



University of
Salford
MANCHESTER

Exploring the female autism phenotype of repetitive behaviours and restricted interests (RBRIs) : a systematic PRISMA review

Allely, CS

<http://dx.doi.org/10.1108/AIA-09-2018-0030>

Title	Exploring the female autism phenotype of repetitive behaviours and restricted interests (RBRIs) : a systematic PRISMA review
Authors	Allely, CS
Type	Article
URL	This version is available at: http://usir.salford.ac.uk/id/eprint/50308/
Published Date	2019

USIR is a digital collection of the research output of the University of Salford. Where copyright permits, full text material held in the repository is made freely available online and can be read, downloaded and copied for non-commercial private study or research purposes. Please check the manuscript for any further copyright restrictions.

For more information, including our policy and submission procedure, please contact the Repository Team at: usir@salford.ac.uk.

**Exploring the Female Autism Phenotype of Repetitive Behaviours and Restricted Interests (RBRI):
A Systematic PRISMA Review**

Abstract

Purpose: There is a need for increased understanding, awareness and recognition of the autism female phenotype in terms of Repetitive Behaviours and Restricted Interests (RBRIs).

Design/methodology/approach: A systematic PRISMA review was conducted. The main aim of the present systematic review is to identify studies which have investigated RBRIs in females with ASD or the differences in RBRIs between males and females with ASD.

Findings: Nineteen relevant articles were identified. Five studies found no significant evidence to support the notion of sex differences in RRBIs in ASD. One study did not report any differences in RRBIs between males and females with ASD. Twelve studies found evidence that males with ASD had significantly more RRBIs compared to females with ASD. Lastly, one study found that girls with ASD have features of RRBIs which are exhibited more compared to boys with ASD.

Practical implications: The RBRIs exhibited in autistic females are not sufficiently captured by most currently diagnostic instruments. Clinicians are less likely to identify the RBRIs in females as they tend not to be the typical repetitive behaviours commonly associated with ASD. It has been recommended that clinicians consider 'females as a whole' in terms of their clinical presentation and look for any indication of RBRIs, even repetitive interests which appear clinically innocuous.

Research limitations/implications: There is a real lack of in-depth knowledge and understanding of the female phenotype of ASD and such lack of knowledge has a detrimental impact on the identification of autistic females and a lack of identification can have negative consequence. This is important to address in future research as it is well-established that the earlier the diagnosis the better the outcomes due to the timely access to appropriate interventions.

Originality/value: There is relatively little research investigating RBRIs in autistic women and girls. There is a real need to highlight the importance of understanding and recognising how RBRIs can differ between males and females with ASD.

Keywords: RBRIs; Repetitive behaviours and restricted interests; Autism spectrum disorder; autism; females; women; woman; girls; diagnosis; gender

Autism spectrum disorder (ASD) is a neurodevelopmental condition which is characterised by social communication and social interaction difficulties in addition to restricted, repetitive behaviours or interests (RBRIs) (American Psychiatric Association, APA, 2013). There is significant variability in the clinical presentation of the ASD symptomology across individuals even though they all share the same core symptoms (Veselinova, 2014). In the Diagnostic and Statistical Manual of Mental Disorders, 4th Edition (DSM-IV, APA, 1994), autistic disorder was considered to be one of four categorical diagnoses that consisted of a group of disorders referred to as pervasive developmental disorders (PDD). As well as autistic disorder, the PDD group comprised Asperger's disorder, childhood disintegrative disorder, Rett's disorder and pervasive developmental disorder not otherwise specified (APA, 2013). In the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) the subtypes of ASD have been removed (e.g., autistic disorder and Asperger disorder). In the DSM-5 there is now just a single category of ASD (Maenner et al., 2014). ASD has an early developmental onset of persistent, typically lifelong symptoms. About four males are diagnosed with ASD for every female (e.g., Fombonne 2009). The possible explanations for this male-to-female ratio remain elusive (Adamou, Johnson, & Alty, 2018).

Repetitive Behaviours and Restricted Interests (RBRIs): Current RRB Diagnostic Criteria

Repetitive behaviours and restricted interests (RBRIs) characterise behaviours that can include repetitive motor movements, sensory reactions, rituals, routines, and restricted interests. RBRIs are common during early typical development (e.g., Arnott et al., 2010; Leekam et al., 2007). What makes the RBRIs which can be exhibited in early typical development different from those RBRIs exhibited in autistic individuals is the *intensity* of these behaviours. A study by South and colleagues (2005) showed how debilitating RBRIs can be (even in a very high-functioning sample comprising of Asperger's syndrome and high functioning autism individuals) with respect to the frequency of occurrence and the level of distress these RBRIs can cause for individuals as well as their families (South, Ozonoff, & McMahon, 2005). As highlighted above, RBRIs form an essential domain for a diagnosis of ASD (APA, 2013). Moreover, RBRIs have been found to be one of the earliest predictors exhibited in infants of a later diagnosis of ASD (e.g., Ozonoff et al., 2008; Wolff et al., 2014). Research has indicated that there exist two main subtypes of RBRIs (see Leekam, Prior, & Uljarevic, 2011). One being repetitive sensory and motor (RSM) behaviours which consist of repetitive motor behaviours and unusual sensory responses such as simple motor stereotypies and excessive smelling or touching of objects. The other subtype being the insistence on sameness (IS) which includes

routines, rigid behaviours and restricted interests (e.g., Bishop et al., 2013; Honey, McConachie, Turner, & Rodgers, 2012).

For the RBRI category in the DSM-5 criteria for ASD, there are four symptoms and the individual must exhibit at least two of the four, currently or by history. The four symptoms are:

1. Stereotyped or repetitive motor movements, use of objects or speech (such as simple motor stereotypies, echolalia, repetitive use of objects, lining up toys or flipping objects, or idiosyncratic phrases).
2. An insistence on sameness, excessive adherence to routines, ritualised patterns of verbal or nonverbal behaviour, or excessive resistance to change (such as motoric rituals, insistence on same route or food, rigid thinking patterns, repetitive questioning or extreme distress at small changes).
3. Highly restricted, fixated interests that are abnormal in intensity or focus (such as strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interests).
4. Hyper- or hypo-reactivity to sensory input or unusual interest in sensory aspects of environment; (such as an apparent indifference to pain/heat/cold, adverse response to specific sounds or textures, excessive smelling or touching of objects, fascination with lights or spinning objects). (see DSM-5, 2013, pp. 50).

Repetitive behaviours and restricted interests (RBRI) between males and females with ASD

Numerous researchers and clinicians have argued that one of the potential explanations for more males being diagnosed with ASD is that males display more (on average) RBRI than females. RBRI are an ASD symptomology which may be recognised more easily (Hartley & Sikora, 2009; Van Wijngaarden-Cremers, van Eeten, Groen, Van Deurzen, Oosterling, & Van der Gaag, 2014; Zwaigenbaum et al., 2012; Mandy, Chilvers, Chowdhury, Salter, Seigal, & Skuse, 2012; Koenig & Tsatsanis, 2005; Kreiser & White, 2014; Rivet & Matson, 2011). In females with a higher IQ or with less extreme stereotypies ASD often goes undetected (e.g., Baird et al., 2011). In the 22 studies that van Wijngaarden-Cremers and colleagues (2014) included in their meta-analysis it is possible that autistic females with a higher IQ may have been missed. If this was the case, then the authors argue that the meta-analysis would have *overestimated* problems in females in the domain of communication, social behaviour and RBRI restricted interests which was not the case. Instead,

autistic males and females exhibited similar symptom severity on communication and social behaviour. However, autistic girls exhibited less RBIs compared to autistic boys. In their meta-analysis, van Wijngaarden-Cremers and colleagues (2014) were not able to include intellectual disability as a confounder because of the lack of specific data on this in the original articles they identified. The key finding from the meta-analysis was that autistic boys exhibited more RBIs compared to autistic girls. RBIs are not unique to ASD as they can be found in children with an intellectual disability and severe deprivation and in typically developing children with intelligence which is within the normal range (Muthugovindan & Singer, 2009).

Limitations with the current RRBI diagnostic criteria

A limitation of the current RRBI diagnostic criteria is that it does not represent the full range of RBIs types (Mandy et al., 2012). Many autistic females may have very extreme interests or behaviours but in areas which fall out with the 'typical' ASD interests (which are so stereotypical and commonly found in ASD males), which would exclude them from fulfilling the criteria for RBIs for a diagnosis of ASD (Hull, Mandy, & Petrides, 2017). Or they may have interests which are a preoccupation with parts of an object which is less obvious than it is in males, less rituals, routines and stereotypical mannerisms (Nicholas et al., 2008), less factual expertise (e.g., knowledge of subway or train routes) and less oddly formal play (Mandy et al., 2012). An example which clearly demonstrates how the quality of RBIs may be different in autistic females is a young woman who always carried a number of well-worn books wherever she went. She would constantly read the books at the expense of all social interactions. This may be a type of repetitive behaviour which is not easily identified or recognised as being a circumscribed or 'special interest' (Halladay et al., 2015).

Present Study

Some studies support the notion that the sex differences appear to emerge only later in development as several studies have found that there are no differences in the behavioural presentation between ASD male and female toddlers (e.g., Reinhardt, Wetherby, Schatschneider, & Lord, 2015; Postorino, Fatta, De Peppo, Giovagnoli, Armando, Vicari, & Mazzone, 2015). This suggests that females may learn to mask or camouflage their autistic traits, which would support the female phenotype theory. However, if the difference only emerges later in development the question this raises is why are girls not diagnosed earlier like boys. However, there are a number of

studies which do show differences between autistic boys and girls (e.g., Rynkiewicz et al., 2016). Therefore, to date, the literature on whether there are sex/gender differences in ASD symptomology is inconsistent. The main aim of the present systematic review is to identify studies which have investigated RBRI in females with ASD or the differences in RBRI between males and females with ASD.

Methods

A total of five internet-based bibliographic databases were searched in order to identify studies which empirically investigated camouflaging or masking behaviour in females with ASD. Specifically, PsycARTICLES Full Text; AMED (Allied and Complementary Medicine) 1985 to November 2018; PsycEXTRA 1908 to December 10, 2018; PsycINFO 2002 to December Week 5 2018 and Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily 1946 to January 04, 2019. The search on the five databases was conducted on 7th January 2019. The search followed PRISMA guidelines (see Liberati et al., 2009; Moher, Liberati, Tetzlaff, & Altman, 2009). The search was not restricted by date. Search terms were applied to title. The following search criteria were entered into the five databases: ("repetitive behavio* and restricted interests" or RBRI* or "repetitive/stereotyped behavio*" or RRB* or "restricted and repetitive behavio*" or "restricted, repetitive behavio* and interests" or RRBI* or "restricted and repetitive behavio*" or "stereotypic and repetitive behavio*" or "repetitive sensory and motor behavio*" or "stereotypic behavio*" or "repetitive and stereotyped behavio*" or "restricted repetitive pattern of interests and behavio*" or "stereotypic/ repetitive movement*" or "motor stereotypies" or "repetitive and/or restricted behavio* and interests").m_titl. AND (ASD* or "autis* spectrum disorder*" or autis* or "autis* spectrum condition*" or asperger*).m_titl. AND (gender or sex or female* or women or woman or girl*).m_titl.

This search returned a total of nine articles. Following the removal of duplications there were five articles which were all relevant for the review. As well as the searches carried out on the five databases listed above, a variety of permutations of ASD in relation to female differences in RBRI were entered into Google Scholar and thoroughly screened for any potentially relevant articles not identified through the database searches. For instance, RBRI AND female AND autism; "repetitive behaviours" AND autism AND female; "repetitive behaviors" AND autism AND female; gender AND autism AND repetitive; "restricted and repetitive behaviour" AND autism AND female; etc. This resulted in fourteen further studies which were identified as being relevant to the present review (see Figure 1. For PRISMA Flow Diagram of this process). Lastly, because this is a relatively

under-researched area within autism research, the decision was made for the present systematic review to adopt an inclusive approach. No exclusion criteria were implemented for the studies identified which have explored RRBIs in females with ASD or the differences in RRBIs between males and females with ASD. All papers published since 2008 will be considered for inclusion in the present review.

