploring 'female' images compared to autistic boys ( $p < .05$ ) and autistic boys perseverated more to 'male' images compared to autistic females ( $p < .05$ ). Furthermore, autistic boys were significantly more detail oriented than autistic females ( $p = .05$ ). The researchers concluded that these findings suggest that different toys/interests capture the attention of autistic boys and girls, in line with the gender differences observed in typical development.

**Hattier et al., (2011)** used the Diagnostic Assessment for the Severely Handicapped- Second Edition (DASH-2; Matson, 1998) to explore the prevalence of 'stereotypic disorder' (defined as "motor behaviour that is repetitive, often seemingly driven, and non-functional"; American Psychiatric Association, 1994) in 140 autistic adults (77 male; 63 female; mean age 49.28 years), who also had profound intellectual disability (ID), and found that autistic women had lower scores on the 'Stereotypies' subscale compared to autistic men ( $p < .05$ ). Whilst this suggests that repetitive motor behaviours are more prevalent in males, it is difficult to confidently conclude whether this reflects a sex/gender difference within the autistic population or that of the ID population, given 'lower order' behaviours such as repetitive motor movements commonly occur in individuals with low IQ (Turner, 1999), regardless of autism diagnosis.

**Hiller, Young, and Weber (2014)** In a sample of 138 autistic children (69 male; 69 female; mean age 8.76 and 8.06 respectively) with a diagnosis of 'high functioning ASD' (the DSM-4-TR criterion used to make official diagnosis), explored reports of restricted interests in children's diagnostic assessment reports and concluded a significant sex difference in restricted interests ( $p < .001$ ). Being reported to have 'seemingly random' special interests (e.g., interest in rocks, pens, stickers) was significantly predictive of being a girl, with 60% of girls endorsing this category compared to only 29% of boys. In contrast, a special interest focussing on 'screen time' (e.g., obsessive gaming, iPad, other screen technology) was more predictive of being a boy, with 38% of boys reporting this obsessional interest compared to only 9% of girls. A sex/gender difference was also identified for special interests focussing on a certain TV programme or character, with 17% of girls endorsing this interest compared to 10% of boys. This study also explored the potential influence of age on sex/gender differences in the type of restricted interest by categorising participants into groups of <7 years old and >7 years old. This analysis concluded that an interest in wheeled toys was prevalent in a larger percentage of younger boys meanwhile older boys had a greater fixation with screens. In contrast, autistic girls remained mostly endorsing the 'seemingly random' across both age groups. Hiller et al (2014) also reported that, based on the DSM-5



criteria, autistic boys were more likely to present with stereotyped use of objects (e.g., lining up or sorting) compared to autistic girls.

**Hiller et al. (2016)** explored the predictive odds ratios for pre-diagnosis concerns of parents/carers relating to restricted and repetitive behaviours and subsequent autism diagnosis. In a sample of 152 children (mean age 10.94 years), with 92 boys and 60 girls, a backward stepwise logistic regression identified that 'interest in parts of mechanical objects' significantly predicted sex alongside four other items (not RRBI related) ( $p < .01$ ). When controlling for age, this item remained a strong predictor of sex (alongside imitation ability). Most girls in the sample were rated as having 'little or no interest' in parts of mechanical objects (48% of girls compared to 15% boys;  $p < .01$ ). In contrast, 55% of boys were rated as 'fascinated' compared to only 26% of girls ( $p = .096$ ).

**Hus et al., (2007)** explored parent/carer reports of insistence on sameness displayed by their children, based on responses to items relating to these behaviours on the ADI-R. The sample consisted of parents/carers of 812 autistic boys and 171 autistic girls (mean age 7.75 years). The results indicated no significant difference between autistic boys and girls for insistence on sameness at any severity level (e.g., low, medium, high).

**Knutsen et al (2019)** explored the restricted and repetitive behaviour subscale on the ADOS-2, including behavioural exemplars such as stereotyped/idiosyncratic use of words or phrases, with a sample of 1024 IQ-matched autistic boys ( $n=512$ ) and girls ( $n=512$ ; aged 2-12 years) and concluded no significant sex/gender differences between boys and girls, irrespective of age or IQ level. They also reported no significant difference in reports of behavioural exemplars, such as hand and finger mannerisms, for either  $<6$  years or 6-12 year groups or  $IQ < 70$  or  $IQ > 70$  groups, suggesting that no sex/gender difference in this specific repetitive behaviour, regardless of age or IQ. However, they did report that younger high-functioning autistic girls and older lower-functioning autistic girls reported less repetitive interests than boys ( $p < .05$ ) however, this category also encompasses 'stereotyped behaviours' which makes it difficult to extrapolate findings specifically for repetitive interests.

