REVIEW PAPER

The Early Childhood Signs of Autism in Females: a Systematic Review

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Abstract
Compared to males, females are at a much greater likelihood of receiving a later diagnosis of autism, which impacts their opportunity to receive early support. To assist with early identification, this systematic literature review aimed to ascertain whether females differ from males in the early childhood signs of autism. The small number of heterogeneous studies made it difficult to draw conclusions, although it appears that females and males under 6 years of age are more similar than different in terms of their expression of autistic behaviours. Given the discrepant sex/gender ratio in autism, these findings highlight the importance of exploring whether there are different and/or specific, not yet identified, early signs of autism in females and males.

Keywords Autism · Gender differences · Sex differences · Female · Early signs

Autism spectrum disorder (ASD; referred to as 'autism' hereafter) is a heterogeneous neurodevelopmental disorder characterised by the early developmental onset of persistent differences and/or difficulties in social communication and interaction, and the presence of restricted, repetitive patterns of behaviour and/or interests (American Psychiatric Association, 2022). The prevalence of autism is estimated to be one in 44 children (Maenner et al., 2021), with approximately four males to every female receiving a diagnosis of autism (Fombonne, 2009; Maenner et al., 2021). When synthesising population-based/non-referred samples, however, a recent meta-analysis found the male-to-female ratio was closer to three males to every female (Loomes et al., 2017). Such findings suggest that a diagnostic sex/gender[1], bias exists,

resulting in some autistic females[2] going undiagnosed (Dworzynski et al., 2012), misdiagnosed (Kentrou et al., 2019; Kirkovski et al., 2013), or diagnosed later in life compared to males (Begeer et al., 2013; Giarelli et al., 2010; Rivet & Matson, 2011).

Several theories have been proposed to better understand the nature of this diagnostic sex/gender bias. Due to the underrepresentation of females in both research and clinical practice, one hypothesis is that our conceptualisation of autism and the development of diagnostic criteria and assessment tools is based upon the 'male' presentation (Kopp & Gillberg, 1992; Kreiser & White, 2014). Additionally, many females are not referred for autism assessments until later in life, as their differences may not be attributed to autism due to it being recognised as a 'male' condition (Bargiela et al., 2016; Whitlock et al., 2020). A second hypothesis is the 'female protective effect' which proposes that autistic females are genetically shielded against autism (Robinson et al., 2013). There is also emerging evidence to suggest that some females present with a different phenotype of autism to males (Hull et al., 2020; Kirkovski et al., 2013; Wilson et al., 2016). The female autism phenotype is thought to be characterised by an altered expression of the core traits of autism, as well as possible additional characteristics that are not currently recognised by

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[1] The term "sex/gender" is used when discussing the differences between females and males, as the individual effects of biological sex and social gender constructs on the presentation of autism have not yet been determined (Lai et al., 2015).

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[2] In keeping with the preferred language of those within the autism community, "identity-first" language (i.e., "autistic person") rather than "person-first" language (i.e., "person with autism") is used throughout this review (Bury et al., 2020; Kenny et al., 2016).

diagnostic criteria (Dworzynski et al., 2012; Hull et al., 2017, 2020; Kirkovski et al., 2013; Kopp & Gillberg, 2011; Kreiser & White, 2014; Lai et al., 2015; Rivet & Matson, 2011; Van Wijngaarden-Cremers et al., 2014). Although no conclusive framework has yet been derived, all proposed theories may contribute to this diagnostic sex/gender bias, delaying early detection in many females and their access to early supports and services, and consequently hindering their development, mental health, and well-being (Pellicano et al., 2014).

To improve early detection of autism in females, understanding sex/gender differences is important. Signs of autism are said to largely manifest within the first 2 years of a child's life (Fombonne, 2009; Rogers & Dilalla, 1990; Werner et al., 2000); however, evidence for sex/gender differences at a young age is conflicting. When investigating social communication and interactions (SCI), several studies have found that females have greater difficulties than males (Hartley & Sikora, 2009; Lawson et al., 2018; Rahaman et al., 2021; Ros-Demarize et al., 2020; Wang et al., 2017). In contrast, one study found that autistic females have less social communication difficulties (as per lower social affect scores) than males (Craig et al., 2020), while others found no sex/gender differences (Andersson et al., 2013; Carter et al., 2007; Reinhardt et al., 2015; Van Wijngaarden-Cremers et al., 2014). Additionally, some studies have reported fewer restricted and repetitive behaviours and/or interests (RRBIs) in autistic females compared with males (Hartley & Sikora, 2009; Sipes et al., 2011; Wang et al., 2017), whereas others found no sex/gender differences in this domain (Carter et al., 2007; Craig et al., 2020; Fulceri et al., 2016; Reinhardt et al., 2015; Siracusano et al., 2021).

