

# Research Review: A systematic review and meta-analysis of sex differences in narrow constructs of restricted and repetitive behaviours and interests in autistic children, adolescents, and adults

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**Background:** Evidence that autism often manifests differently between males and females is growing, particularly in terms of social interaction and communication, but it is unclear if there are sex differences in restricted and repetitive behaviours and interests (RRBIs) when rigorously focusing on the narrow construct level (i.e., stereotyped behaviour, restricted interests, insistence on sameness, and/or sensory experiences). **Methods:** We conducted a systematic review and four random effects meta-analyses investigating sex differences in narrow construct measures of RRBIs in autistic children, adolescents, and adults (Prospero registration ID: CRD42021254221). Study quality was appraised using the Newcastle-Ottawa Quality Assessment Scale. **Results:** Forty-six studies were narratively synthesised and 25 of these were included in four random effects meta-analyses. Results found that autistic males had significantly higher levels of stereotyped behaviours (SMD = 0.21, 95% confidence interval (CI) [0.09, 0.33],  $p < .001$ ) and restricted interests (SMD = 0.18, 95% CI [0.07, 0.29],  $p < .001$ ) compared to autistic females. In contrast, there were no significant sex differences for sensory experiences (SMD = -0.09, 95% CI [-0.27, 0.09],  $p = .32$ ) and insistence on sameness (SMD = 0.01, 95% CI [-0.03, 0.05],  $p = .68$ ). The findings from the narrative synthesis were generally consistent with those from the meta-analyses and also found qualitative sex differences in the way RRBIs manifest. **Conclusions:** Our findings show significant differences in narrowly defined RRBIs in males and females. Practitioners need to be aware of such differences, which could be contributing to the under-recognition of autism in females and may not be captured by current diagnostic instruments. **Keywords:** Autism; sex differences; gender differences; systematic review; meta-analysis; restricted and repetitive behaviours and interests.

## 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 has a diagnosis of autism (Roman-Urrestarazu et al., 2021) and the sex<sup>1</sup> ratio is around four males to every one female (Fombonne, Quirke, & Hagen, 2009; Kreiser & White, 2014; Maenner et al., 2020; Solmi et al., 2022). However, the ratio is less extreme (around 3:1) in population-based studies, suggesting that there is a proportion of women and girls reporting high autism traits that are not receiving a diagnosis, despite meeting clinical criteria (Loomes, Hull, & Mandy, 2017).

There is research to suggest autism manifests differently in females, sometimes described as the Female Autism Phenotype (Hull, Petrides, & Mandy, 2020) which may contribute to their under-diagnosis. For example, on average, autistic females

are more likely to use strategies that mask autistic features or compensate for underlying difficulties, known as camouflaging, which would make them less likely to receive a diagnosis (Cook, Hull, Crane, & Mandy, 2021; McQuaid, Raitano Lee, & Wallace, 2021). Females may also have more age and gender-appropriate restricted and repetitive interests compared to males, such as those involving animals and fictional characters as opposed to vehicles, computers, or physics, which again could contribute to under-recognition of autism in females (Grove, Hoekstra, Wierda, & Begeer, 2018; Nowell, Jones, & Harrop, 2019).

## Sex 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, Zimmermann, & Poling, 2014). As a result, current

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understandings of autism may be biased towards a male-specific manifestation of the condition (Thompson, Caruso, & Ellerbeck, 2003). The notion of autism as a male condition has reduced 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, Young, & Weber, 2014; Mandy et al., 2012) or misinterpreted by key adults, including educational practitioners (Aggarwal & Angus, 2015; Holtmann & Bölte, 2007). The misinterpretation of diagnostically relevant behaviours 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, Ronald, Bolton, & Happé, 2012; Mandy et al., 2012). Additionally, the potential presence of camouflaging in schools may further hinder the early identification of autistic girls (Attwood et al., 2006; Dean, Harwood, & Kasari, 2017).

Due to a lack of reliable genetic biomarkers for autism (Goldani, Downs, Widjaja, Lawton, & Henden, 2014), the assessment of autism relies on reported and observed behaviours, often using 'gold standard' tools (Falkmer, Anderson, Falkmer, & Horlin, 2013; Ozonoff, Goodlin-Jones, & Solomon, 2005) such as the Autism Diagnostic Interview Revised (ADI-R; Rutter, Le Couteur, & Lord, 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 samples (Bargiela, Steward, & Mandy, 2016; Lai & Baron-Cohen, 2015; McCrimmon & Rostad, 2014) and therefore may not be sensitive to how autism presents in females (Wood-Downie, Wong, Kovshoff, Cortese & Hadwin, 2021). The sensitivity of the ADOS to autistic female characteristics has also been challenged by research findings suggesting that adult autistic females (diagnosed in childhood) are less likely to meet ADOS cut-off scores compared to males (Lai et al., 2011). Girls with comparable levels of autism trait severity to boys have also been found to be less likely to receive a diagnosis of autism (Russell, Steer, & Golding, 2011), suggesting that females need to surpass a higher threshold of severity to meet diagnostic criteria (Dworzynski et al., 2012; Kreiser & White, 2014). 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, Enticott, & Fitzgerald, 2013; Rivet & Matson, 2011).

### *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, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 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.