Results

A total of nineteen articles were identified as relevant to the present review.

Sex Differences in RRBIs in ASD

Out of the total of 19 articles, five found no significant evidence to support the notion of sex differences in RRBIs in ASD (Solomon, Miller, Taylor, Hinshaw, & Carter, 2012; Andersson, Gillberg, & Miniscalco, 2013; Harrop, Gulsrud, & Kasari, 2015; Reinhardt, Wetherby, Schatschneider, & Lord, 2015; Knutsen, Crossman, Perrin, Shui, & Kuhlthau, 2018). It is important to emphasise one of these studies here. The study carried out by Knutsen and colleagues (2018) revealed more similarities than differences between males and females with ASD in the core diagnostic domain of RRBIs based on clinical observations. However, they did find something interesting. Compared to similar males, younger higher functioning and older lower functioning females exhibited reduced rates on the Autism Diagnostic Observation Schedule restricted and repetitive behaviour subcategory *unusually repetitive/excessive, stereotyped behaviours*. It is important to highlight that this study conducted by Knutsen and colleagues (2018) uses the biggest known sample to date of 1024 individually matched female and male children with ASD to investigate sex differences in RRBIs based on clinician observation. One study did not report any differences in RRBIs between males and females with ASD (Chowdhury, Benson, & Hillier, 2010). However, they only had one female with ASD in their sample and therefore they could not carry out any analysis looking at differences between males and females.

Twelve studies found evidence that males with ASD had significantly more RRBIs compared to females with ASD (Hartley & Sikora, 2009; Bölte, Duketis, Poustka, & Holtmann, 2011; Hattier, Matson, Tureck, & Horovitz, 2011; Sipes, Matson, Worley, & Kozlowski, 2011; Mandy, 2012; Park, Cho, Cho, Kim, Kim, Shin et al., 2012; Szatmari, Liu, Goldberg, Zwaigenbaum, Paterson, Woodbury-Smith et al., 2012; Frazier, Georgiades, Bishop, & Hardan, 2014; Hiller, Young, & Weber, 2014; Wilson, Murphy, McAlonan, Robertson, Spain, Hayward et al., 2016; Supekar & Menon, 2015;

Dean, Harwood, & Kasari, 2017). Lastly, one study found that girls with ASD have features of RRBI which are exhibited more compared to boys with ASD (Antezana et al., 2018). Specifically, they found that the items that were found to best-discriminate gender were heightened stereotyped behaviours and restricted interest items in the boys and compulsive, sameness, restricted, and self-injurious behaviour items in the girls. This study is the first to find that girls with ASD may have increased compulsive, sameness, and restricted RRBI compared to boys (Antezana et al., 2018).

The study conducted by Hiller, Young and Weber (2014) is worth pointing out here as it found that girls presented with both less and *different* restricted interests. A major contribution of this work is its exploration of the specific types of restricted interests displayed by boys and girls. For the 89 % of boys and 58 % of girls who did present with a fixated interest, the findings indicated that girls and boys present differently in terms of the types of fixations that they have. Specifically, compared to girls, boys were more likely to demonstrate fixated interests with televisions or video games, while girls were more likely to demonstrate interests around random objects. This included animals, rocks, shells, or books. Interestingly, when the sample was split into older and younger children, these seemingly random fixations held by many girls, remained the most common category (Hiller, Young, & Weber, 2014). Compared to males, the restricted and repetitive interests among females were thus more difficult to categorise and “identify as atypical” (Hiller et al., 2014, pp. 1391). Results supported the finding that, compared to boys, fewer girls with ASD exhibited restricted interests and other behaviours such as lining up or sorting objects (Hartley & Sikora 2009; Mandy et al., 2012).

The study by Mandy and colleagues (2012) also revealed differences between males and females with ASD on certain items within the RRBI domain. They found that, compared to females, males were especially likely to score on items measuring ‘oddly formal play’ involving lining up toys and having ‘a large store of factual information’. The authors suggest that these are both behaviours which are relevant to the systematising construct which has been advocated by Baron-Cohen (2002). Mandy and colleagues go on to suggest that their findings are consistent with Baron-Cohen’s ‘extreme male brain’ theory of ASD which would predict that, even amongst individuals with ASD, males would exhibit higher scores for systemising (Mandy et al., 2012).

Interestingly, Bölte and colleagues (2011) in a sample of 35 males and 21 females with higher functioning ASD and unaffected sibling controls investigated visual attention to detail (ATTD) and selected executive functions (EF). Based on the Autism Diagnostic Interview-Revised (ADI-R) or the Autism Diagnostic Observation Schedule (ADOS), Bölte and colleagues (2011) found that EF impairments in males were correlated with more RRBI. The findings suggested that RRBI are more

pronounced in males compared to females and that the observed association between EF and RBIs (stereotypic behaviours and interests) is stronger in the autistic males. Autistic females exhibited a better EF which was found to be associated with less RBIs. The authors state that the “identified association between EF and stereotypic behaviours and interests is indeed a ‘possible’ one” (Bölte, Duketis, Poustka, & Holtmann, 2011, pp. 507).

Lastly, it is important to point out here that one study found that the amount of change also differed according to different subtypes of the RBIs. Chowdhury and colleagues (2010) investigated age-related changes in RBIs in 34 high-functioning adults with ASDs at current age and retrospectively at age 4–5 years using the Autism Diagnostic Interview—Revised, and the Repetitive Behavior Scale—Revised (RBS-R). They found evidence of significant changes in all RBIs over time, the only exception being the Self-injurious Behavior subscale of the RBS-R. Chowdhury and colleagues, from childhood to adulthood, found a 75% improvement in compulsions, a 71% improvement in stereotypies and a 53.6% improvement in self-injurious behaviours. They found that about 44% exhibited improvement on the Restricted Behavior subscale (44.1%) (Chowdhury et al., 2010).

Neuroanatomical Findings

Only one study was identified which used not just clinical judgement, psychological and behavioural assessments, etc. but also neuroanatomical data. Supekar and Menon (2015) analysis of neuroanatomical data revealed, for the first time, that girls and boys with ASD differ in the organization of cortical and subcortical motor systems and that RRBI severity is associated with sex differences in gray matter (GM) morphometry in distinct motor systems (Supekar & Menon, 2015).

Genetic Liability

Another study reported findings which supported the hypothesis of a multiple threshold model of genetic liability of ASD with females having a higher liability for affectation status, at least on the repetitive behaviour dimension of ASD (Szatmari et al., 2012). Szatmari and colleagues’ (2012) sample included individuals with ASD (970 families, 2,028 individuals) who were recruited as part of the Autism Genome Project (AGP). They differentiated the families into families containing a female (either female-female or male-female) and those with only males. Szatmari and colleagues argued that if the sex with the lower prevalence is associated with a greater genetic liability necessary to cross sex-specific thresholds, the males from female containing families should be more severely

affected than males from male only families. Affected subjects from the different types of families with ASD were sampled and compared on the social reciprocity and repetitive behaviour scores from the Autism Diagnostic Interview-Revised (ADI-R). In general, females were found to have lower repetitive behaviour scores compared to males. Additionally, males from female containing families were found to have higher repetitive behaviour scores when compared to males from male-male families (Szatmari et al., 2012).

Discussion

This review highlighted the lack of consistency across the studies in terms of whether there are sex differences in RBIs in ASD. The review also highlights the relatively little research attention that has been given to this particular area with only 19 studies being identified. Out of the total of 19 articles, five found no significant evidence to support the notion of sex differences in RBIs in ASD (Solomon et al., 2012; Andersson et al., 2013; Harrop et al., 2015; Reinhardt et al., 2015; Knutsen et al., 2018). It is important to emphasise one of these studies here. Knutsen and colleagues (2018) study revealed more similarities than differences between males and females with ASD in the core diagnostic domain of RBIs based on clinical observations. One study did not report any differences in RBIs between males and females with ASD as they only had one female with ASD in their sample. As a result, analysis looking at the difference between males and females with ASD could not be carried out (Chowdhury, Benson, & Hillier, 2010).

Twelve studies identified in the present review found evidence that males with ASD had significantly more RBIs compared to females with ASD (Hartley & Sikora, 2009; Bölte et al., 2011; Hattier et al., 2011; Sipes et al., 2011; Mandy et al., 2012; Park et al., 2012; Szatmari et al., 2012; Frazier et al., 2014; Hiller et al., 2014; Wilson et al., 2016; Supekar & Menon, 2015; Dean et al., 2017). Lastly, one study found that girls with ASD have features of RBIs which are exhibited more compared to boys with ASD (Antezana et al., 2018). Specifically, they found that the items that were found to best-discriminate gender were greater stereotyped behaviours and restricted interests in the boys and compulsive, sameness, restricted, and self-injurious behaviour items in the girls (Antezana et al., 2018). Antezana and colleagues (2018) discuss some of the potential limitations with their study. For instance, the data used in the study was gathered from numerous studies (with few overlapping measures), therefore specific exclusion/inclusion criteria may impact the ability to generalise from the findings. For instance, IQ data was only available for a subset of the sample. A large age range was adopted in the study, and age differences in RBI may impact on the findings. As mentioned later, The Repetitive Behavior Scale—Revised (RBS-R, Bodfish et al., 2000) is a parent-

report measure. Therefore, it is subject to bias as a result of the different interpretations that can be made of items and scores assigned by each parent. For example, “pulling hair” could be interpreted a number of ways such as compulsive (i.e., trichotillomania), sensory-based or self-injurious. However, it is important to point out here that this is the first study of its kind as it investigated gender differences in RRBI using a *comprehensive* RRBI measure in children with ASD. Only two other studies identified in the present review used the Repetitive Behavior Scale-Revised (RBS-R) (Chowdhury et al., 2010; Solomon et al., 2012; Frazier et al., 2014).

The results from the study conducted by Knutsen and colleagues (2018) identified more RRBI similarities than differences between females and males. They had expected that RRBI domain differences would emerge among primary school-aged higher or lower functioning groups. However, this was not found. Such a finding would have been consistent with the findings from other studies (e.g., Szatmari et al., 2012; Supekar & Menon, 2015). The findings by Knutsen and colleagues are consistent with the findings from earlier studies which found similar RRBI domain scores on clinician-reported diagnostic measures between female and male children with ASD (Andersson et al., 2013; Harrop et al., 2015; Reinhardt et al., 2015).

Four of the five studies identified in the present review which found no significant differences in RRBI between males and females with ASD overall (Andersson et al., 2013; Harrop et al., 2015; Reinhardt et al., 2015; Knutsen et al., 2018) are inconsistent with the findings from a meta-analysis by Van Wijngaarden-Cremers and colleagues (2014). In their systematic review and meta-analysis of 22 peer reviewed original publications which investigated gender differences in the core triad of impairments in ASD, they observed lower rates of RRBI in females compared to males aged between 6 and 12 years of age (a similar age range to the samples in the four studies identified in this review which found no significant differences). However, as pointed out by Knutsen and colleagues (2018) in their paper, the meta-analysis by Wijngaarden-Cremers and colleagues (2014) included findings from both clinician (ADOS) and caregiver (ADI-R) report. Whereas the study by Knutsen and colleagues (2018) included a clinical sample which was only based on direct clinical observation (ADOS). Knutsen and colleagues acknowledge the inherent bias in their sample which only included children who were referred to an Autism Treatment Network (ATN) site. (The ATN registry is the first and one of the largest autism data registries in North America). Additionally, they highlight the potential issues surrounding direct clinician observation as opposed to relying on historical report (e.g., from the parents) which may be another potential contributory factor resulting in the discrepancies in findings in relation to whether there are differences in RRBI between males and females with ASD. They highlight that there may be differences between clinical

and caregiver perspectives of RRBIs (e.g., Le Couteur et al., 2008; Lemler, 2012; Ventola et al., 2006) and in exclusively female samples (Kopp et al., 2010).