**Lam (2004)** focussed their dissertation on investigating the validity of the RBS-R as well as exploring sex/gender differences in the subscales of the RBS-R using a sample of 305 children and adolescents (253 male, 53 female; mean age of 15.34 years) and concluded no significant sex/gender difference for any of the subscales of relevance to this review.

**Lane et al., (2014)** explored sex differences in autistic hyposensitivity using subscales of the Short Sensory Profile with 228 autistic infants (mean age; 60.71 months) and concluded no

difference between boys ( $n=203$ ) and girls ( $n=25$ ) on any of the sensory subscales, including hyposensitivity. A critique of this particular study is the noticeable dominance of males in the sample.

**Lawrence (2017)** examined the relationship between self-injurious behaviour and a range of factors, including stereotypies and sensory experiences, by analysing client records of 145 children and young adults (114 male, 31 female; mean age 16 years) who were diagnosed autistic and who also had comorbid intellectual disabilities. Analysing mean scores from the Autism Spectrum Rating System (ASRS), Lawrence reported that autistic males displayed more stereotypies (mean: 17.11) compared to autistic females (mean: 12.52). Lawrence also reported that autistic females had higher mean sensory processing difficulties (mean: 72.18) compared to autistic males (mean; 71.89).

**Lee (2009)** investigated sex/gender differences in relation to areas of development, cognition, behaviour, and sensory experiences, in a sample of 116 males and 33 females (aged 12-18 years) who self-reported a diagnosis of Aspergers' syndrome, confirmed using parental completion of the Asperger Syndrome Diagnostic Scale (ASDS; Myles, Bock, & Simpson, 2001). Lee reported significantly lower scores for sensory sensitivity on the Adolescent/Adult Sensory Profile compared to females ( $p<.01$ ).

**Lever et al., (2018)** conducted a cross-sectional study investigating self- and other-reported autistic traits in both autistic and non-autistic adults in order to explore changes across the lifespan. They concluded that autistic women ( $n= 56$ ) reported higher total scores on the Sensory Sensitivity Questionnaire compared to autistic men ( $n=116$ ), indicating greater sensory sensitivity ( $p<.001$ ).

**Mandy et al., (2012)** investigated sex differences in repetitive stereotyped behaviours, as reported by parents on the 3Di and observed in the ADOS, for 325 children aged three to eighteen years old (273 male, 52 female). They concluded that autistic boys had more repetitive stereotyped behaviours than girls on the 3Di ( $p=.03$ ) with boys more frequently displaying 'oddly formal play' that involves systematically lining up toys. On the ADOS, repetitive stereotyped behaviour scores differed between boys and girls, with boys showing greater impairment than girls ( $p=.04$ ). Using the 'activities and interest' subscale of the Children's Communication Checklist (Bishop, 1998), Mandy et al (2012) also concluded no significant difference between autistic males and females for the item 'endlessly and exactly repeating words/phrases'.

**May et al., (2016)** investigated sex/gender differences in repetitive motor movements in 64 (32 male, 32 female) children, aged 7-12 years, who were diagnosed as autistic or having

Asperger's syndrome, using the mean scores from the Repetitive Behaviours Questionnaire (RBQ) and found that autistic males displayed more repetitive motor movements than females ( $p=.05$ ). However, this was also the case for the non-autistic group featured in the study, which suggests that this gender difference is not unique to autistic children. No significant sex/gender difference was found for 'rigidity and adherence to routine'.