Reviews and large-scale studies into sex differences in autistic RRBIs at the broad construct consistently find that autistic females display fewer RRBIs than males (Frazier, Georgiades, et al., 2014; Frazier, Ratliff, et al., 2014; Supekar & Menon, 2015; Szatmari et al., 2012; see Lai et al., 2015; and van Wijngaarden-Cremers et al., 2014 for reviews). For example, a meta-analysis of 22 studies identified that females had, on average, fewer RRBIs (based on ADI-R and ADOS overall scores) compared to males (van Wijngaarden-Cremers et al., 2014).

A criticism of research focussing only on the broad construct of RRBIs 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). Consistently, studies that have explored sex differences in narrow constructs of RRBIs produce more mixed results, for example, no sex difference on ADI-R items such as stereotyped language, unusual sensory interests, and resistance to change (McLennan, Lord, & Schopler, 1993). Lai et al. (2011) also reported autistic females as having more 'lifetime sensory issues' compared to autistic boys. These studies suggest that sex differences in RRBIs may depend on the specific domain in question, however there is yet to be a systematic review and meta-analysis of narrow constructs of RRBIs based upon DSM-5 criteria (see Table S1), which provides the most recent diagnostic classification of autism. The current DSM-5 also includes sensory symptoms as a narrow construct within RRBIs and there has not yet been a systematic synthesis of sex differences in this specific domain. Accordingly, we conducted a systematic literature review and meta-analysis to explore sex differences in narrow constructs of RRBIs in autistic individuals.

### **Method**

The review was prospectively registered on PROSPERO (registration number: CRD42021254221). The review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA; Page et al., 2021).

## Search strategy

We searched 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 (see Table S1), including the population terms 'autism spectrum disorder' and 'autism spectrum condition'; comparator terms 'sex' and 'gender'; and outcome terms 'repetitive behavi?r\*', 'restricted interest', 'insistence on sameness', 'sensory', and 'circumscribed interest\*' (Appendix S1). We re-ran the search on 21st October 2022, to see whether there were any additional articles that reported data that could be included in the meta-analyses, though none were identified. An English language restriction was applied. In addition, reference lists of included studies were hand-searched. This process was first conducted by the first author. To check for reliability, a second author (HWD) independently completed abstract screening for 25% of studies. Cohen's Kappa test indicated substantial agreement between both reviewers (0.68). HWD also assessed the full-texts of 25% of studies for eligibility and again there was substantial agreement ( $\kappa = .74$ ).

## Eligibility criteria

Cross-sectional, peer-reviewed, articles including autistic males and females, of any age, and including an outcome measure of subdomains of RRBI based upon the DSM-5 autism diagnostic criteria, were included. Studies including participants with high autistic traits and/or participants self-identifying as autistic were also included. Studies that did not include a measure reporting a subdomain of RRBI according to the DSM-5 diagnostic criteria were excluded. See Table 1 for further details on inclusion/exclusion criteria and rationale. Further details (i.e., specific examples of outcome measures excluded) can be found in Appendices S2 and S3.

## Data extraction

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

## Study quality appraisal

The included studies were evaluated for quality using the Newcastle-Ottawa Quality Assessment Scale (N-OQAS, see Appendix S4), adapted for cross-sectional studies (Herzog et al., 2013).

## Data synthesis strategy

Findings from included studies are described in the narrative synthesis, which have been categorised into narrow constructs corresponding to the four RRBI subdomains. The key findings are described in the results section below (with a focus on qualitative differences between males and females which would not be apparent from the meta-analytic findings alone).

Random-effect meta-analyses were performed using Comprehensive Meta-Analysis (CMA; Borenstein, Hedges, Higgins, & Rothstein, 2013) for the four narrow construct measures of RRBI. Standardised mean differences (SMD) were calculated for autistic males and females, converting from other effect size metrics (e.g., odds ratio) if needed, using CMA. Where more than one measure was used in a study, the measure most closely reflecting the narrow construct being explored was

**Table 1** 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 RRBI 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 reviews (van Wijngaarden-Cremers et al., 2014) 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

used. Publication bias was assessed via visual inspection of funnel plots and through Egger's test. Heterogeneity was assessed using chi-squared tests and interpretation of the  $I^2$  statistic. We planned to conduct moderator analyses for different age groups and IQ levels. We also conducted a post-hoc sensitivity analysis restricted to high-quality studies and a meta-regression with year of publication as covariate, to assess whether study quality or year of publication were significant moderating variables.