The study by Reinhardt and colleagues (2015) found no sex differences in relation to RRBIs in males and females with ASD. However, they acknowledge a potential limitation with their study that may explain this. Their sample consisted of 511 children (288 of whom were diagnosed with ASD). However, only 54 females with ASD were included (Reinhardt et al., 2015). A larger sample may have resulted in a stronger significance level between the groups with respect to differences in the amount of RRBIs. Indeed, the issues of underpowered sample sizes is not unique to this particular study. Solomon and colleagues (2012) examined phenotypic differences between boys and girls based on a sample of 8-18 year-old autistic girls ($n = 20$) and boys ($n = 20$) and typically developing girls ($n = 19$) and boys ($n = 17$). Only marginally significant differences in the domain of RRBIs were reported in this study (Solomon et al., 2012). Such a limitation was emphasised by Mandy and colleagues (2012) (who did find sex differences in RRBIs in individuals with ASD. They argue that the reason for some of the studies not finding sex differences in RRBIs is likely to be due to their methodological characteristics as opposed to a type I error in their own study. They highlighted examples of earlier studies (published before 2008) where they suggest that their null findings may be due to group comparisons which lack sufficient statistical power in order to detect the moderately sized effects they found (e.g., Carter et al., 2007; Holtmann et al., 2007).

Another important consideration when looking at potential explanations for why some studies may report null findings are the possible differences in growth trajectories that may occur across the life course. Indeed, Harrop and colleagues (2015) have postulated that girls and boys may exhibit differential growth trajectories which change over the course of the lifespan (which was found in the study by Frazier and colleagues published in 2014). In their study they found a trend towards this. However, it was found to be statistically non-significant (which again, may be due to underpowered sample. Their sample comprised of only 29 girls with ASD and 29 boys with ASD) (Harrop et al., 2015). Although the study by Harrop and colleagues did not find any statistically significant sex difference in the young girls and boys on lower order RRBIs. However, these young boys and girls RBRI profile may change throughout the lifespan and gender differences may present in older, higher-functioning children with ASD. Differences in growth trajectories across the lifespan was found by Chowdhury and colleagues (2010) in individuals with ASD as a whole (both males and females). Moreover, Knutsen and colleagues (2018) found that younger higher functioning and older lower functioning females exhibited reduced rates on the Autism Diagnostic Observation Schedule restricted and repetitive behaviour subcategory *unusually repetitive/excessive, stereotyped behaviours* when compared to similar males. Such findings have obvious clinical and developmental

implications for females with ASD (Harrop et al., 2015). Interestingly, Hattier and colleagues (2011) found that, irrespective of age or gender, the frequency of RRBIs do not appear to change between young adulthood and later adulthood which is consistent with earlier findings by Gillberg and Steffenburg (1987) who argued that there is no discernible trajectory in ASD symptoms. Specifically, they argue that some ASD symptoms may plateau while some others may become more pronounced over time.

The suggestion that there is a higher liability threshold for expression of RRBIs in autistic females is important to consider given the studies discussed above showing, overall, lower levels of RRBIs in autistic females (particularly in high functioning females) compared to autistic males. As mentioned earlier, this may be one potential explanation for the very high sex ratios at the high end of the spectrum as RRBIs are considered to be a crucial behavioural symptom for the identification of ASD (Frazier, Georgiades, Bishop, & Hardan, 2014). If repetitive behaviour is used as critical diagnostic criteria, females with ASD will potentially not be identified by existing diagnostic assessments (Rynkiewicz, Schuller, Marchi, Piana, Camurri, Lassalle, & Baron-Cohen, 2016). Mandy and colleagues (2012) have detailed some prospective solutions to this. First, that there could be a lowering of the diagnostic threshold for clinical significance of RRBIs in females. Second, current RBRI scales/measurements could be modified so that they exclude the items which have been found to be sex-biased or, alternatively, creating sex-specific algorithms with differential item weighting. Or combining these two recommendations. Before any of these prospective solutions can be put in place, there needs to be much more research in order to further delineate sex differences in ASD (Mandy et al., 2012).

Interestingly, Frazier and colleagues have also highlighted that this raises the issues of whether high functioning females who do not exhibit restricted interests but have impairments in the social communication/interaction domain and the need for sameness would meet the diagnostic criteria for Social Communication (Pragmatic) Disorder or whether there needs to be a 'relaxation' of the DSM-5 criteria for ASD for females (Frazier et al., 2014). In current diagnostic assessments, symptoms exemplars which are specific to the female phenotype are not explicitly found. Frazier and colleagues (2014) have argued that behaviour exemplars which are specific to the female phenotype need to be included in commonly-used assessment tools. This may result in more females being correctly identified and diagnosed (Frazier et al., 2014). As pointed out by Solomon and colleagues (2012), sex-specific diagnostic criteria for neuropsychiatric disorders (such as ASD) would be more precise and clinically useful (see Hartung & Widiger, 1998).

Limitations

There are some potential limitations with the present systematic review. Primarily, there is the potential that relevant articles have not been identified in the search carried out on the databases. It is important to note that there are very few papers which have focused specifically on sex differences in RBRIs in ASD populations. Typically, studies focus on ASD symptoms more broadly and RBRIs is a subgroup analysis. This means that it is challenging to identify all studies which have included analysis of sex differences in RBRIs in populations with ASD because it was not the primary focus of their study and therefore this key wording is not included in the title of the paper for identification in databases searches, etc. However, in order to reduce the risk of this the 'Google Scholar' search was carried out in addition to the database search. All relevant papers were reviewed (including reference sections) for the purposes of identifying any potentially relevant articles which were not identified during the database searches. Every attempt was made to ensure that there were no inherent biases in the identification of papers for inclusion in this review.

Clinical Implications and Recommendations

Clinical considerations when assessing possible RBRIs and avoiding stereotyping

There is need for increased understanding, awareness and recognition of the female phenotype in terms of RBRIs (Wilson et al., 2016; Gould, 2017). The RBRIs exhibited in autistic females are not sufficiently captured by most currently used diagnostic instruments. Moreover, clinicians are less likely to identify the RBRIs in females as they tend not to be the typical repetitive behaviours commonly associated with ASD (Gould, 2017). Clinicians need to be cautious about potentially stereotyping observed behaviours. Identifying the typical types of RBRIs which can be found in both males and females (across the lifespan) is one step forward to address these identified issues (Wilson et al., 2016).

Kreiser and White (2014) recommend that clinicians consider the following questions when assessing a female for possible ASD: "Is there any negative impact on social, academic, or occupational activities as a result of engaging in the activity or interest?" and "What happens when the engagement in the activity or interest is interrupted or stopped?". Importantly, it is the *quality* and *intensity* of these activities or interests, in addition to the amount of time spent engaged with them that is important to consider as opposed to the special interests (Gould & Ashton-Smith, 2011). It is recommended that clinicians consider 'females as a whole' in terms of their clinical presentation

and look for any indication of RBRI, even repetitive interests which appear clinically innocuous. Additionally, so that symptoms can be accurately recognised, clinicians should be encouraged, if they do not do so already, to gain as much clinical experience as possible observing the male and female phenotypes of ASD (Halladay et al., 2015).

Future Research Directions

Firstly, it is worth pointing out that there is a need for future studies to include females with varying levels of 'severity' of ASD symptomology (Bargiela et al., 2016).

Exploring the gender differences on measures of RBRI in more detail

Autistic females tend to score lower on measures of RBRI compared to ASD males. However, there is a need for empirical research to explore whether this gender difference is due to actual differences in these traits or if females are scoring lower on the measures because they are 'simply' not captured by the measures (Van Wjingaarden-Cremers et al., 2014; Bargiela et al., 2016).

Investigating the clinical utility of the ASSQ-REV in female populations

As mentioned earlier, ASD screening tools have been developed (and therefore normed) based on the male phenotype which questions the validity of these tools for autistic females. In order to investigate this Kopp and Gillberg (2011) identified and evaluated 18 items which are thought to be sensitive to the female phenotype of ASD. These 18 items were integrated into the Autism Spectrum Screening Questionnaire (ASSQ, Ehlers, Gillberg, & Wing, 1999). This instrument was developed to screen for Asperger syndrome and high-functioning autism. Findings revealed that a number of items on the newly named ASSQ-Revised Extended Version exhibited a higher sensitivity to autistic females. Additionally, there were four questions on the ASSQ-REV which girls would typically provide an affirmative response to, namely, avoiding demands, difficulty completing daily activities due to repetitive behaviours, interacting mostly with younger children, or having a different voice or speech when compared to boys (Kopp & Gillberg, 2011; Haney, 2016). It would be useful to investigate how this tool captures female exemplars of RBRI across the lifespan – from childhood to adulthood.

Gould has already recommended that current diagnostic instruments and/or manuals need to be adapted to include symptom exemplars which capture the female phenotype of ASD – e.g., the types of RBRI exhibited in ASD females (Gould, 2017). Future studies could investigate the clinical

utility of the new screening tool 'The autism spectrum screening questionnaire-revised extended version (ASSQ-REV, Kopp & Gillberg, 2011) in how sensitive it is to female features of ASD (using samples of girls and women). In their sample, Kopp and Gillberg (2011) found that certain single ASSQ-GIRL items were much more typical of autistic girls compared to autistic boys. The single items which were most marked included: "avoids demands", "very determined", "careless with physical appearance and dress" and "interacts mostly with younger children".

Adapted Version of the Repetitive Behavior Questionnaire-2 (RBQ-2)

It would be useful for future research to investigate the RBQ-2 in females compared to males using measures specifically designed to investigate this behaviour. One measure that would be worth considering is the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007; Lidstone et al., 2014) which is a twenty-item questionnaire. The items are directly derived from a standardised clinical interview tool, the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002). Barrett and colleagues (2015) investigated an adapted version of the RBQ-2. They adapted it into an adult self-report questionnaire which they called the Adult RBQ-2 (RBQ-2A). The authors emphasise that, given that the RBQ-2A has been adapted into a self-report measure, it is only accessible to participants who have sufficient cognitive resources and verbal ability to enable them to complete the questionnaire (Barrett, Uljarević, Baker, Richdale, Jones, & Leekam, 2015). Their findings indicated that the RBQ-2A has utility as a self-report questionnaire measure of RBQ-2 for adults (Barrett et al., 2015).

Neurobiological Substrates of RBQ-2 in Autistic Females Compared to ASD Males

As pointed out by Van Wijngaarden-Cremers and colleagues (2014), the "male-skewed bias towards restricted interests and behaviors and stereotypes has not been precisely elucidated by biological theories. The underlying mechanisms are yet to be identified" (pp. 633). Future studies could investigate this using functional magnetic resonance imaging (fMRI) or other less expensive, relatively motion-tolerant and more transportable measures of neurobiological activity such as functional near-Infrared spectroscopy (fNIRS). fNIRS measures brain activity through the hemodynamic responses associated with neuron behaviour. There is increasing use of fNIRS in autism research (see Mazzone, Grove, Eapen, Lenroot, & Bruggemann, 2018). Additionally, Supekar and Menon (2015) recommend more research is needed to explore how the observed sex differences in neuroanatomy that they found in their study are associated with current ADI-R RBQ-2

scores, current ADI-R RRB subscales scores repetitive, sensory motor behaviours (RSM), insistence on sameness (IS) and circumscribed interests, and/or other measures of RRB including the Repetitive Behaviors Scale-Revised (RBS-R). In their study, Supekar and Menon (2015) studied volume (gray matter). They suggest that there is a need for further research to explore any sex differences in the cortical surface area and cortical thickness (which are the two components of volume) (Supekar & Menon, 2015).