**McFayden et al., (2019)** reported that, in a sample of 72 autistic young people (mean age 12.33 years), males ( $n=55$ ) scored higher on the 'restricted behaviour' subscale of the RBS-R than females ( $n=20$ ) to a level of significance ( $p<.05$ ). However, the restricted interest subscale on the RBS-R includes items relating to restricted behaviours, for example 'preoccupation with part of an object', which do not align with the restricted interests narrow construct, as defined in this review. Although items pertaining to relevant restricted interests do feature within the subscale, it is not possible to separate these for analyses of explicitly restricted interests. Alongside the use of the RBS-R, McFayden et al (2019) also coded the type of restricted interests reported by autistic males and females from ADI-R and/or ADOS reports. Ten restricted interest categories emerged which were then assigned a number and then diagnostic reports were re-coded and then co-coded by two researchers blind to participant sex. Multiple interests in one diagnostic report were coded individually and interests spanning categories were dually coded. The results indicated that males and females displayed the same average number of interests. Equally, a similar proportion of males and females were reported to have no restricted interests (34% and 31% respectively). Similar rates of interest in electronics (video games, TV, iPads) and reading were reported across sexes/genders. However, males were more likely to report interests in vehicles (13%) and history (10%) compared to females (0% and 4% respectively) whereas females were more likely to report interests in animals (27%), people (7%) and science (8%) relative to males (9%, 2%, 3%, respectively). McFayden et al. (2019) also concluded that the interests of autistic females had more of a social quality to them and were more often related to living constructs (e.g., animals and people) compared to males whose interests were often object-related constructs.

**Sipes (2011)** investigated autism symptom endorsement using parent/carer responses on the Baby and Infant Screen for Children with aUtism Traits-Part 1 (BISCUIT-Part 1; Matson, Boisjoli & Wilkins, 2007) assessment tool. Data regarding 390 infants (294 male, 96 female; mean age 29 months) were categorised into low and average development quotient (DQ) scores using the Battelle Developmental Inventory, 2nd Edition (BDI-2; Neborg, 2005) and an interesting difference was reported between the parental endorsement of echolalia (defined as 'saying words or phrases repetitively') between low and average DQ autistic children. Whilst 21.4% of parents to low DQ autistic girls reported echolalia compared to 12.2% of parents to low DQ boys, only 7.7%

## Appendix C

of parents to average DQ autistic girls reported echolalia compared to 20.5% of parents to average DQ boys. This suggests that girls with average DQ are particularly less likely to present with echolalia. However, it is possible that the types of echolalia displayed by average DQ girls are less obvious and detectable rather than less present entirely. There was also little difference in the endorsement of repetitive hand or arm movements between boys and girls with low DQ (33.1% and 30% respectively). With regards to whole body movements, low DQ boys were more likely to endorse this behaviour (32.6%) followed by low DQ girls (30%) and boys with average DQ (22.9%). Considerably less girls with average DQ displayed whole body repetitive movements (3.8%). It is possible that the reduced presentation of repetitive motor movements by average DQ girls could be due to greater social awareness and motivation to 'blend in' resulting in autistic girls with average or higher-than-average DQ engaging in masking behaviours to suppress visible repetitive motor movements. The percentage of boys displaying 'abnormal preoccupation with the parts of an object or objects' was similar for those with low DQ (36.2%) and average DQ (31.5%) however noticeably fewer girls with average DQ endorsed this behaviour (7.6%) compared to girls with low DQ (40%) (Sipes et al., 2011). This suggests that cognitive functioning may play a particular role in the presence of repetitive use of objects, especially for autistic girls. The percentage of boys displaying 'Sticking to odd routines or rituals that don't have a purpose or make a difference' was somewhat similar for those with low DQ (23.5%) and average DQ (34.3%) however noticeably fewer girls with average DQ endorsed this behaviour (15.3%) compared to girls with low DQ (25.7%), suggesting that DQ may moderate the display of this behaviour, particularly in females[HW21]. However, Sipes et al also reported no significant difference in parental endorsement for items 'upset with change in routine' and 'needs reassurance if events don't go as planned', therefore the overall findings relating to IS in this study remain mixed. At the behaviour exemplar level, Sipes et al (2011) reported significantly less parent endorsement for 'limited number of interests' (7.6%), 'interest in a highly restricted set of activities' (0%) and 'restricted interests and activities' (19.2%) for autistic girls with average DQ compared to all other groups, supporting the above findings that age and IQ seem to moderate levels of restricted interest, at least for girls, where certain interests may be less likely to be identified by observers as being limited, restricted or atypical for their gender. Finally, parent endorsement of the item 'abnormal fascination with the movement of spinning objects' was lower for those of autistic girls with average DQ (3.8%) compared to other groups.

**Siracusano et al., (2021)** recruited a sample of parents/carers of 219 pre-school and school-aged autistic children (125 male, 65 female) with an average age of 9.1 years, to complete the Italian version of the RBS-R to explore sex/gender differences. They reported non-significant

results for all RBS-R subscales of relevance for this review, including stereotyped behaviours, insistence on sameness, and restricted interests.