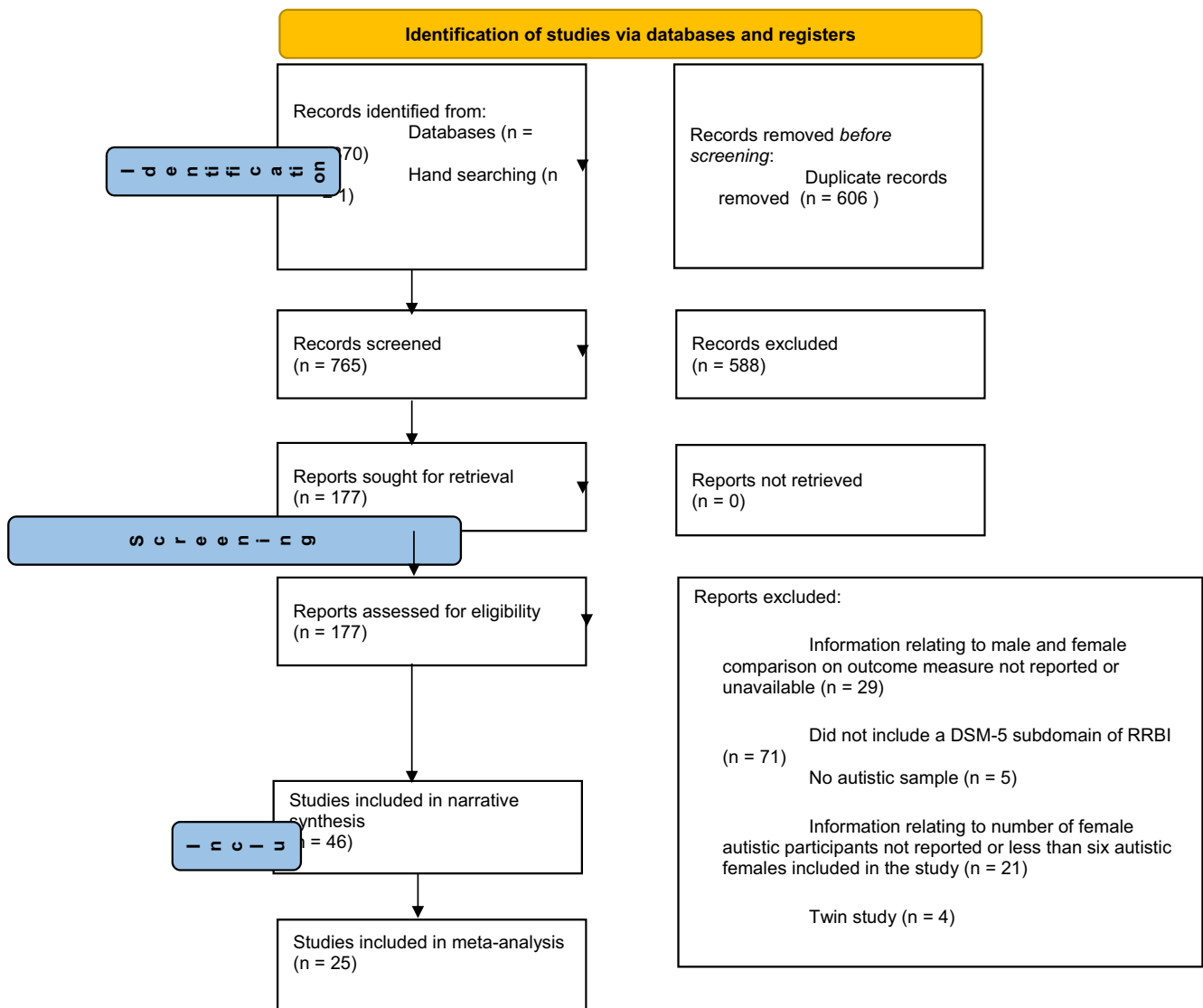
## Results

### Search results

As shown in Figure 1, from an initial 1,370 possible relevant references, a total of 46 unique studies were included within the narrative synthesis, of which 25 were included in the meta-analyses (further search details are provided in Appendix S5). Characteristics of all included studies are presented in Table S2.

### Characteristics of included studies

Stereotyped or repetitive motor movements, use of objects, or speech<sup>2</sup> ( $N = 29$ ) and highly restricted, fixated interests that are abnormal in intensity or focus<sup>3</sup> ( $N = 27$ ) 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<sup>4</sup> ( $N = 25$ ) and hyper- or hypo-reactivity to sensory input or unusual interest in sensory aspects of the environment<sup>5</sup> ( $N = 21$ ).



**Figure 1** PRISMA flow diagram of study identification and selection. From: Page et al. (2021). For more information, visit: <http://www.prisma-statement.org/>

The majority of studies included children ( $n = 42$ ) with four studies exploring RRBI outcomes exclusively in adults (those over 18 years; Aita et al., 2019; Barrett, Uljarević, Jones, & Leekam, 2018; English et al., 2021; Weiland, Polderman, Hoekstra, Smit, & Begeer, 2020). Diagnosis of autism was confirmed in 24 studies, often via clinical assessment and/or ADOS assessment ( $n = 15$ ) or autism trait screening tools or review of diagnostic reports ( $n = 9$ ).

### Study quality

A total of 10 studies received a score of 'good', 16 'satisfactory' and 20 'unsatisfactory' on the *N*-OQAS. Of the 20 'unsatisfactory' studies, studies 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. Additional details of study quality can be found in Appendix S6.

### Key findings from narrative synthesis

*Stereotyped or repetitive motor movements, use of objects, or speech.* 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., 2015). 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).

*Insistence on sameness, inflexible adherence to routine, or ritualised patterns of verbal or nonverbal behaviour.* 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. Antezana et al. (2019) reported higher endorsement of 'distress at small changes' in autistic females compared to males.

*Highly restricted, fixated interests that are abnormal in intensity or focus.* 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, Albright, Muskett, & Scarpa, 2019; Sutherland, Hodge, Bruck, Costley, & Klieve, 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 restricted interest in relation to the collection of 'seemingly random' items such as rocks, pens and stickers compared to males (Hiller et al., 2014, 2016). Grove et al. (2018) also found differences in the types of special interests men and women held (e.g., women more likely to report being interested in autism, whereas men were more likely to report interests in computers and gaming) though, of note, found no sex difference in terms of the intensity of restricted interests, suggesting that whilst the content or the way the interest is expressed might be different, the intensity may not be.