*The role of neuropeptides in RBRI*s

Solomon and colleagues (2012) highlighted in their paper some of the studies which have linked differences in RBRI and variations in neuropeptides including oxytocin and vasopressin (e.g., Carter, 2007; Hollander et al., 2003; Insel, O'Brien, & Leckman, 1999). Moreover, there have been some small scale studies which have found that infusions of oxytocin reduce RBRI in adult autistic males (Hollander et al., 2003). More recently Yang and colleagues (2015) found that cortisol, serotonin and oxytocin may all have a contributory role in the presentation of RBRI in autistic individuals. Further research could investigate the role of these neuropeptides in RBRI in ASD (males and females) across the lifespan and investigate treatment implications for more severe cases (e.g., of particularly extreme self-injurious behaviour such as head banging).

Conclusions

In the present review only nineteen studies were identified which looked at sex differences in RBRI in males and females with ASD. Twelve studies found evidence that males with ASD had significantly more RBRI compared to females with ASD. This review highlighted the lack of consistency across the studies in terms of whether there are sex differences in RBRI in ASD with five of the nineteen studies finding no statistically significant sex differences. There is a real need to highlight the importance of understanding and recognising how RBRI can differ between males and females with ASD. This is important to address in future research as it is well-established that the earlier the diagnosis the better the outcomes due to the timely access to appropriate interventions (Begeer et al., 2013; Mademtzi, Singh, Shic, & Koenig, 2018).

Conflicts of Interest

The author(s) have no conflicts of interest to declare.

References

Adamou, M., Johnson, M., & Alty, B. (2018). Autism Diagnostic Observation Schedule (ADOS) scores in males and females diagnosed with Autism: a naturalistic study. *Advances in Autism*, (just-accepted), 00-00.

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4th ed. – text revision). Washington, DC: Author.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Andersson, G. W., Gillberg, C., & Miniscalco, C. (2013). Pre-school children with suspected autism spectrum disorders: do girls and boys have the same profiles?. *Research in Developmental Disabilities*, *34*(1), 413-422.

Antezana, L., Factor, R. S., Condy, E. E., Strege, M. V., Scarpa, A., & Richey, J. A. (2018). Gender differences in restricted and repetitive behaviors and interests in youth with autism. *Autism Research*.

Arnott, B., McConachie, H., Meins, E., Fernyhough, C., Le Couteur, A., Turner, M., ... & Leekam, S. (2010). The frequency of restricted and repetitive behaviors in a community sample of 15-month-old infants. *Journal of Developmental and Behavioral Pediatrics*, *31*(3), 223-229.

Attwood, T. (2006). *Asperger's and girls*. London: Jessica Kingsley Publications.

Baird, G., Douglas, H. R., & Murphy, M. S. (2011). Recognising and diagnosing autism in children and young people: summary of NICE guidance. *British Medical Journal*, *343*(d6360), 10-1136.

Bargiela, S., Steward, R., & Mandy, W. (2016). The experiences of late-diagnosed women with autism spectrum conditions: An investigation of the female autism phenotype. *Journal of Autism and Developmental Disorders*, *46*(10), 3281-3294.

Baron-Cohen, S. (2002). The extreme male brain theory of autism. *Trends in Cognitive Sciences*, *6*(6), 248-254.

- Barrett, S. L., Uljarević, M., Baker, E. K., Richdale, A. L., Jones, C. R., & Leekam, S. R. (2015). The Adult Repetitive Behaviours Questionnaire-2 (RBQ-2A): a self-report measure of restricted and repetitive behaviours. *Journal of Autism and Developmental Disorders*, *45*(11), 3680-3692.
- Begeer, S., Mandell, D., Wijnker-Holmes, B., Venderbosch, S., Rem, D., Stekelenburg, F., & Koot, H. M. (2013). Sex differences in the timing of identification among children and adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *43*(5), 1151-1156.
- Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., ... & Lord, C. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *43*(6), 1287-1297.
- Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders*, *30*, 237–243.
- Bölte, S., Duketis, E., Poustka, F., & Holtmann, M. (2011). Sex differences in cognitive domains and their clinical correlates in higher-functioning autism spectrum disorders. *Autism*, *15*(4), 497-511.
- Carter, A. S., Black, D. O., Tewani, S., Connolly, C. E., Kadlec, M. B., & Tager-Flusberg, H. (2007). Sex differences in toddlers with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *37*(1), 86-97.
- Carter, C. S. (2007). Sex differences in oxytocin and vasopressin: Implications for autism spectrum disorders? *Behavioural Brain Research*, *176*, 170–186.
- Chowdhury, M., Benson, B. A., & Hillier, A. (2010). Changes in restricted repetitive behaviors with age: A study of high-functioning adults with Autism Spectrum Disorders. *Research in Autism Spectrum Disorders*, *4*, 210–216.
- Dean, M., Harwood, R., & Kasari, C. (2017). The art of camouflage: Gender differences in the social behaviors of girls and boys with autism spectrum disorder. *Autism*, *21*(6), 678-689.
- Ehlers, S., Gillberg, C., & Wing, L. (1999). A screening questionnaire for Asperger syndrome and other high-functioning autism spectrum disorders in school age children. *Journal of Autism and Developmental Disorders*, *29*(2), 129-141.
- Fombonne, E. (2009). Epidemiology of pervasive developmental disorders. *Pediatric Research*, *65*(6), 591–598.

- Frazier, T. W., Georgiades, S., Bishop, S. L., & Hardan, A. Y. (2014). Behavioral and cognitive characteristics of females and males with autism in the Simons Simplex Collection. *Journal of the American Academy of Child and Adolescent Psychiatry, 53*(3), 329-340.
- Gillberg, C. (1993). Autism and related behaviours. *Journal of Intellectual Disability Research, 37*(4), 343-372.
- Gillberg, C., & Steffenburg, S. (1987). Outcome and prognostic factors in infantile autism and similar conditions: A population based study of 46 cases followed through puberty. *Journal of Autism and Developmental Disorders, 17*, 273–287.
- Gould, J. (2017). Towards understanding the under-recognition of girls and women on the autism spectrum. *Autism, 21*(6), 703-705.
- Gould, J., & Ashton-Smith, J. (2011). Missed diagnosis or misdiagnosis? Girls and women on the autism spectrum. *Good Autism Practice (GAP), 12*(1), 34-41.
- Halladay, A. K., Bishop, S., Constantino, J. N., Daniels, A. M., Koenig, K., Palmer, K., ... & Taylor, J. L. (2015). Sex and gender differences in autism spectrum disorder: summarizing evidence gaps and identifying emerging areas of priority. *Molecular Autism, 6*(1), 36.
- Hollander, E., Novotny, S., Hanratty, M., Yaffe, R., DeCaria, C. M., Aronowitz, B. R., et al. (2003). Oxytocin infusion reduces repetitive behaviors in adults with autism spectrum disorders. *Neuropsychopharmacology, 28*, 193–198.
- Haney, J. L. (2016). Autism, females, and the DSM-5: Gender bias in autism diagnosis. *Social Work in Mental Health, 14*(4), 396-407.
- Harrop, C., Gulsrud, A., & Kasari, C. (2015). Does gender moderate core deficits in ASD? An investigation into restricted and repetitive behaviors in girls and boys with ASD. *Journal of Autism and Developmental Disorders, 45*(11), 3644-3655.
- Hartley, S. L., & Sikora, D. M. (2009). Sex differences in autism spectrum disorder: an examination of developmental functioning, autistic symptoms, and coexisting behavior problems in toddlers. *Journal of Autism and Developmental Disorders, 39*(12), 1715-1722.
- Hartung, C. M., & Widiger, T. A. (1998). Gender differences in the diagnosis of mental disorders: conclusions and controversies of the DSM-IV. *Psychological Bulletin, 123*(3), 260–278.

Hattier, M. A., Matson, J. L., Tureck, K., & Horovitz, M. (2011). The effects of gender and age on repetitive and/or restricted behaviors and interests in adults with autism spectrum disorders and intellectual disability. *Research in Developmental Disabilities, 32*(6), 2346–2351.

Hiller, R. M., Young, R. L., & Weber, N. (2014). Sex differences in autism spectrum disorder based on DSM-5 criteria: evidence from clinician and teacher reporting. *Journal of Abnormal Child Psychology, 42*(8), 1381-1393.

Holtmann, M., Bölte, S., & Poustka, F. (2007). Autism spectrum disorders: Sex differences in autistic behaviour domains and coexisting psychopathology. *Developmental Medicine and Child Neurology, 49*(5), 361-366.

Honey, E., McConachie, H., Turner, M., & Rodgers, J. (2012). Validation of the repetitive behaviour questionnaire for use with children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 6*(1), 355-364.

Hull, L., Mandy, W., & Petrides, K. V. (2017). Behavioural and cognitive sex/gender differences in autism spectrum condition and typically developing males and females. *Autism, 21*(6), 706-727.

Hull, L., Petrides, K. V., Allison, C., Smith, P., Baron-Cohen, S., Lai, M. C., & Mandy, W. (2017). “Putting on my best normal”: social camouflaging in adults with autism spectrum conditions. *Journal of Autism and Developmental Disorders, 47*(8), 2519-2534.

Insel, T. R., O’Brien, D. J., & Leckman, J. F. (1999). Oxytocin, vasopressin, and autism: Is there a connection? *Biological Psychiatry, 45*, 145–157.

Kim, Y. S., Fombonne, E., Koh, Y. J., Kim, S. J., Cheon, K. A., & Leventhal, B. L. (2014). A comparison of DSM-IV pervasive developmental disorder and DSM-5 autism spectrum disorder prevalence in an epidemiologic sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 53*(5), 500-508.

Kim, Y. S., Leventhal, B. L., Koh, Y.-J., Fombonne, E., Laska, E., Lim, E.-C., et al. (2011). Prevalence of autism spectrum disorders in a total population sample. *The American Journal of Psychiatry, 168*(9), 904–912.

Kim, J., & Shin, M. (2005). A study of reliability & validity for the Korean version of Asperger Syndrome Diagnostic Scale. *Korean Journal of Child and Adolescent Psychiatry, 16*, 98–105.

Knutsen, J., Crossman, M., Perrin, J., Shui, A., & Kuhlthau, K. (2018). Sex differences in restricted repetitive behaviors and interests in children with autism spectrum disorder: An Autism Treatment Network study. *Autism, 1362361318786490*.