**Smerbeck (2019)** developed the Survey of Favorite Interests and Activities (SOFIA) and used it to examine restricted and repetitive behaviours and interests in a sample of 121 parents of high-functioning autistic boys and 49 parents of high-functioning autistic girls. They found that autistic females scored higher on the Atypicality subscale of the SOFIA, which refers to interests that are deemed unusual in nature, compared to males.

**Solomon et al., (2012)** used the RBS-R with a sample of 40 autistic children (20 male, 20 female), aged between 12-18 years, and 36 non-autistic children, to compare autism and internalising symptoms. They found no significant sex/gender differences for any of the subscales, including stereotyped behaviours, insistence on sameness, and restricted interests. However, they commented that the results were suggestive of higher scores on the restricted interests subscale in boys ( $U=77.50$ ,  $z=-2.43$ ,  $p=.015$ ).

**Stephenson et al., (2021)** used the Autism Spectrum Rating Scale (ASRS) to investigate sex/gender differences in stereotypies with a sample of 381 autistic boys and 100 autistic girls (mean age 3.41 years) and reported significant results, with autistic boys displaying more stereotypies than autistic females ( $p<.01$ ). However, they reported non-significant results when exploring 'atypical language' and 'behavioural rigidity' between autistic males and females.

**Sutherland et al., (2017)** recruited a sample of 334 parents of autistic children (aged 5-18 years) and explored the topics of special interests and the predictive odds of being male or female. Results indicated that boys were significantly more likely to be interested in technology ( $p<.01$ ), dinosaurs ( $p<.05$ ) and transport ( $p<.001$ ) whilst girls demonstrated greater interest in art ( $p<.01$ ), books ( $p<.05$ ) and singing/music ( $p<.001$ ). The researchers also developed a multiple-choice questionnaire to explore the sensory sensitivities and seeking behaviours across the senses (vision, hearing, smell, taste, touch, proprioception, and vestibular) and found that sensory sensitivities were more often reported by parents than sensory seeking behaviours (71% compared to 29% respectively). However this could reflect the reliance on parental interpretation of sensory sensitivities, and behavioural responses (such as covering ears) might be easier to observe than sensory seeking behaviours such as seeking out textures or sounds. A sex/gender difference was reported in only one element of sensory sensitivity (taste) with girls displaying more sensitivity than boys, with an odds ratio of 0.263 ( $p<.01$ ).

**Tang et al., (2021)** reviewed and interpreted autism diagnostic reports of 195 autistic children (145 male and 50 female) and categorised their parent-reported behaviours and severity

into the two categories of the DSM-5 criterion (A and B). They concluded that significantly more boys (72%) were reported to have displayed 'repeated or learned phrases' compared to only 54% of girls ( $p < .05$ ). There was no significant sex/gender difference in the presence of echolalia. No significant difference between males and females was also reported for motor stereotypies, with 82% of males and 72% of females reportedly presenting with this behaviour. Tang et al (2021) reported that 70% of autistic boys displayed repetitive use of objects compared to 68% of autistic girls. They also reported non-significant sex/gender differences in autistic children in relation to behavioural exemplars 'distress at small changes', 'difficulties with transition' and 'ritualistic behaviours'. Regarding restricted interests, the most frequently reported special interest for autistic females was animals (31%) whilst males most reported interests in transport (35%). Finally, when exploring sensory experiences, the researchers reported non-significant results for a range of sensory manifestations (e.g., auditory, movement, olfactory, and tactile). No significant difference between males and females for 'fascination with spinning objects, lights and/or mirrors' was also reported. However, autistic males were significantly less likely to present with 'fears reflecting sensory avoidance' compared to females ( $p < .05$ ). That being said, this is open to clinician interpretation of 'fears' and assessments, such as the ADOS and ADI-R, are reliant on subjective reports and the restricted opportunities clinical assessment environments provide to observe sensory avoidance.

**Uljarevic et al., (2020)** examined clinician-rated severity scores of each of the DSM-4 items relating to the restricted and repetitive behaviours domain, including repetitive motor behaviours and insistence on sameness, with a large sample of 3647 children (3007 male, 640 female), with a mean age of 6.6 years. Using odds ratios, they found that autistic males had higher ratings of repetitive motor behaviours compared to females (OR 1.81, 96% CI 1.54, 2.12) but that there was no sex/gender differences for insistence on sameness (OR 1.01; 95% CI 0.86, 1.18).