*Hyper- or hypo-reactivity to sensory input or unusual interests in sensory aspects of the environment.* 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). 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).

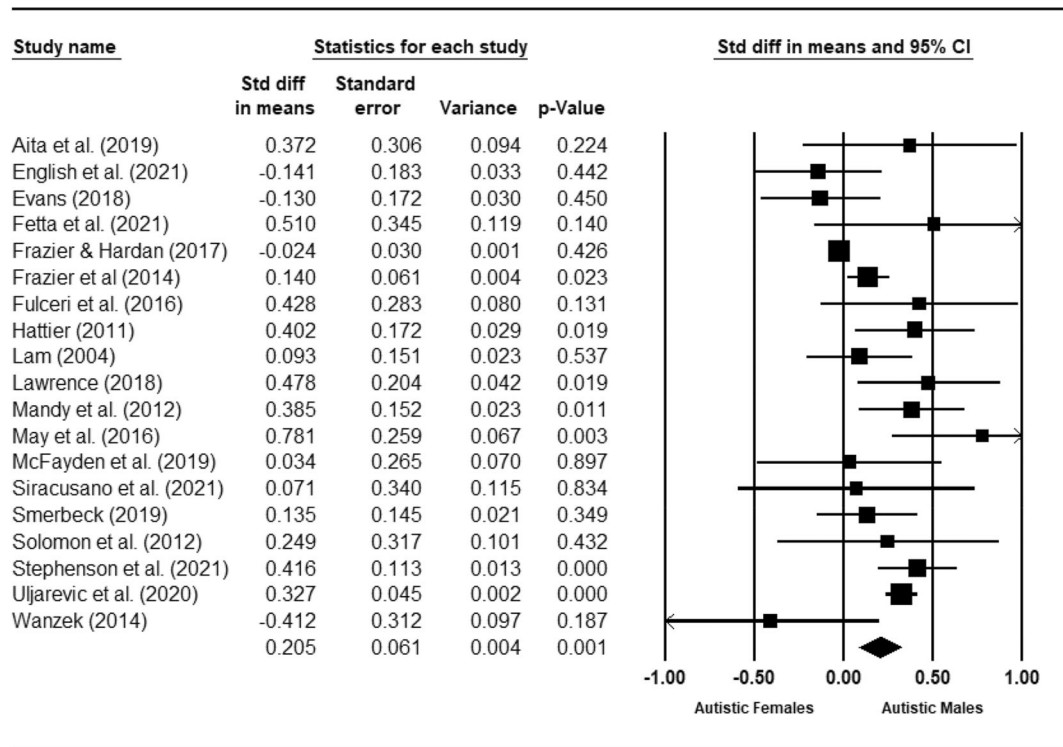
### Meta-analyses

*Publication bias and heterogeneity.* One plot (for 'restricted interests') appeared asymmetrical and Eggers test was significant ( $p < .001$ ), indicating publication bias (see Figure S1). Three other plots (stereotyped behaviour, insistence on sameness, and sensory experiences) appeared symmetrical, with most studies clustered around the overall SMD, suggesting no publication bias (see Figures S2–S4). Consistently, Eggers test was non-significant for these three analyses (for stereotyped behaviour,  $p = .16$ ; for insistence on sameness,  $p = .62$ ; and sensory,  $p = .22$ ).

*Stereotyped or repetitive motor movements, use of objects, or speech.* Significant differences were found, SMD = 0.21, 95% confidence interval (CI) [0.09, 0.33],  $p = .001$ , indicating that autistic males had significantly higher rates of stereotyped behaviour than autistic females (see Figure 2). Heterogeneity tests were significant,  $Q = 77.25$ ,  $p < .001$ , with  $I^2$  indicating moderate to substantial variance due to true heterogeneity ( $I^2 = 76.70$ ).

As Wanzek (2014) reported very different results to the majority of all other studies, a post-hoc sensitivity analysis was conducted by re-running the analysis with the removal of this data point. Results did not substantially change, with autistic males having significantly higher rates of stereotyped behaviour than autistic females, SMD = 0.22, 95% CI [0.10, 0.34],  $p < .001$ , though heterogeneity remained significant,  $Q = 74.35$ ,  $p < .001$ , with  $I^2$  again indicating substantial variance due to true heterogeneity ( $I^2 = 77.13$ ).

*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 = 0.01, 95% CI [-0.03, 0.05],  $p = .68$ , indicating that autistic males and females had similar rates of insistence on sameness (see Figure 3). Heterogeneity tests were not significant,  $Q = 11.19$ ,  $p = .80$ , with  $I^2 = 0.00$ .



**Figure 2** Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of stereotyped behaviours. <sup>a</sup>The data from McFayden et al. (2019) used in this meta-analysis are not the final data set used and reported in their published paper. <sup>b</sup>McFayden et al. (2019) data used in meta-analysis refer 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) refer to autistic (diagnosed and self-identifying) only

*Highly restricted, fixated interests that are abnormal in intensity or focus.* A random-effects meta-analysis found significant differences between autistic males and females,  $SMD = 0.18$ , 95% CI [0.07, 0.29],  $p = .001$ , indicating that autistic males had significantly higher rates of restricted interests than autistic females (see Figure 4). Heterogeneity tests were significant,  $Q = 25.57$ ,  $p < .01$ , with  $I^2$  indicating moderate variance due to true heterogeneity ( $I^2 = 53.07$ ).