- Koenig, K., & Tsatsanis, K. D. (2005). Pervasive developmental disorders in girls. In: Bell DJ, Foster SL and Mash EJ (eds) *Handbook of Behavioral and Emotional Problems in Girls*. New York: Kluwer Academic/Plenum Publishers, pp.211–237.
- Kopp, S., & Gillberg, C. (2011). The Autism Spectrum Screening Questionnaire (ASSQ)-Revised Extended Version (ASSQ-REV): an instrument for better capturing the autism phenotype in girls? A preliminary study involving 191 clinical cases and community controls. *Research in Developmental Disabilities, 32*(6), 2875-2888.
- Kopp, S., Kelly, K. B., & Gillberg, C. (2010). Girls with social and/or attention deficits: a descriptive study of 100 clinic attenders. *Journal of Attention Disorders, 14*(2), 167–181.
- Kopp, S., & Gillberg, C. (1992). Girls with social deficits and learning problems: Autism, atypical Asperger syndrome or a variant of these conditions. *European Child and Adolescent Psychiatry, 1*(2), 89-99.
- Kreiser, N. L., & White, S. W. (2014). ASD in females: are we overstating the gender difference in diagnosis?. *Clinical Child and Family Psychology Review, 17*(1), 67-84.
- Lai, M. C., Lombardo, M. V., Ruigrok, A. N., Chakrabarti, B., Auyeung, B., Szatmari, P., ... & MRC AIMS Consortium. (2017). Quantifying and exploring camouflaging in men and women with autism. *Autism, 21*(6), 690-702.
- Lai, M.-C., Lombardo, M. V., Auyeung, B., Chakrabarti, B., & Baron-Cohen, S. (2015). Sex/gender differences and autism: setting the scene for future research. *Journal of the American Academy of Child and Adolescent Psychiatry, 54*(1), 11–24.
- Lai, M. C., Lombardo, M. V., Chakrabarti, B., & Baron-Cohen, S. (2013). Subgrouping the Autism "Spectrum": Reflections on DSM-5. *PLoS biology, 11*(4), e1001544.
- Lai, M. C., Lombardo, M. V., Suckling, J., Ruigrok, A. N., Chakrabarti, B., Ecker, C., ... & MRC AIMS Consortium. (2013). Biological sex affects the neurobiology of autism. *Brain, 136*(9), 2799-2815.
- Lai, M. C., Lombardo, M. V., Ruigrok, A. N., Chakrabarti, B., Wheelwright, S. J., Auyeung, B., ... & MRC AIMS Consortium. (2012). Cognition in males and females with autism: similarities and differences. *PLoS One, 7*(10), e47198.
- Lai, M. C., Lombardo, M. V., Pasco, G., Ruigrok, A. N., Wheelwright, S. J., Sadek, S. A., ... & MRC AIMS Consortium. (2011). A behavioral comparison of male and female adults with high functioning autism spectrum conditions. *PloS one, 6*(6), e20835.

- Le Couteur, A., Haden, G., Hammal, D., & McConachie, H. (2008). Diagnosing autism spectrum disorders in pre-school children using two standardised assessment instruments: the ADI-R and the ADOS. *Journal of Autism and Developmental Disorders*, 38(2), 362-372.
- Lemler, M. (2012). Discrepancy between parent report and clinician observation of symptoms in children with autism spectrum disorders. *Discussions*, 8(2), 1-3.
- Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: a review of research in the last decade. *Psychological Bulletin*, 137(4), 562.
- Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., ... & Couteur, A. L. (2007). Repetitive behaviours in typically developing 2-year-olds. *Journal of Child Psychology and Psychiatry*, 48(11), 1131-1138.
- Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P., ... & Moher, D. (2009). The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. *PLoS Medicine*, 6(7), e1000100.
- Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., et al. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, 8(2), 82–92.
- Mademtzi, M., Singh, P., Shic, F., & Koenig, K. (2018). Challenges of females with autism: A parental perspective. *Journal of Autism and Developmental Disorders*, 48(4), 1301-1310.
- Mandy, W., Chilvers, R., Chowdhury, U., Salter, G., Seigal, A., & Skuse, D. (2012). Sex differences in autism spectrum disorder: evidence from a large sample of children and adolescents. *Journal of Autism and Developmental Disorders*, 42(7), 1304-1313.
- Mazzoni, A., Grove, R., Eapen, V., Lenroot, R. K., & Bruggemann, J. (2018). The promise of functional near-infrared spectroscopy in autism research: What do we know and where do we go?. *Social Neuroscience*, 1-14.
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Annals of Internal Medicine*, 151(4), 264-269.
- Muthugovindan, D., & Singer, H. (2009). Motor stereotypy disorders. *Current Opinion in Neurology*, 22(2), 131-136.

- Nicholas, J. S., Charles, J. M., Carpenter, L. A., King, L. B., Jenner, W., & Spratt, E. G. (2008). Prevalence and characteristics of children with autism-spectrum disorders. *Annals of Epidemiology*, *18*, 130–136.
- Ozonoff, S., Macari, S., Young, G. S., Goldring, S., Thompson, M., & Rogers, S. J. (2008). Atypical object exploration at 12 months of age is associated with autism in a prospective sample. *Autism*, *12*(5), 457-472.
- Park, S., Cho, S.-C., Cho, I. H., Kim, B.-N., Kim, J.-W., Shin, M.-S., et al. (2012). Sex differences in children with autism spectrum disorders compared with their unaffected siblings and typically developing children. *Research in Autism Spectrum Disorders*, *6*(2), 861–870.
- Postorino, V., Fatta, L. M., De Peppo, L., Giovagnoli, G., Armando, M., Vicari, S., & Mazzone, L. (2015). Longitudinal comparison between male and female preschool children with autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *45*(7), 2046-2055.
- Reinhardt, V. P., Wetherby, A. M., Schatschneider, C., & Lord, C. (2015). Examination of sex differences in a large sample of young children with autism spectrum disorder and typical development. *Journal of Autism and Developmental Disorders*, *45*(3), 697-706.
- Rivet, T. T., & Matson, J. L. (2011). Review of gender differences in core symptomatology in autism spectrum disorders. *Research in Autism Spectrum Disorders*, *5*(3), 957-976.
- Rynkiewicz, A., Schuller, B., Marchi, E., Piana, S., Camurri, A., Lassalle, A., & Baron-Cohen, S. (2016). An investigation of the 'female camouflage effect' in autism using a computerized ADOS-2 and a test of sex/gender differences. *Molecular Autism*, *7*(1), 10.
- Sipes, M., Matson, J. L., Worley, J. A., & Kozlowski, A. M. (2011). Gender differences in symptoms of autism spectrum disorders in toddlers. *Research in Autism Spectrum Disorders*, *5*(4), 1465–1470.
- Solomon, M., Miller, M., Taylor, S. L., Hinshaw, S. P., & Carter, C. S. (2012). Autism symptoms and internalizing psychopathology in girls and boys with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, *42*(1), 48-59.
- South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders*, *35*(2), 145-158.
- Supekar, K., & Menon, V. (2015). Sex differences in structural organization of motor systems and their dissociable links with repetitive/restricted behaviors in children with autism. *Molecular Autism*, *6*(1), 50.

- Szatmari, P., Liu, X. Q., Goldberg, J., Zwaigenbaum, L., Paterson, A. D., Woodbury-Smith, M., ... & Thompson, A. (2012). Sex differences in repetitive stereotyped behaviors in autism: implications for genetic liability. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics*, *159*(1), 5-12.
- Szatmari, P., Chawarska, K., Dawson, G., Georgiades, S., Landa, R., Lord, C., ... & Halladay, A. (2016). Prospective longitudinal studies of infant siblings of children with autism: lessons learned and future directions. *Journal of the American Academy of Child and Adolescent Psychiatry*, *55*(3), 179-187.
- van Wijngaarden-Cremers, P. J., van Eeten, E., Groen, W. B., van Deurzen, P. A., Oosterling, I. J., & van der Gaag, R. J. (2014). Gender and age differences in the core triad of impairments in autism spectrum disorders: A systematic review and meta-analysis. *Journal of Autism and Developmental Disorders*, *44*(3), 627–635.
- Ventola, P. E., Kleinman, J., Pandey, J., Barton, M., Allen, S., Green, J., ... & Fein, D. (2006). Agreement among four diagnostic instruments for autism spectrum disorders in toddlers. *Journal of Autism and Developmental Disorders*, *36*(7), 839-847.
- Veselinova, C. (2014). Introductory awareness of autistic spectrum conditions. *Nursing and Residential Care*, *16*(1), 40-44.
- Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The diagnostic interview for social and communication disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry*, *43*, 307–325.
- Wilson, C. E., Murphy, C. M., McAlonan, G., Robertson, D. M., Spain, D., Hayward, H., ... & Zinkstok, J. (2016). Does sex influence the diagnostic evaluation of autism spectrum disorder in adults?. *Autism*, *20*(7), 808-819.
- Wolff, J. J., Botteron, K. N., Dager, S. R., Elison, J. T., Estes, A. M., Gu, H., ... & Zwaigenbaum, L. (2014). Longitudinal patterns of repetitive behavior in toddlers with autism. *Journal of Child Psychology and Psychiatry*, *55*(8), 945-953.
- Yang, C. J., Tan, H. P., Yang, F. Y., Wang, H. P., Liu, C. L., He, H. Z., ... & Du, Y. J. (2015). The cortisol, serotonin and oxytocin are associated with repetitive behavior in autism spectrum disorder. *Research in Autism Spectrum Disorders*, *18*, 12-20.

Zwaigenbaum, L., Bryson, S. E., Szatmari, P., Brian, J., Smith, I. M., Roberts, W., et al. (2012). Sex differences in children with autism spectrum disorder identified within a high-risk infant cohort. *Journal of Autism and Developmental Disorders*, 42(12), 2585–2596.

Table 1. Studies identified in the review which have investigated RBRI in females with ASD or the differences in RBRI between males and females with ASD (n = 19).

Authors	Sample Characteristics	Aims	Measures	Main Findings
<p>Andersson, Gillberg, & Miniscalco (2013)</p>	<p>20 girls (1.8–3.9 years of age) matched for chronological and developmental age with 20 boys with suspected ASD.</p> <p>Mean age 37 months (range 21–45 months).</p>	<p>To investigate whether very young girls and boys, identified at general population Child Health Care (CHC) screening of all children <3 years of age and referred for assessment with suspected ASD, have the same clinical, developmental, social and language profiles.</p>	<p><i>Diagnostic Process</i></p> <p>All children included in the study underwent the following assessments: (a) medical-neurological-psychiatric examination of the child; (b) child and family medical/psychiatric history taken from parent; (c) Griffiths’ Developmental Scales (Arvidsson & Köröndi, 2011), and, when appropriate, according to developmental age, the Wechsler Preschool and Primary Scale of Intelligence, third edition (WPPSI-III) (Wechsler, 2005); (d) Vineland Adaptive Behaviour Scales (VABS) (Sparrow, Balla, & Cicchetti, 1984); (e) MacArthur Communicative Development Inventory (Eriksson & Berglund,</p>	<p>No significant difference between the girls and the boys on RRB (ADOS) were found.</p>

			<p>2002; Fenson et al., 1994); (f) Reynell Developmental Language Scales III (RDLS) (Edwards et al., 1997); (g) Diagnostic Interview for Social and Communication disorders (DISCO-11) (Wing, Leekam, Libby, Gould, & Larcombe, 2002); (h) pre-school observation (if the child did not attend a pre-school, an observation of the child in the Home was carried out); (i) Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2000); (j) Children's Global Assessment Scale (CGAS) (Shaffer et al., 1983). All test results were then evaluated in relation to overall clinical judgement (Charman & Baird, 2002).</p> <p><i>Test instruments</i></p> <p>ADOS (Lord et al., 2000)</p> <p>ADOS Revision (ADOS-R) (Gotham, Risi, Pickles, & Lord,</p>	
--	--	--	--	--

			<p>2007).</p> <p>The Griffiths' Developmental Scales I and II (Alin-A° kerman & Nordberg, 1991)</p> <p>WPPSI-III (Wechsler, 2005)</p> <p>The VABS (Sparrow et al., 1984)</p> <p>Language comprehension was tested with the RDLS-III (Edwards et al., 1997).</p> <p>The CGAS (Schorre & Vandvik, 2004; Shaffer et al., 1983)</p>	
Antezana et al. (2018)	<p>615 individuals with ASD (507 boys; 82.4%), ages 3-18 years of age (M = 10.26, SD = 4.20).</p> <p>Intelligence Quotient</p>	<p>To investigate whether specific RRBI (i.e., stereotyped, self-injurious, compulsive, insistence on sameness, ritualistic, and restricted) can distinguish girls with</p>	<p><i>Cognitive Measures</i></p> <p>Repetitive Behavior Scale-Revised (RBS-R, Bodfish et al., 2000).</p>	<p>RBS-R items were found to significantly differentiate girls from boys with ASD.</p> <p>The study found no gender differences for total RBS-R symptom severity ($p > 0.67$). However, there were significant gender</p>