**Uljarevic et al., (2021)** conducted a very comprehensive study, with a large sample size (17,581 children and adolescents with ASD), in order to characterise how restricted and repetitive behaviour domains differ according to a range of individual characteristics, including sex/gender. Using the RBS-R, they concluded no significant sex difference on the insistence on sameness/ritualistic subscale of the RBS-R in their sample of autistic children and young adults (14,186 male, 3,395 female), average age 8.24 years. However male sex was a predictor of stereotypy ( $p < .001$ ) and restricted interests ( $p < .001$ ).

**Wanzek (2014)** used the RBS-R to explore sex/gender differences in stereotyped behaviours (and associated behavioural exemplars) in 42 autistic participants (21 male, 21 female) using a mixture of both parent/carer report and self-report. They concluded no significant

sex/gender differences in stereotyped behaviours when reported by parents/carers, but they did report patterns of difference between sex/genders when reviewing self-report data. Autistic males (mean age; 18 years, 5 months) appeared to show a pattern towards being 'somewhat more likely' to report moderate to severe levels of repetitive behaviours involving objects (e.g., spinning objects, letting objects fall out of hands) compared to females. Meanwhile autistic females (mean age; 19 years, 6 months) were 'somewhat more likely' to report mild to moderate problems relating to repetitive head movements compared to autistic males. Non-significant results for parental reports of their child's insistence on sameness on the RBS-R was reported, however Wanzek did report a trend towards autistic females reporting higher levels of sameness behaviours (e.g., preferring things to remain in the same place, objecting to visiting new places, becoming upset if interrupted in what they are doing, liking the same music or videos played repeatedly, or insisting on using a particular door) compared to males.

**Weiland et al., (2020)** aimed to explore sensory symptoms using perceptual measures, using the Sensory Perception Quotient, with a sample of autistic ( $n=657$ ) and non-autistic adults. As part of this, they collected data for males and females and reported that autistic women ( $n=340$ ; mean age; 43.2 years) self-reported higher sensory sensitivities, based on the total Sensory Perception Quotient- Short measure, compared to 316 autistic males.

**Williams et al., (2019)** explored the limits to which autistic and non-autistic participants could detect warm and cool temperatures, using a thermal perceptual thresholds test. As part of their study, they also collected outcome measures related to sensory experiences, including the 'sensory' subscale of the Social Responsiveness Scale-2, which was included in this meta-analysis. Their sample consisted of 62 autistic males (mean age 16.77 years) and 21 autistic females (mean age 18.53 years). Data used in the meta-analysis revealed no significant sex/gender difference for the 'sensory' subscale on the SRS-2.