*Hyper- or hypo-reactivity to sensory input or unusual interests in sensory aspects of the environment.* A random-effects meta-analysis found no significant difference between autistic males and females for sensory experiences,  $SMD = -0.09$ , 95% CI [-0.27, 0.09],  $p = .31$  (see Figure 5). However, heterogeneity tests were significant,  $Q = 49$ ,  $p < .001$ , with  $I^2$  indicating substantial variance due to true heterogeneity ( $I^2 = 77.60$ ).

*Moderators.* 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 as planned. Only three studies (Aita et al., 2019; Hattier, Matson, Tureck, & Horovitz, 2011; Lawrence, 2017) featured participants with identified  $IQ < 70$ , and therefore we were not able to investigate cognitive functioning as a

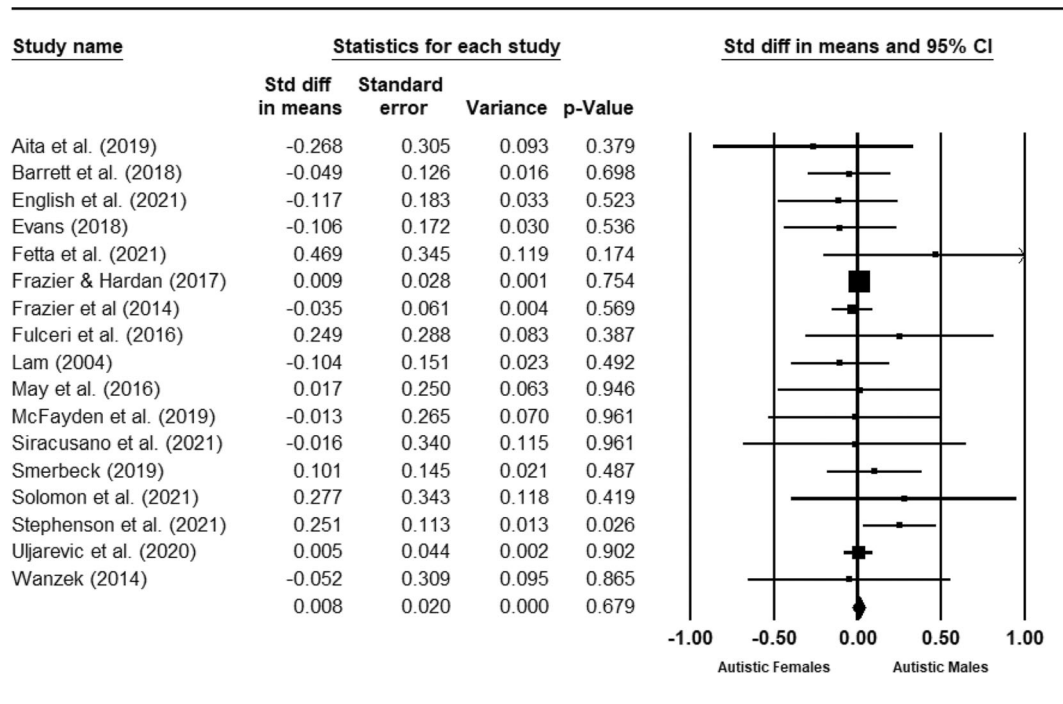
possible moderating variable as planned, nor were there sufficient studies for each behavioural exemplar to investigate this as a potential moderating variable using meta-regression.

A post-hoc sensitivity analysis was conducted restricted to studies judged as good or very good, which led to the same pattern of results as in the main analysis, suggesting that study quality was not a moderator. Specifically, males had significantly higher levels of stereotyped behaviours ( $p < .001$ ) and restricted interests ( $p = .001$ ). No significant differences were found for insistence on sameness ( $p = .52$ ) or sensory experiences ( $p = .91$ ).

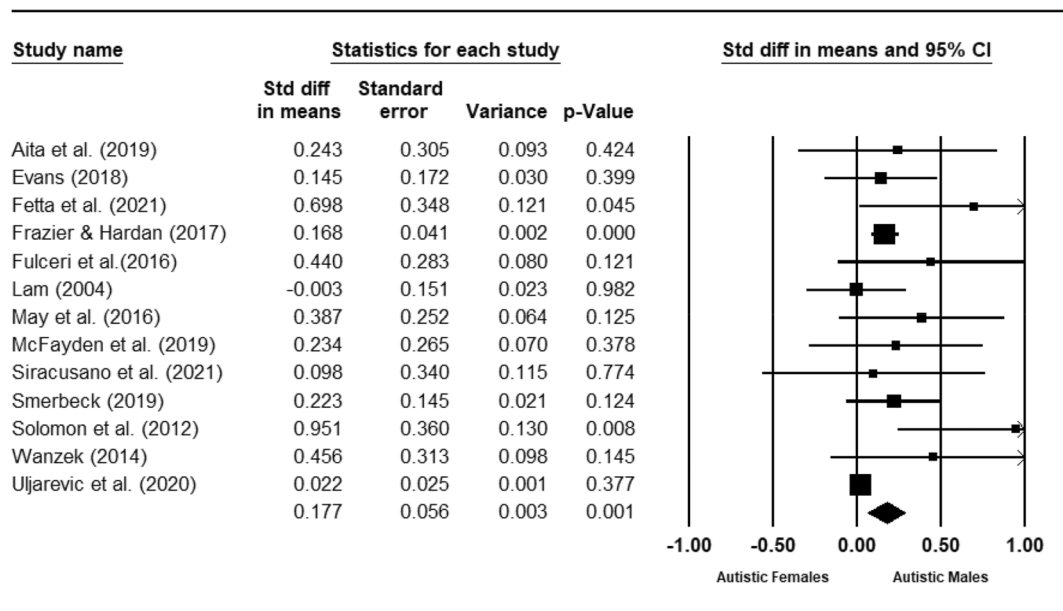
*Meta-regression.* A meta-regression was run with year as the covariate for the four analyses, which were all non-significant, suggesting that year of publication was not a significant moderator (stereotyped behaviours,  $p = .80$ ; insistence on Sameness,  $p = .18$ ; restricted interests,  $p = .91$ ; sensory experiences,  $p = .95$ ).