	<p>(IQ) data were available for 495 of the 615 participants (80.8%; M = 88.14, SD = 25.00).</p>	<p>ASD from boys with ASD.</p>	<p><i>ASD severity</i></p> <p>Each participant in National Database for Autism Research (NDAR) was given a clinical ASD severity score which was derived from all available diagnostic and adaptive assessments.</p>	<p>differences at the level of RBS-R items ($F(1,546) = 2.44, p < 0.001$).</p> <p>The items which were identified as being the best at distinguishing males and females with ASD were heightened stereotyped behaviours and restricted interests items in the boys and compulsive, sameness, restricted, and self-injurious behaviour items in the girls.</p> <p>Specifically, Bonferroni-corrected univariate ANOVAs showed that there were significant gender differences for 8 of the 43 RBS-R items. Namely, (item 11) "Pulls hair/skin" ($F(1,588) = 17.32, p < 0.001$), (43) "Fascination with movement of object" ($F(1,588) = 9.41, p < 0.01$), (3) "Hand/Finger" ($F(1,588) = 8.71, p < 0.01$), (20) "Hoarding/Saving" ($F(1,588) = 7.71, p < 0.01$), (12) "Rubs or scratches self" ($F(1,588) = 6.17, p = 0.01$), (5) "Object Usage" ($F(1,588) = 5.05, p = 0.03$), (33) "Insists on sitting at the same place" ($F(1,588) = 4.28, p < 0.05$), and (42) "Preoccupation with parts of an object"</p>
--	---	--------------------------------	--	---

				<p>(F(1,588) = 4.07, p < 0.05).</p> <p>Four other items showed a trend towards statistical significance (all Ps < 0.10). These included: (17) “Washing/Cleaning” (F(1,588) = 2.84), (25) Ritualistic “Self-Care—Bathroom/Dressing” (F(1,588) = 3.13), (34) “Dislikes changes in appearance/behavior” (F(1,588) = 2.93), (41) “Strongly attached to one object” (F(1,588) = 3.05).</p> <p>Strongly differentiating RBS-R items had greater success in correctly classifying boys (67.90%) compared to girls (61.00%).</p>
<p>Bölte, Duketis, Poustka, & Holtmann (2011)</p>	<p>35 males and 21 females with higher functioning ASD and unaffected sibling controls.</p> <p>Control sample comprised 23 males and 35 females.</p>	<p>To investigate sex differences in cognitive domains and their clinical correlates in higher-functioning ASD. To investigate this issue using a hypothesis-driven choice of attention to detail (ATTD) and executive function (EF)</p>	<p>The Autism Diagnostic Interview-Revised (ADI-R)</p> <p>Autism Diagnostic Observation Schedule (ADOS)</p> <p>The Child Behaviour Checklist</p>	<p>Findings showed that males exhibited more stereotyped behaviours and interests compared to the females on the ADOS (F(1, 54) = 5.6; p = .02; partial η^2 = .09).</p> <p>Stereotyped behaviours and interests on the ADI-R and ADOS (r = .45 and .42) (p < .01) were found to correlate moderately</p>

	<p>Females with ASD (n = 21) Mean age 14.3 (SD = 2.7).</p> <p>Males with ASD (n = 35) Mean age 14.0 (SD = 3.0)</p> <p>Females Siblings (n = 35) Mean age 14.8 (SD = 5.3)</p> <p>Males Siblings (n = 23) Mean age 14.4 (SD = 4.0).</p> <p>Exclusion criteria included mental retardation (IQ < 70).</p>	tests as well as gold standard clinical scales for ASD.	<p>(CBCL)</p> <p>Young Adult Behaviour Checklist (YABCL)</p> <p>The Wisconsin Card Sorting Test (WCST)</p> <p>Tower of Hanoi (ToH)</p> <p>Trailmaking Test (TMT)</p> <p>Embedded Figures Test (EFT)</p>	<p>(but robustly) with the performance on the TMT-B-A (r = .37 and .46) and ToH (moves) (r = .31 and .42) (p < .01) (however this correlation was not found with the WCST (r = .09 and .10)).</p> <p>Correlations were higher in males (r = .37 to .51) compared to females (r = .25 to .37) (p < .01).</p>
Chowdhury, Benson, & Hillier (2010)	34 (33 males, 1 female) high-functioning adults with ASDs at current age and retrospectively at age 4–5 years using the Autism Diagnostic	To examine Restricted Repetitive Behaviours (RRBs) symptom change for a sample of high-functioning adults with ASD.	<p>Autism Diagnostic Interview—Revised (ADI-R, Lord et al., 1994).</p> <p>The Repetitive Behavior Scale—Revised (RBS-R, Bodfish et al.,</p>	Findings showed significant changes in all RRBs over time. The only exception to this being of the Self-injurious Behavior subscale of the RBS-R.

	Interview—Revised, and the Repetitive Behavior Scale—Revised. Ages ranged from 19 to 28 years (mean age = 22.5, SD = 2.5). Nonverbal IQs ranged from 72 to 124 (mean nonverbal IQ = 98.8, SD = 15.7).		2000). A demographic form was used to collect information on the participant and the parent informant (e.g., the participant’s date of birth, gender, ethnicity, educational history, interventions received since age 4–5, psychiatric and medical diagnoses, and current work placement if any, informant’s age, relationship to participant, and highest level of education).	Specifically, from childhood to adulthood findings demonstrated a 75% improvement in compulsions, a 71% improvement in stereotypies and a 53.6% improvement in self-injurious behaviours. Approximately 44% exhibited improvement on the Restricted Behavior subscale (44.1%). Findings also revealed a low base rate for specific symptoms (such as self-injurious behavior, unusual preoccupations, and unusual sensory interests). Analysis not conducted on males and females separately. Only one female.
Dean, Harwood and Kasari (2017)	96 elementary school children during recess (ASD = 24 girls and 24 boys, typically developing = 24 girls and 24 boys). Children with ASD had average intelligence (IQ ≥ 70).	To investigate to what extent environmental factors such as gender-related social behaviours and activities play a role in helping girls with ASD to mask their symptoms. To investigate if girls with ASD are more effective at “camouflaging” their ASD	<i>Eligibility criteria</i> The ADOS (Lord et al., 2002) The Stanford–Binet Intelligence Scale: Fifth Edition (SB-5) <i>Primary outcome variables</i>	Findings showed that out of the groups only one boy in the ASD group was observed engaging in repetitive behaviour (n = 1) 18%.

		<p>symptoms and adopting compensatory behaviours in order to mitigate their social impairments.</p> <p>To investigate if the symptoms of ASD more obvious and easier to identify in boys.</p>	<p>The Playground Observation of Peer Engagement (POPE, Kasari et al., 2011, 2015)</p>	
<p>Frazier, Georgiades, Bishop, & Hardan (2014)</p>	<p>2,418 ASD-affected individuals (304 females, 2,114 males; age range = 4–18 years)</p>	<p>To investigate the differences in behavioural symptoms and cognitive functioning between males and females with ASD.</p>	<p><i>Core autism symptoms</i></p> <p>Diagnostic Interview–Revised (ADI-R)</p> <p>Autism Diagnostic Observation Schedule (ADOS)</p> <p>Social Responsiveness Scale (SRS, Constantino & Gruber, 2005).</p> <p>Repetitive Behavior Scale–</p>	<p>Findings showed that females with ASD had significantly lower repetitive behavior symptom levels on the ADI-R repetitive domain score and the RBS-R restricted interests subscale.</p> <p>RBS-R restricted interests survived false discovery rate correction within the repetitive behavior domain. This suggests that females with ASD are likely to exhibit fewer circumscribed interests.</p> <p>In females with ASD, lower levels of restricted interests were not found to be moderated by any demographic or clinical characteristic (all $p > .05$).</p>

			<p>Revised (RBS-R, Bodfish et al., 2000)</p> <p><i>Cognitive and motor</i></p> <p>Cognitive data included full scale intelligence quotient (FSIQ), verbal IQ, and nonverbal IQ derived from multiple instruments (Elliott, 1990; Wechsler, 1999, 2004).</p> <p>Peabody Picture Vocabulary Test–Fourth edition (PPVT, Dunn & Dunn, 2007) scaled scores from the non-word repetition subtest of the Comprehensive Test of Phonological Processing (CTOPP, Wagner, Torgesen, & Rashotte, 1999).</p> <p>Motor functioning was assessed using the total number of pegs completed using the dominant (Pegs Dominant) and nondominant (Pegs Non-</p>	<p>In females with ASD, lower restricted interests were found to be independent of reductions in IQ (standardised direct effect =-.065, standardised indirect effect < .001).</p>
--	--	--	---	--

			<p>Dominant) hands in the Grooved Pegboard test (Lezak, 1995).</p> <p>Caregiver reports of motor function were obtained using the fine motor, coordination during movement, general coordination, and composite scores from the Developmental Coordination Disorder Questionnaire (DCDQ, Wilson et al., 2009).</p> <p><i>Adaptive behavior and associated behavior problems</i></p> <p>Vineland Adaptive Behavior Scale—Second Edition (Sparrow et al., 2005).</p> <p>Child Behavior Checklist (CBCL, Achenbach & Rescorla, 2001).</p> <p>Aberrant Behavior Checklist (ABC, Aman et al., 1985a,1985b).</p>	
--	--	--	--	--

<p>Harrop, Gulsrud, & Kasari (2015)</p>	<p>29 girls with ASD were matched to 29 boys based on ASD severity.</p> <p>Chronological age (months) for girls = 38.81 (SD = 8.71)</p> <p>Chronological age (months) for boys = 35.83 (SD = 6.49)</p>	<p>To investigate potential differences in the overall frequency of observer coded RRBs between girls and boys with ASD.</p> <p>To examine potential differences in the categories of RRBs between girls and boys with ASD.</p> <p>To determine if the associations between developmental variables (non-verbal and verbal) and chronological age and RRBs differ between girls and boys with ASD.</p>	<p>Autism Diagnostic Observation Schedule (ADOS-2; Lord et al., 2012).</p> <p>Mullen Scales of Early Learning (MSEL; Mullen, 1995).</p> <p>A videotaped caregiver–child interaction (CCX) with a standardised set of toys.</p> <p><i>Ratings of Restricted and Repetitive Behaviours (RRBs)</i></p> <p>Coding of child RRBs was based on the scheme developed by Harrop and colleagues (2014).</p>	<p>Findings showed that while boys demonstrated higher frequencies of RRBs compared to girls within the CCX this difference was not significant [$F(1,56) = 2.37, p = .13, g2 = .04$].</p> <p>A trend toward higher frequencies of visual RRBs in boys was found [$F(1,56) = 3.89, p = .05, g2 = .06$]. While boys exhibited more object and sensory seeking RRBs, these differences were not significant [object: $F(1, 56) = 1.62, p = .21, g2 = .02$; sensory seeking: $F(1, 56) = 1.72; p = .19, g2 = .03$].</p> <p>The remaining categories of sensory aversion, motor/mannerisms and verbal were not significantly different between girls and boys.</p> <p>Girls and boys did not differ on ADOS-2 RRB algorithm scores in the whole sample ($n = 58$) or by individual study.</p> <p>The findings suggest that girls and boys</p>
--	--	--	--	--

				under five are more similar than dissimilar with respect to RRBs.
Hartley and Sikora (2009)	157 boys and 42 girls with ASD aged 1.5–3.9 years.	To investigate developmental profiles, autistic symptoms, and coexisting behaviour problems in boys and girls with ASD.	<p>Vineland Adaptive Behavior Scales, Second Edition, Survey Interview Form (Sparrow et al., 2005).</p> <p>Age, sex, ethnicity, and whether or not the child was receiving Early Intervention/Early Childhood Education Services were reported by Parents.</p> <p>The Mullen Scales of Early Learning (MSEL; Mullen, 1995)</p> <p>ADOS-G (Lord et al., 2000).</p> <p>A semi-structured diagnostic interview of the Diagnostic and Statistical Manual, Fourth</p>	Findings revealed that boys, when compared to girls, had a significantly higher ADOS-G Restricted/Repetitive/Stereotyped Interests, Behaviours or Activities score ($F(1, 261) = 5.60, p = .03, \text{partial } \eta^2 = .03$).