## Appendix D Funnel plots

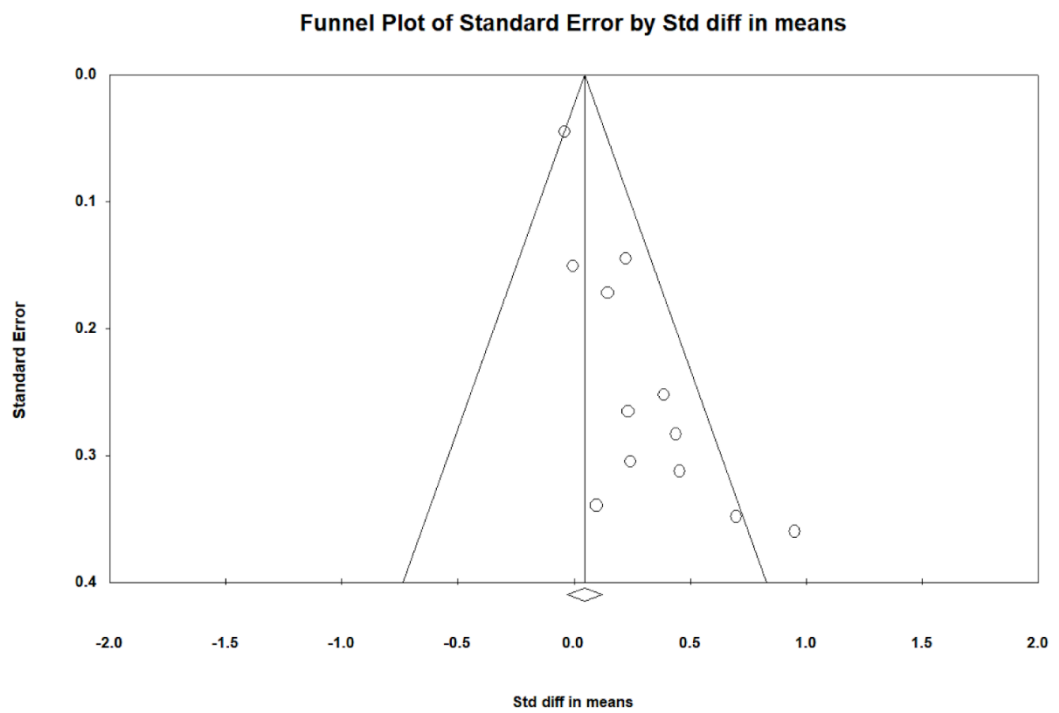


Figure 7.