## Discussion

This systematic review and meta-analysis explored, for the first time, sex differences in the narrow constructs of autistic RRBIs. Data from 46 studies were included in a narrative synthesis, of which 25 of these were included in four random-effects meta-analyses, in line with the subdomains of RRBIs



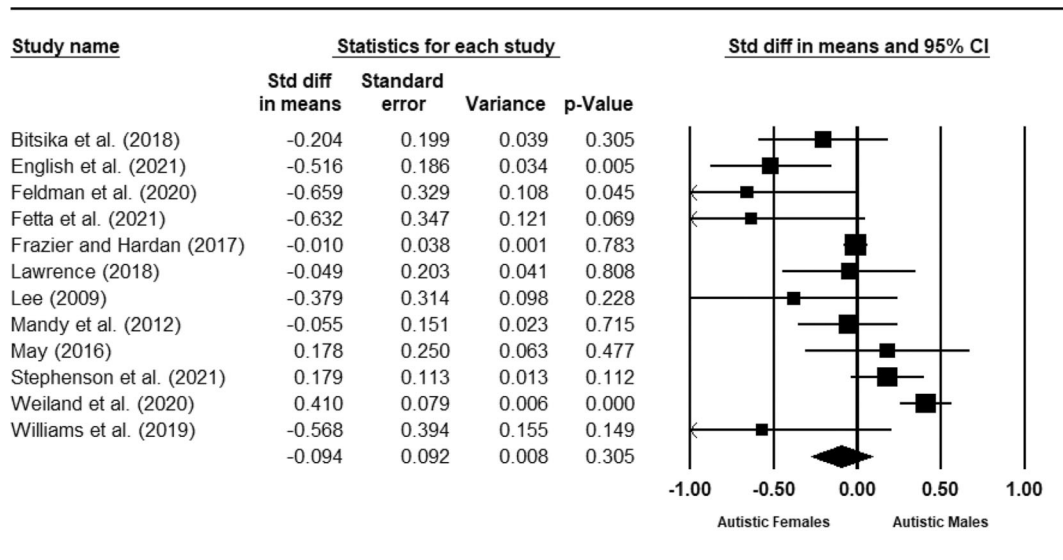
**Figure 3** Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of insistence on sameness. <sup>a</sup>The data from McFayden et al. (2019) used in this meta-analysis are not the final data set used and reported in their published paper. <sup>b</sup>McFayden et al. (2019) data used in meta-analysis refer 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) refer to autistic (diagnosed and self-identifying) only. <sup>d</sup>The *p*-value for Stephenson et al. (2021) is different from 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)



**Figure 4** Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of restricted interests. <sup>a</sup>The data from McFayden et al. (2019) used in this meta-analysis are not the final data set used and reported in their published paper. Likewise, data in this meta-analysis refer 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) are prior to controlling for autism severity

outlined by the DSM-5. These findings indicate significant differences between autistic males and females in the presentation of stereotyped behaviours and restricted interests, with autistic males presenting with higher levels of these behaviours

than autistic females, even if these findings should be considered with caution due to significant inter-study heterogeneity. No significant sex differences were identified for insistence on sameness or sensory experiences. The findings from the narrative



**Figure 5** Forest plot for meta-analysis comparing autistic males and females on narrow construct measures of sensory experiences. 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 are not the final data set used and reported in their published paper. Likewise, data in this meta-analysis refer 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) refer to autistic (diagnosed and self-identifying) only

synthesis were generally consistent with those from the meta-analyses, as well as some qualitative sex differences being reported, such as females being more likely to report having interests related to people, relationships and/or living beings (e.g., psychology; animals), whereas males reported interests related to technology, objects and/or mechanical topics (e.g., science; transportation).

The sex differences found for restricted interests are in line with previous studies indicating fewer restricted interests in autistic females compared to males (Uljarević, Alvares, et al., 2021; Uljarević, Frazier, et al., 2021). The fact that autistic females' restricted interests appear to be more socially appropriate and developmentally normative than males' interests (McFayden et al., 2019; Sutherland et al., 2017), for example in animals, could contribute towards autistic females being missed by practitioners and less likely to be referred for diagnostic assessment. Consistently, Whitlock, Fulton, Lai, Pellicano, and Mandy (2020) found that, when educational staff were presented with vignettes of the female autism phenotype (which included social or relational restricted interests), they reported that they were unlikely to seek support from an external professional. The type of restricted interests displayed by autistic females may also be difficult to identify using diagnostic tools, 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.