			<p>Edition (American Psychiatric Association 2000) criteria for Autistic Disorder was completed with parents.</p> <p>Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2000; Holtmann et al., 2007).</p>	
<p>Hattier, Matson, Tureck, & Horovitz (2011)</p>	<p>140 adults with ASDs and severe or profound intellectual disability (ID). (M = 49.28 years, SD = 11.22). All participants had previous diagnoses of severe to profound ID and an ASD.</p> <p>55% of participants were male and 45% were female.</p>	<p>To investigate the associations of gender and age on the frequency of RRBIs in a sample of adults with ASD and either severe or profound intellectual disability (ID).</p>	<p>Diagnostic Assessment for the Severely Handicapped-Second Edition (DASH-II; Matson, 1998).</p> <p>The DASH-II is an 84 item, informant-based measure used to screen for symptoms of psychopathology in adults with severe to profound ID. The scale is divided into 13 subscales: Impulse, Organic, Anxiety, Mood, Mania, ASD/Autism, Schizophrenia, Stereotypies, Self-Injurious Behavior, Elimination, Eating, Sleep, and Sexual.</p>	<p>Findings showed that scores on the stereotypies subscale were significantly lower for females (M = 2.079, SD = 2.623) compared to the males (M = 3.221, SD = 3.012).</p>

<p>Hiller, Young, & Weber (2014)</p>	<p>69 girls and 69 boys all diagnosed with high-functioning ASD.</p> <p>There was no significant difference</p> <p>in the age of the girls (M=8.06 years, SD=4.03) and boys (M=8.76 years, SD=3.91), $t(136)=1.03$, $p=0.31$, $d=0.17$</p>	<p>To investigate sex differences in the presentation of children and adolescents with ASD, based on both DSM-IV-TR and DSM-5 criteria.</p>	<p><i>Diagnostic process</i></p> <p>DSM-IV-TR (American Psychiatric Association, 2000) and DSM-5 (American Psychiatric Association, 2013).</p> <p>All assessments comprised a formal diagnostic interview with the child and parent, which followed a standard procedure, as per the clinic's protocol.</p> <p>Autism Detection in Early Childhood (ADEC; Young, 2007),</p> <p>Childhood Autism Rating Scale (CARS; Schopler et al., 1986)</p> <p>Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994)</p>	<p>The study found that girls exhibited with both less and <i>different</i> restricted interests.</p> <p><i>Sex Differences Based on DSM-IV-TR Criteria</i></p> <p>On the Repetitive/Restricted Interests domain, the only category which was found to significantly predict sex was the presence of restricted or fixated interests ($p < 0.001$). If a child did not meet this criterion, the predicted odds ratio showed the child was 10 times more likely to be a girl than boy.</p> <p>Routine adherence, stereotyped movement, and preoccupation with parts of objects all failed to significantly predict whether the child was a girl or a boy.</p> <p><i>Sex Differences Based on DSM-5 Criteria</i></p> <p>Restricted, Repetitive Behaviour Domain</p> <p>Differences were found in the stereotyped use of objects. Girls were found to be</p>
---	---	---	--	---

			<p>Autism Detection Observation Schedule (ADOS; Lord et al., 1989).</p> <p><i>IQ Information</i></p> <p>Standardised IQ data from the Wechsler Intelligence Scale for Children (Wechsler 2003), or the Wechsler Preschool and Primary Scale of Intelligence (Wechsler, 2002).</p> <p><i>Family History</i></p> <p>Information on family history of ASD was available for 61 girls and 57 boys.</p>	<p>substantially less likely to exhibit stereotyped use of objects (such as lining up or sorting behaviour). 27% of girls and 6% of the boys did not meet criterion for this impairment. 22% girls and 31 % of the boys somewhat met criterion. This indicates that the behaviour was present sometimes although infrequently. If the child did not meet this criterion the predicted odds ratio showed they were at least 8 times more likely to be a girl.</p> <p><i>Types of Restricted Interests</i></p> <p>Girls were most commonly rated as having restricted interests in the ‘seemingly random’ category (60 % girls, 29 % boys). Thus, being rated as having an apparent random restricted interest (e.g., rocks, stickers, pens) significantly predicted the child was a girl.</p> <p>The category which was found to most strongly predict that the child was a boy was fixations with screen time. Screen time fixations were mainly obsessive gaming (but it also included obsessions</p>
--	--	--	--	--

				<p>with iPads or other such screen technology). The results showed that 38% boys and only 9% of girls reportedly exhibited obsessional interests in screens.</p> <p>17% girls and 10% of the boys were reported to exhibit obsessional behaviour primarily around a specific program or character. 8 % of girls and 5 % of boys were reported to display an obsessional behaviour towards a toy.</p> <p>The authors split the sample up in younger (<7 years old) and older (>7 years old) group analysis which showed that the largest percentage of girls' restricted interests remained in the category of 'random' irrespective of age group.</p>
<p>Knutsen, Crossman, Perrin, Shui, & Kuhlthau (2018)</p>	<p>1024 children with ASD (512 female, 512 male; age range 2–<12 years)</p> <p>ADOS RRB domain score was described overall and by</p>	<p>To provide a comprehensive analysis of the ADOS RRB domain, focusing on the RRB subcategories among four individual groups of female and male children with ASD matched on age</p>	<p>ADOS-2 (Lord et al., 2012).</p> <p>Cognitive data for the majority of participants are available in the form of an overall composite IQ score derived</p>	<p>The results from this study showed that there were no sex differences on the ADOS RRB domain score across the full sample and for each of the stratified age/IQ groups.</p> <p>Interestingly, the study found that</p>

	<p>gender, and subcategory item scores were described by gender only in the following 4 individual age- and IQ-matched groups: (1) $IQ \geq 70$ and 2–<6 years, (2) $IQ \geq 70$ and 6–<12 years, (3) $IQ < 70$ and 2–<6 years, and (4) $IQ < 70$ and 6–<12 years. The two groups with ID (<70) are considered lower functioning and the two without ID (≥ 70) are considered higher functioning (Volkmar et al., 2004).</p>	<p>and IQ.</p>	<p>from two well-known standardised IQ instruments: the Stanford–Binet Edition, Fifth Edition (Roid, 2003); and Mullen Scales of Early Learning (Mullen, 1995).</p> <p>IQ for a minority of participants are derived from the following standardised IQ measures: Differential Ability Scales, Second Edition (DAS-II; Elliott, 1990); Wechsler Intelligence Scales (Third, Fourth, and Preschool Eds; Wechsler, 1991, 2002, 2003); and Bayley Scales of Infant Development (Bayley, 1993).</p>	<p>younger higher functioning and older lower functioning females presented reduced rates on the Autism Diagnostic Observation Schedule restricted and repetitive behavior subcategory unusually repetitive/excessive, stereotyped behaviours compared to similar males.</p> <p>Findings showed differences for the unusually repetitive interest or stereotyped behaviours subcategory in two groups: (1) younger higher functioning females ($IQ \geq 70$) had less (about half the odds) repetitive interests/behaviours than males (McNemar $S = 4.17$, odds ratio (OR) = 0.45, $p = 0.04$); and (2) older lower functioning females ($IQ < 70$) also had less (about 70% less odds) repetitive interests/behavior when compared to males (McNemar $S = 4.57$, OR = 0.27, $p = 0.03$).</p> <p>No differences were found for stereotyped/idiosyncratic use of words or phrases, unusual sensory interest in play material/person, or hand and finger and other complex mannerisms.</p>
--	---	----------------	---	--

<p>Mandy, Chivers, Chowdhury, Salter, Seigal, & Skuse (2012)</p>	<p>52 girls and 273 boys (Age range = 3–18 years) who consecutively received an ASD diagnosis at a clinic for assessing high-functioning ASD (mean verbal IQ = 92.6).</p> <p>Females: mean age: 10.2 (SD = 3.5)</p> <p>Males: mean age: 9.7 (SD = 3.1)</p>	<p>To investigate the female ASD phenotype amongst predominantly high-functioning children and adolescents.</p>	<p>The Developmental, Dimensional and Diagnostic Interview (3Di, Skuse et al., 2004).</p> <p>The Autism Diagnostic Observation Schedule (ADOS)</p> <p>The ADOS (Lord et al., 2000).</p> <p>The Strengths and Difficulties Questionnaire (SDQ) The SDQ comprises 25 items in 5 subscales: conduct problems, emotional problems, hyperactivity, peer problems and prosocial behaviour (Goodman, 1997).</p> <p>Intelligence Quotient (IQ)</p> <p>IQ data were collected as part of clinical assessment over the time frame of the study. A range of measures were used including: the British Picture Vocabulary Scale (Dunn et al., 1982), the</p>	<p><i>Parent Report</i></p> <p>Boys were found to have more RSBs compared to girls based on the 3Di. The frequency with which individual RSBs were reported by parents of males and females was also examined. Boys were found to be more likely to ‘have a large store of factual information’ ($p < .006$) and to exhibit ‘oddly formal play’ ($p < .026$) that involved systematically lining up toys when compared to girls.</p> <p><i>Direct Observation Using the ADOS</i></p> <p>As with parent report, the groups differed on the ADOS RSB score. The males were found to exhibit greater impairment when compared to the females. No age-by-gender interactions for the ADOS data was found.</p> <p>Repetitive and stereotyped behaviour</p>
---	--	---	--	---

			Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999) and the Wechsler Intelligence Scale for Children—Third (Wechsler et al., 1991) and Fourth Editions (Wechsler, 2003).	ADOS Females: Mean: 14.44 (SD = 17.31) Males: Mean: 22.69 (SD = 22.14) .42 t = 2.06, p = .04.
Park et al. (2012)	<p>ASD group comprised age- and IQ-matched boys (n = 91) and girls (n = 20) diagnosed with ASD by a child psychiatrist based on DSM-IV-R criteria.</p> <p>Group of unaffected siblings comprised age- and IQ-matched male siblings (n = 47) and female siblings (n = 51).</p> <p>Group of typically developing (TD) children comprised age- and IQ-matched TD boys (n =</p>	<p>To investigate the nature of cognitive and behavioural sex differences in children with ASD and two comparison groups: a group of TD children and a group of unaffected siblings of ASD children.</p>	<p><i>Diagnosis and core autistic symptoms</i></p> <p>The Korean versions of the Social Communication Questionnaires (SCQ) (Yoo, 2008) and the Asperger Syndrome Diagnostic Scale (ASDS) (Kim & Shin, 2005).</p> <p><i>Intelligence</i></p> <p>For intellectual ability, all participants were assessed using the Korean version of the Leiter International Performance Scale (Roid, Miller, & Leiter, 1997; Shin & Cho, 2009).</p>	<p>The findings indicate that, compared to females with ASD, the males with ASD exhibited significantly higher scores on the repetitive stereotyped behaviour (RSB) domain of the ADI-R (t = 2.03, p = 0.045).</p> <p>Also, findings suggested that male siblings exhibited significantly higher scores on the RSB domains of ADI-R (t = 4.17, p < 0.001) when compared to female siblings.</p>

	<p>26) and TD girls (n = 25).</p> <p>Mean age of the participants was 8.49 (SD = 2.89, age range = 4 to 15 years), and their mean IQ levels were average (ASD children) or above-average (unaffected siblings and TD children).</p>		<p><i>Co-occurring psychopathology</i></p> <p>The Korean version of the Child Behavior Checklist (K-CBCL) (Oh & Lee, 1990).</p> <p><i>Cognitive style</i></p> <p>The parents of participants completed 3 questionnaires measuring aspects of their children's cognitive style, preferences, and traits. The children's version of the Autism Spectrum Quotient (AQ-C, Auyeung et al., 2008).</p> <p>The children's version of the Empathy Quotient (EQ-C, (Auyeung et al., 2009).</p>	
--	---	--	---	--