Funnel plot for restricted interests meta-analysis

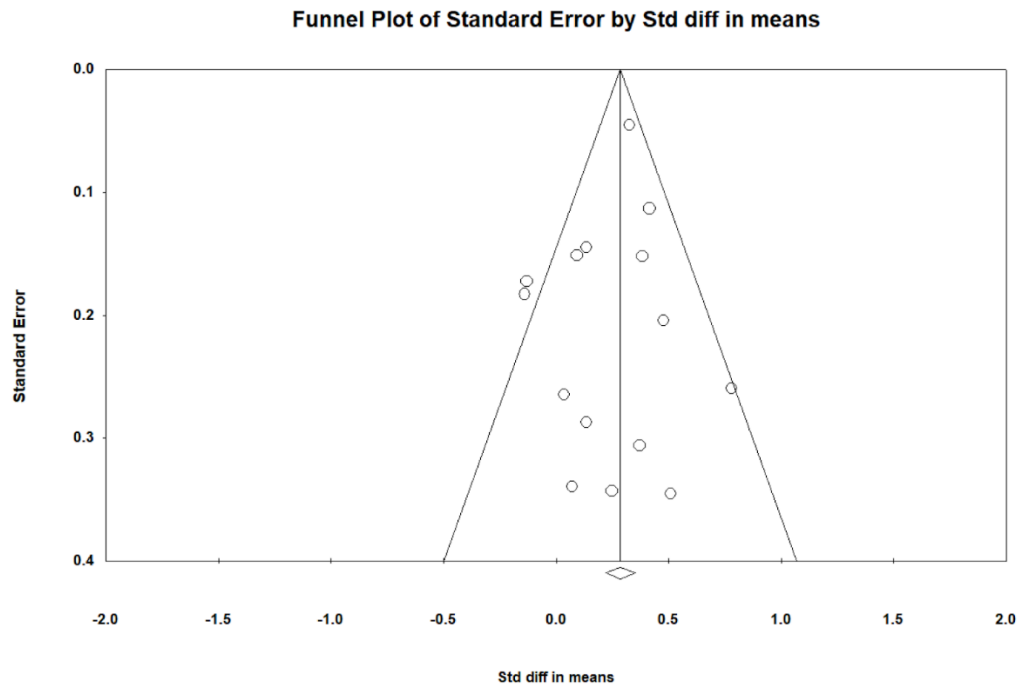


Figure 8  
Funnel plot for stereotyped behaviour meta-analysis

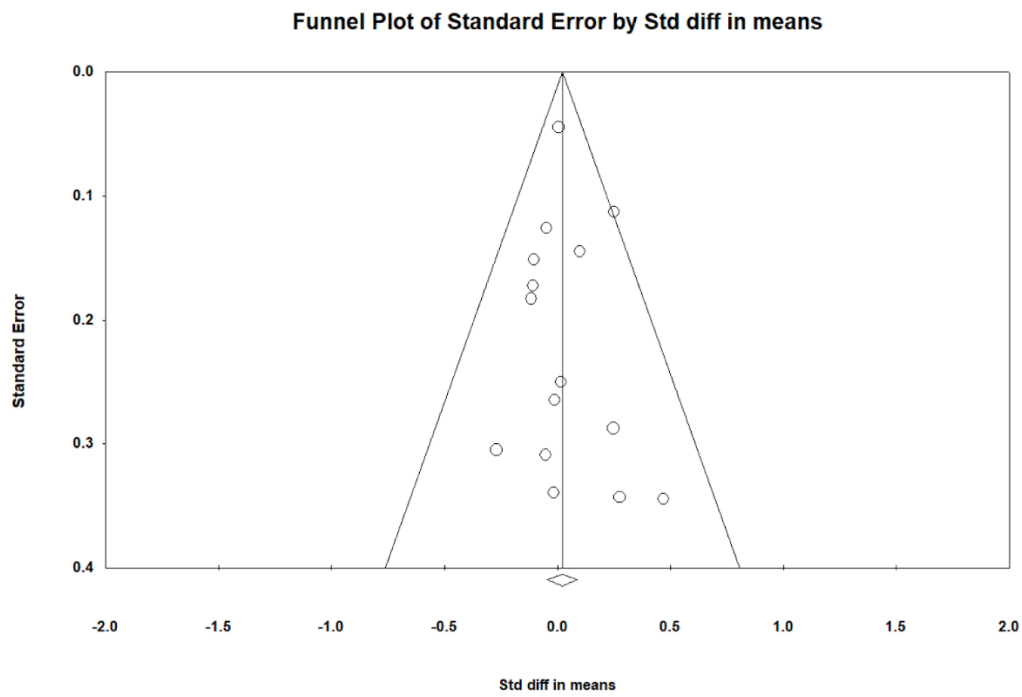


Figure 9  
Funnel plot for insistence on sameness meta-analysis

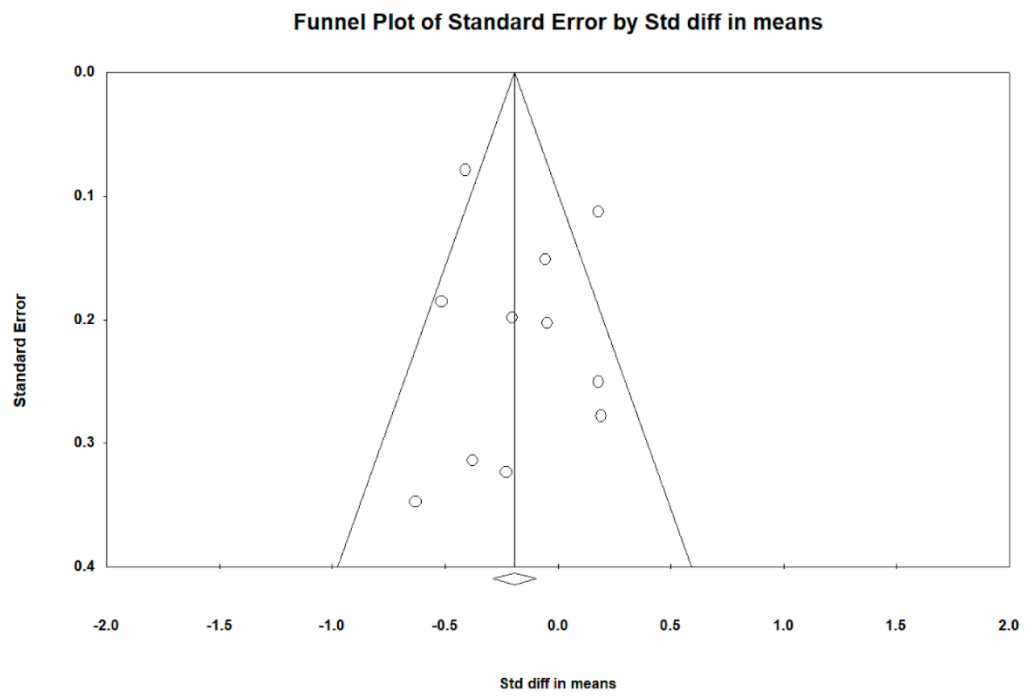


Figure 10

Funnel plot for sensory meta-analysis



## Appendix E Questionnaires

### E.1 The Autism Symptom Self-Report for adolescents and adults (Posserud et al., 2013)

1. Do you find it difficult to socialise with, or to get in touch with people, especially people your own age?
2. Do you prefer to be alone rather than being together with other people?
3. Do you have difficulties perceiving social cues?
4. Do other people tell you that your behaviour or your emotional responses are inappropriate or hurtful?
5. Do you have a strong interest or hobby that absorbs so much of your time that it hampers other activities?
6. Do you or do other people feel that you have very set routines or that you are very immersed in your own interests?
7. Do you or do other people feel that you impose your routines or interests on others?

Response options were “not true” (score 0), “somewhat true” (score 1), “certainly true” (score 2).