Although our findings suggest that restricted 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, Clarke et al. (2021) coded ADOS-2 administrations – using the newly developed Gendered Autism Behavioural Scale (GABS) – and found that females (for one of the two cohorts) had higher levels of relational interest on the GABS than males, despite there being no overall sex difference on the originally scored ADOS-2, as well as low correlations between the GABS and ADOS domain scores. Overall, these findings suggest that conventional diagnostic instruments may not be sensitive to specific autism presentations, including relational interests, which could therefore mask nuanced sex differences, which has also previously been demonstrated in the social communication and interaction domains (Wood-Downie et al., 2021).

Future research exploring autistic restricted interests will need to ascertain the respective levels of restricted 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) restricted interests (Frazier, Ratliff, et al., 2014; Solomon, Miller, Taylor, Hinshaw, & Carter, 2012; Sutherland et al., 2017), which likely will require the adaptation and/or development of new instruments.

The significant sex differences identified for stereotyped behaviours are 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, Young, & Weber, 2021). Lower levels of stereotyped behaviours in autistic females could contribute towards their under-recognition, particularly as these behaviours are readily externally observable and perhaps more in accordance with the male-stereotyped nosology of autism than other RRBI narrow constructs. 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. Culture-oriented judgements could also influence how stereotyped behaviours are interpreted, for example lining up toys being interpreted as atypical in boys whilst a sign of neatness or organisation when displayed by girls (Gal, 2011).

There is also emerging research suggesting that autistic females might be motivated to mask autistic features, known as camouflaging (Hull et al., 2020), which could be contributing towards lower levels of stereotyped behaviours observed by others. The majority of research into camouflaging has focussed on the social interaction and communication domain (Cook et al., 2021, for a review), however autistic adults have described suppressing stereotyped behaviours as a camouflaging strategy (e.g., Hull et al., 2017). Future research needs to understand how camouflaging could be impacting sex differences in stereotyped behaviours, and RRBI as a whole. Exploration as to how stereotyped behaviours are observed and interpreted by those key to the 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 in females, particularly during childhood where observation reports are more heavily relied upon.

Our meta-analytical findings suggest there are no significant differences between autistic females and males for sensory experiences, though some differences were reported in the narrative synthesis for highly specific aspects of such experiences (e.g., taste), which may have contributed to the substantial amount of heterogeneity in this analysis. Considering the ADOS-2 and ADI-R only capture 'unusual sensory interests' and 'undue sensitivity to noise' (e.g., hypersensitivity), such differences could be missed during diagnostic assessments, highlighting the importance of individuals being asked about aspects of sensory experiences for all key senses.

### Implications for practice

One of the key implications for practice is the need for professionals to be aware and assess for potential nuanced RRBI sex differences. For example, our findings suggest that females may hold more gender-normative restricted interests than autistic males, which may not be captured by conventional instruments used as part of autism assessments (Clarke et al., 2021). Accordingly, females may be less likely to be referred for an assessment (e.g., by school staff), as well as less likely to receive a diagnosis if assessed, particularly if not presenting with additional cognitive and/or behavioural difficulties (Dworzynski et al., 2012; Whitlock et al., 2020).

The moderating influence of biological and environmental sex and gender-related factors at different timepoints within development is a possible reason why some behavioural presentations are more or less identifiable at different age ranges, based on how sex or gender-normative or impactful on functioning they are considered to be (Lai, Lin, & Ameis, 2022). For example, a female child with a restricted interest in an age-appropriate toy or topic (e.g., My Little Pony) may be less likely to be identified, based on the content of the interest alone, compared to a female adult with the same interest, due to differences in what is deemed contextually typical for the age of the individual, illustrating the importance of assessing the functional impact of interests (even if they appear age-appropriate), as well as future research investigating age as a moderating variable.

Females may also have similar levels of insistence on sameness and sensory symptoms to autistic males, though these may be less externally observable than, for example, stereotyped behaviours, compounding difficulties in identifying females on the autism spectrum. As a result, it is important for clinicians to be aware of these potential differences, particularly in relation to developmental stage, and consider them as part of diagnostic assessments, such as by asking the individual and their caregivers about sensory experiences, and whether they impact on everyday functioning (Lai et al., 2022).

As our findings provide robust evidence that RRBI sex differences vary depending on the specific narrow construct in question, clinicians and researchers need to adopt fine-grained assessment of RRBI during assessments and research. For example, by considering each of the four subdomains of the DSM-5 separately, as measuring overall levels could lead to subtler differences being overlooked. Additional training for educational staff and clinicians on the topic of autistic sex differences in these sub-domains, through a neurodiversity lens that does not pathologise autistic behaviours and interests, is also required to improve clinician confidence levels and support earlier identification of autistic females.

## Limitations

A number of limitations should be considered, some related to the systematic review and meta-analysis per se, and others related to methodological issues of the included studies. In relation to limitations of the systematic review, due to financial constraints, we limited the search to studies in English, as we could not fund paper translation. However, research has indicated that excluding non-English studies had minimal effect on overall conclusions in systematic reviews (Nussbaumer-Streit et al., 2020).

Regarding limitations of the included studies, a proportion of studies included in the meta-analyses (36%) were rated as 'unsatisfactory' in quality analysis, primarily 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.