			<p>The children’s version of the Systemizing Quotient (SQ-C, Auyeung et al., 2009).</p> <p>The Korean versions of the AQ-C, EQ-C, and SQ-C were translated and validated by Ghim and colleagues (2011).</p>	
<p>Reinhardt, Wetherby, Schatschneider, & Lord (2015)</p>	<p><i>ASD Group</i></p> <p>288 participants (54 female)</p> <p><i>Typically developing group (TD)</i></p> <p>59 females and 164 males.</p>	<p>To investigate sex differences in early social communication and developmental functioning in children with ASD and TD and adaptive behavior and autism symptoms in children with ASD.</p>	<p>Communication and Symbolic Behavior Scales Developmental Profile (CSBS) (Wetherby, Allen, Cleary, Kublin & Goldstein, 2002; Wetherby & Prizant, 2002).</p> <p>Mullen Scales of Early Learning (MSEL)</p> <p>Vineland Adaptive Behavior Scales, Second Edition (VABS)</p> <p>Autism Diagnostic Observation Schedule—Modules 1, 2, 3 or the Toddler Module (ADOS-T; Luyster</p>	<p>Males and females with ASD were compared on the ADOS domain scores (Restricted and Repetitive Behaviours). Findings revealed no significant sex differences. The effect size was small 0.15.</p>

			et al., 2009).	
Sipes et al. (2011)	<p>390 caregivers of infants and toddlers aged between 17 to 36 months who were enrolled in an early intervention program funded by the State of Louisiana.</p> <p>Mean age of the sample was 26.09 months (SD = 4.65), and 75% were males.</p> <p>Mean developmental quotient (DQ) for the whole sample was 74.6 (SD = 14.13).</p> <p>4 groups were made based on gender and DQ level. Using BDI-2 scores, participants were classified as</p>	<p>To investigate gender differences in symptom endorsements of ASD.</p>	<p>Battelle Developmental Inventory, Second Edition (BDI-2; Newborg, 2005)</p> <p>The Modified Checklist for Autism in Toddlers (M-CHAT; Charman et al., 2001; Robins, Fein, Barton, & Green, 2001).</p> <p>Criteria from the DSM-IV-TR (APA, 2000)</p> <p>Clinical judgment</p> <p>Baby and Infant Screen for Children with Autism Traits-Part 1 (BISCUIT-Part 1; Matson, Boisjoli, & Wilkins, 2007).</p>	<p>The results found gender differences in regard to the restricted interests and repetitive behaviour domain.</p> <p>Females with an average DQ were found to have significantly fewer endorsements on items related to restrictive and repetitive behaviours (RRBs).</p> <p>Groups significantly differed on the Repetitive Behavior/Interest subscale (of the BISCUIT-Part 1.) $F(3, 385) = 5.96, p < .001$.</p> <p>Also, on the third domain of the BISCUIT-Part 1. which assesses for RRBs, only females with average DQ were found to differ significantly from the other groups. Females with average DQ were found to endorse significantly fewer items related to RRBs compared all other groups. This would suggest the existence of a potential gender and DQ effect.</p>

	average or low DQ groups. Low DQ was defined as being more than one standard deviation below the mean. The 4 groups: (1) males with average DQ, (2) males with low DQ, (3) females with average DQ, and (4) females with low DQ.			
Solomon, Miller, Taylor, Hinshaw, & Carter (2012)	8-18 year-old girls (n = 20) and boys (n = 20) with ASD and typically developing (TD) girls (n = 19) and boys (n = 17).	<p>To investigate whether the clinically-referred high-functioning sample of boys and girls differed in ASD symptoms based on independent assessments of language, social, and repetitive behaviour symptoms that were not used when making the diagnosis of ASD.</p> <p>To investigate whether girls with ASD were at greater risk for</p>	<p>Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler 1999).</p> <p>Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al. 2000).</p> <p>Social Communication Questionnaire (SCQ; Rutter et al. 2003).</p> <p>Social Responsiveness Scale (SRS; Constantino 2002),</p>	<p>There was a main effect of group on all RBS-R scores: Stereotyped behavior, $\eta^2 = 38.48$; self-injurious behavior, $\eta^2 = 25.80$; compulsive behaviour, $\eta^2 = 32.25$; ritualistic behaviour, $\eta^2 = 39.65$; sameness behavior, $\eta^2 = 45.93$; restricted interests, $\eta^2 = 43.34$; and overall scores, $\eta^2 = 43.40$, $df = 3$, $N = 66$ for all.</p> <p>Follow-up comparisons using an adjusted alpha level of .0018 (.0071/4) revealed that boys and girls with ASD did not differ on any subscale, although results suggestive of higher scores in boys with ASD on the restricted interests subscale, $U = 77.50$, $z = -2.43$, $p = .015$ without such</p>

		internalising problems when compared to TD girls and boys with ASD.	<p>Children’s Communication Checklist-2nd Edition (CCC-2; Bishop, 2003)</p> <p>Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al., 1999).</p> <p>Behavior Assessment System for Children-2nd Edition (BASC2 subscales: Reynolds & Kamphaus, 2004)</p> <p>Children’s Depression Inventory (CDI; Kovacs, 1992).</p>	stringent corrections for multiple comparisons. Girls with ASD had higher scores than TD girls on all subscales (ps<.001) with the exception of the compulsive behaviour subscale. Boys with ASD had higher scores than TD boys (ps = .001). TD girls and TD boys did not differ.
Supekar & Menon (2015)	<p>25 females with ASD (mean age: 10.3 years) and</p> <p>25 males with ASD (mean age: 10.2 years) as well as 19</p> <p>TD females (mean age: 10.2 years) and 19 TD</p>	<p>To explore sex differences</p> <p>in the three core impairments that characterise</p> <p>childhood ASD.</p>	<p>Autism Diagnostic Interview, Revised (ADI-R).</p> <p>Voxel-based morphometry</p> <p>Brain morphometry was assessed using the optimized</p>	<p>This study found that girls with ASD, when compared to boys with ASD, exhibited less severe RRBs.</p> <p>Findings from the neuroanatomical data, showed that gray matter (GM) in the motor cortex, SMA, and crus 1 subdivision of the cerebellum was correlated with</p>

	<p>males (mean age: 10.3 years).</p> <p>Leveraging NDAR and ABIDE, two open-access largescale databases</p>	<p>To investigate whether structural brain organization is different in girls and boys with ASD.</p>	<p>voxel-based morphometry (VBM) method [38] performed with the VBM5 toolbox (http://dbm.neuro.uni-jena.de/vbm).</p>	<p>RRB in girls.</p> <p>GM in the right putamen—the region that discriminated TD girls and boys—was correlated with RRB in boys.</p> <p>In the NDAR dataset, girls with ASD showed less severe RRB, as measured by the ADI-R ($p < 0.01$, $t(740) = -5.19$). Girls with ASD were distinguishable from boys with ASD on the basis of their ADI-R domain scores with an accuracy of 94 %. The ADI-R RRB domain score was found to be the most significant feature that discriminated the two groups.</p> <p>In the ABIDE dataset, girls with ASD showed less severe repetitive/restricted behaviours, as measured by scores on the RRB domain of the ADI-R ($p < 0.01$, $t(45) = -2.78$). Girls with ASD could be distinguished from boys with ASD on the basis of their ADI-R domain scores with an accuracy of 89 %. The ADI-R RRB domain score was found to be the most significant feature that discriminated the two groups.</p>
--	---	--	---	---

				<p><i>Multivariate support vector regression analysis: relationship</i></p> <p>GM volume in the motor cortex, SMA, and crus 1 subdivision of the cerebellum were found to be correlated with scores on the RRB domain of the ADI-R in girls with ASD ($p < 0.05$). However, this relationship was not found in boys or for the social and communication domains in either girls or boys (all p's > 0.48).</p> <p>GM volume in the right putamen was correlated with scores on the RRB domain of the ADI-R ($p < 0.05$). No such relationship was observed in girls or for the social and communication domains in either boys or girls (all p's > 0.64).</p>
Szatmari et al. (2012)	Individuals with ASD (970 families, 2,028 individuals).	To investigate whether the sex differences in severity of quantitative traits seen in ASD are familial. Specifically, are	Autism Diagnostic Interview-Revised (ADI-R, Lord et al., 1994). Different measures of IQ used	Findings showed that, in general, females had lower repetitive behaviour scores when compared to the males.

	<p>The families were differentiated into families containing a female (either female-female or male-female) and those with only males.</p>	<p>they related to differences in genetic liability.</p>	<p>across sites.</p>	<p>Males from female containing families had higher repetitive behaviour scores than males from male–male families.</p> <p>The researchers investigated individual items which comprised the repetitive behaviours (BEH) domain and found that it was mainly items measuring the higher order “insistence on sameness” factor, as opposed to the lower order “sensory motor” behaviours, that resulted in these overall sex differences in mean scores. For example, females had lower scores on unusual preoccupations (P<0.001), circumscribed interests (P=0.002), repetitive use of objects or interest in parts of objects (P=0.03), and the “encompassing preoccupation or circumscribed pattern of interest” subdomain total score (P<.001) (which is the sum of items “unusual preoccupation” and “circumscribed interests”).</p>
<p>Wilson et al. (2016)</p>	<p>1244 adults (935 males and 309 females).</p>	<p>To examine whether sex influenced the diagnostic evaluation of ASD in a</p>	<p><i>Clinical assessment</i></p>	<p><i>Sex differences in core-symptom profiles in</i></p>

	<p>Age range = 18–75 years (inter-quartile range of 22–39 years).</p>	<p>sample of individuals who were referred to a national specialist clinic for an ASD assessment for the first time in adulthood.</p>	<p>Detailed neuropsychiatric assessment by a multidisciplinary clinical team with expertise in ASD: a consultant psychiatrist, +/- junior doctor and a research-reliable ADI-R/ADOS-G administrator.</p> <p>Additional mental health conditions were diagnosed in accordance with the ICD-10R (with the exception of adult attention deficit hyperactivity disorder (ADHD)) which, in keeping with UK guidelines, was assessed using Diagnostic and Statistical Manual of Mental Disorders (4th ed., text rev.; DSM-IV-TR).</p> <p>Neuropsychological testing was completed in 319 participants either for their clinical care if intellectual disability or a significant lacuna in cognitive function was suspected (248</p>	<p><i>high-functioning ASD (N = 827)</i></p> <p>The results showed that males had significantly more repetitive behaviours/restricted interests (based on the repetitive behaviours and restricted interests domain of the ADI-R), $t(526) = 3.27, p = 0.001, d = 0.33$.</p> <p><i>Interactions between sex, diagnostic subtype and core-symptoms</i></p> <p>On average, findings showed that the full-ASD participants scored significantly higher compared to the partial-ASD participants in all of the domains of the ADI-R (all $ps < 0.001$). Interestingly, the effect of sex was only significant for the repetitive behaviours and restricted interests domain (male > female; $F(1) = 7.62, p = 0.006$). The average male score was significantly higher compared to the average female score in all ASD subtypes in the repetitive behaviours and restricted interests domain.</p>
--	---	---	--	--

			<p>participants completed the Wechsler Adult Intelligence Scale-III (WAIS-III; Wechsler, 1997)) or as part of associated research projects (71 participants completed the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999)).</p>	<p><i>Asperger syndrome versus childhood/high-functioning autism.</i></p> <p>Based on the ADOS-G, a significant effect of subtype only in the repetitive behaviours/restricted interests domain was found (Asperger > childhood/high-functioning autism; $F(1) = 6.26, p = 0.01$).</p>
--	--	--	---	--