Posserud, M. B., Breivik, K., Gillberg, C., & Lundervold, A. J. (2013). ASSERT - The Autism Symptom Self-Report for adolescents and adults: Bifactor analysis and validation in a large adolescent population. *Research in Developmental Disabilities, 34*(12), 4495–4503.  
<https://doi.org/10.1016/j.ridd.2013.09.032>

### E.2 The Adult Repetitive Behaviour Questionnaire-2A (RBQ-2A)

Individuals often repeat the same behaviour over and over again and some individuals are more repetitive than others. Everybody also has daily routines and rituals. Please rate the repetitive behaviours you have shown over the last month and rate the most usual way that you display the behaviour. Please tick one response for each question.

1. Do you like to arrange items in rows or patterns?

- Never or rarely
- One or more times daily
- 15 or more times daily
- 30 or more times daily

2. Do you repetitively fiddle with items? (e.g. spin, twiddle, bang, tap, twist, or flick anything repeatedly?)

- Never or rarely
- One or more times daily
- 15 or more times daily

## Appendix E

30 or more times daily

3. Do you spin yourself around and around?

- Never or rarely
- One or more times daily
- 15 or more times daily
- 30 or more times daily

4. Do you rock backwards and forwards, or side to side, either when sitting or when standing?

- Never or rarely
- One or more times daily
- 15 or more times daily
- 30 or more times daily

5. Do you pace or move around repetitively? (e.g. walk to and fro across a room, or around the same path in the garden?)

- Never or rarely
- One or more times daily
- 15 or more times daily
- 30 or more times daily

6. Do you make repetitive hand and/or finger movements? (e.g. flap, wave, or flick your hands or fingers repetitively?)

- Never or rarely
- One or more times daily
- 15 or more times daily
- 30 or more times daily

7. Do you have a fascination with specific objects? (e.g. trains, road signs or other things?)

- Never or rarely
- Mild or occasional
- Marked or notable

8. Do you like to look at objects from particular or unusual angles?

- Never or rarely
- Mild or occasional
- Marked or notable

9. Do you have a special interest in the smell of people or objects?

- Never or rarely
- Mild or occasional
- Marked or notable

10. Do you have a special interest in the feel of different surfaces?

- Never or rarely
- Mild or occasional
- Marked or notable

11. Do you have any special objects you like to carry around?

- Never or rarely

- Mild or occasional
- Marked or notable

12. Do you collect or hoard items of any sort?

- Never or rarely
- Mild or occasional
- Marked or notable

13. Do you insist on things at home remaining the same? (e.g. furniture staying in the same place, things being kept in certain places, or arranged in certain ways?)

- Never or rarely
- Mild or occasional (does not affect others)
- Marked or notable (occasionally affects others)
- Serious or severe (affects others on a regular basis)

14. Do you get upset about minor changes to objects? (e.g. flecks of dirt on your clothes, minor scratches on objects?)

- Never or rarely
- Mild or occasional (does not affect others)
- Marked or notable (occasionally affects others)
- Serious or severe (affects others on a regular basis)

15. Do you insist that aspects of daily routine must remain the same?

- Never or rarely
- Mild or occasional (does not affect others)
- Marked or notable (occasionally affects others)
- Serious or severe (affects others on a regular basis)

16. Do you insist on doing things in a certain way or re-doing things until they are “just right”?

- Never or rarely
- Mild or occasional (does not affect others)
- Marked or notable (occasionally affects others)
- Serious or severe (affects others on a regular basis)

17. Do you play the same music, game or video, or read the same book repeatedly?

- Never or rarely
- Mild or occasional (not entirely resistant to change or new things)
- Marked or notable (will tolerate changes when necessary)
- Serious or severe (will not tolerate any changes)

18. Do you insist on wearing the same clothes or refuse to wear new clothes?

- Never or rarely
- Mild or occasional (not entirely resistant to change or new things)
- Marked or notable (will tolerate changes when necessary)
- Serious or severe (will not tolerate any changes)

19. Do you insist on eating the same foods, or a very small range of foods, at every meal?

- Never or rarely
- Mild or occasional (not entirely resistant to change or new things)

## Appendix E

- Marked or notable (will tolerate changes when necessary)
- Serious or severe (will not tolerate any changes)

20. What sort of activity will you choose if you are left to occupy yourself?

- A range of different and flexible self-chosen activities
- Some varied and flexible interests but commonly choose the same activities
- Almost always choose from a restricted range of repetitive activities

Barrett, S. L., Uljarević, M., Baker, E. K., Richdale, A. L., Jones, C. R. G., & 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(11), 3680-92.







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