In addition, the majority of studies included participants who already had clinical diagnoses, which limits our ability to generalise the findings to possibly autistic females without diagnoses (e.g., to help explain underdiagnosis), particularly as one hypothesis for their underrecognition is due to presenting with non-traditional autistic features, sometimes referred to as the Female Autism Phenotype (Hull et al., 2020). One proposed aspect of this phenotype is that females are more likely to hold relational interests, whereas males are more likely to hold mechanical interests, which is in accordance with our findings. Again, though, the research which has informed this hypothesised phenotype tends to be based on samples of diagnosed autistic males and females and therefore may not be generalisable to those without diagnoses. However, Bargiela et al. (2016) found that late-diagnosed autistic females believed that stereotypes associated with autism, such as having interests in maths and science, contributed to their underrecognition, providing preliminary evidence that qualitative differences in interests may be a feature of some autistic females who have not yet received diagnoses.

Research that directly compares diagnosed with non-diagnosed autistic individuals is needed to more fully understand how non-typical presentations may contribute to the underrecognition of autism which, to our knowledge, has not yet been done for RRBI. Two studies have researched this in other domains, finding both similarities (e.g., friendship motivation; association between camouflaging and levels of autistic traits) and differences (e.g., social

functioning; overall levels of camouflaging) between females diagnosed with autism and those without diagnoses but high levels of autistic traits (Belcher, Morein-Zamir, Stagg, & Ford, 2022; Milner, Mandy, Happé, & Culvert, 2023), suggesting there may also be similarities and differences in respect to RRBI which is an important avenue for future research.

A significant amount of heterogeneity was identified for restricted interests, stereotyped behaviours, and sensory experiences. Previous research suggests that IQ and age may moderate sex difference in autism (Jiujiu, Kelley, & Hall, 2017; Stratis & Lecavalier, 2013; Wood-Downie et al., 2021) and therefore it is possible that IQ and age account for some of the heterogeneity within the current analyses, which we had planned to investigate through moderator analyses. Unfortunately, due to the under-representation of individuals with lower IQ, we were unable to conduct analyses to see whether IQ was a moderator. Similarly, due to studies including participants with a very wide age range, we were also unable to investigate whether age was a moderating variable. Additionally, due to the small amount of studies for each behavioural exemplar, we were unable to investigate this as a 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 contribute to the variation in results. As such, it will be important for future research to include individuals with intellectual disabilities, narrower age ranges, as well as using a wider range of behavioural exemplars as outcome measures (such as hyper- and hypo-sensitivity and sensory interests), so that future analyses can investigate whether these moderate sex differences. Furthermore, gender-diverse individuals have been reported to present with higher rates of autism, compared to cisgender individuals (Warrier et al., 2020), therefore future research should also incorporate exploration of autistic experiences outside of the gender binary.

## Conclusion

This systematic review and meta-analysis identified significant sex differences in two narrow constructs of RRBI (as defined by the DSM-5) – stereotyped behaviours and restricted interests – with autistic males reporting higher rates compared to autistic females. Some qualitative differences in the way in which RRBI manifest between sexes were also reported, such as in the type of restricted interests females hold. In contrast, no significant differences were reported for sensory experiences or insistence on sameness. This highlights the importance of fine-grained analysis into the narrow constructs of RRBI, rather than merely exploring at the broad construct level. These differences could, in part,

contribute to the late and under-diagnosis of autism for females and highlights the importance of developing assessment tools that are sensitive to how autism may manifest in females. More research is needed to explore the potential moderating variables of IQ, age, and specific behavioural exemplars.

## Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article:

**Figure S1.** Funnel plot for restricted interests meta-analysis.

**Figure S2.** Funnel plot for stereotyped behaviours meta-analysis.

**Figure S3.** Funnel plot for insistence on sameness meta-analysis.

**Figure S4.** Funnel plot for sensory meta-analysis.

**Table S1.** Examples of broad/narrow constructs and associated behavioural exemplars based upon DSM-5 diagnostic criteria.

**Table S2.** Study and sample characteristics for all included studies.

**Appendix S1.** Full search terms.

**Appendix S2.** Further details of eligibility criteria.

**Appendix S3.** Further details regarding sensory scores.

**Appendix S4.** Newcastle-Ottawa Quality Assessment Scale with adaptations.

**Appendix S5.** Further information about search results.

**Appendix S6.** Further details of study quality for all studies.

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## Key points

- Previous research into restricted and repetitive behaviours and interests (RRBIs) at the broad construct level tends to find that autistic males display more of these behaviours than autistic females.
- We conducted the first systematic review and meta-analysis focusing on narrow constructs of RRBIs, to investigate possible fine-grained sex differences.
- Autistic males had significantly higher levels of stereotyped behaviours and restricted interests compared to autistic females. No differences were found for sensory experiences or insistence on sameness. Autistic males and females also often hold qualitatively different types of restricted interests.
- Sex differences in RRBIs vary depending on the specific narrow construct, which could contribute to the under-recognition of autism in females, and clinicians need to consider as part of diagnostic assessments.

## Endnotes

1. The effects of biological sex and socially constructed gender are difficult to separate, however, the majority of studies featured in this review refer to biological sex only, therefore the term 'sex' is used throughout.
2. Hereafter referred to as 'stereotyped behaviours' unless other terminology is used in specific studies.
3. Hereafter referred to as 'restricted interests' unless other terminology is used in specific studies.
4. Hereafter referred to as 'insistence on sameness' unless other terminology is used in specific studies.
5. Hereafter referred to as 'sensory experiences' unless other terminology is used in specific studies.

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