The cause of inconsistencies across the sex/gender literature is complex and currently unknown, due to both similarities (e.g., measurement tools) and differences (e.g., heterogeneous sample characteristics) across studies. It has recently been suggested that a qualitative approach may help provide insight, as it can be more sensitive to capturing how females may present their autistic characteristics than a quantitative approach using overall domain/algorithm scores on standard measures (Lai & Szatmari, 2020; Lai et al., 2015). Kopp and Gillberg (2011) introduced the importance of qualitative research via item level analysis when they found that sex/gender differences were only evident after exploring individual behaviours of autism (rather than total scores). Since then, numerous studies have found significant differences between females and males, with respect to narrow constructs (i.e., subdomains of broad diagnostic domains) and/or individual behaviours of SCI (see Wood-Downie et al., 2020 for a review), and RRBIs (Antezana et al., 2019; Ben-Sasson et al., 2019). Generalising to autistic individuals across their lifespan, these results enhance our understanding of where the exact sex/gender differences in autistic traits may lie.

Focusing exclusively on individual signs of autism that emerge early in life, current research has identified a number of behaviours specific to autism across sex/gender. For example, compared to typically developing (TD) peers, young autistic children have been found to display reduced eye contact, response to name, and social smile, as well as more repetitive whole-body movements and behaviours with objects (e.g., spinning, lining up, and visually exploring; see Tanner & Dounavi, 2021; Zwaigenbaum et al., 2015 for reviews). To the best of our knowledge, however, no study to date has synthesised what is currently known about the sex/gender differences of such behaviours, including those pertaining to SCI, RRBIs, and additional behaviours. Therefore, this paper aimed to determine whether females differ from males in the early childhood signs of autism, by reviewing studies that investigated sex/gender differences in behaviours of autism emerging before the age of six.

Methods

This systematic review was conducted following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (Moher et al., 2009) and was registered with PROSPERO (CRD42021273521).

Search Strategy

Three electronic databases (PsycINFO, MEDLINE, and CINAHL) were systematically searched on 4 July 2021. Identical search terms were used across all databases that related to (1) autism, (2) young children, (3) females, and (4) early signs (see Table 1 for complete search strategy). Searches were limited to titles and abstracts, as well as studies published in English and containing human subjects. The reference lists of eligible articles were also hand-searched to identify any further relevant articles.

Eligibility Criteria

To be included in the review, studies were required to meet the following criteria: (1) female participants with a diagnosis of autism and a mean age below 6 years. Retrospective studies, including parents reporting on their child's behaviours between the ages of 0–5, were also included; (2) analysis/investigation of individual behaviours that were consistent with autism diagnostic criteria (i.e., DSM-III, DSM-III-R, DSM-IV, DSM-IV-TR, DSM-5, ICD-9, or ICD-10); and (3) peer-reviewed quantitative and/or qualitative articles.

Studies were excluded from the review if (1) participants had no diagnosis of autism and/or the mean age was 6 years or above; (2) no distinction was made between autistic

Table 1 Search strategy used in electronic databases

1. Autism	Autis* OR ASD OR ASD or asperger* OR pervasive developmental disorder* OR PDD*
2. Young children	Toddler* OR infant* OR baby OR babies OR preschool* OR pre-school* OR child*
3. Female	Gender OR sex OR female OR girl
4. Early signs	"Early sign*" OR "early marker*" OR "early behavioural marker*" OR "early behavioral marker*" OR "early symptoms" OR "early concern*" OR screening OR Point* OR "eye contact" OR eye-contact OR gaze OR waving OR wave* OR gestur* OR play* OR imitat* OR "turn taking" OR turn-taking OR "taking turn*" OR smil* OR "joint attention" OR "social attention" OR "social interaction" OR "social reciprocity" OR social-reciprocity OR "social-emotional reciprocity" OR conversation OR "nonverbal communication" OR "non-verbal communication" OR socialisation OR socialization OR restricted OR repetitive OR interest* OR "hand flapping" OR "hand flicking" OR hand-flapping OR "body rocking" OR "toe walk*" OR spin* OR jump* OR stim* OR echolalia OR sameness OR mouth* OR lick* OR "stereo* movement" OR "stereo* behaviour*" OR "stereo* behavior*" OR blinking OR preoccupation* OR sensory*
5. 1 and 2 and 3 and 4	

females and other participants (e.g., males or TD peers) in the analyses; (3) only broad constructs of autism, including global or domain scores (i.e., SCI and RRBIs) were reported; (4) they were published in languages other than English; and/or (5) other study designs were employed, such as case studies, single-case designs, pre-post and/or longitudinal intervention studies without baseline data, review articles, books or book chapters, dissertations, editorials, commentaries, and grey literature.

Study Selection

All potential studies were independently screened by two authors, TC and NF, to eliminate those that did not meet the inclusion criteria. Inter-rater agreement was very strong: 99% for title and abstract screening, and 88% for full-text screening. Any discrepancies were discussed and consensus was reached.

Data Extraction

Data was extracted from eligible studies by TC and reviewed by NF. Any discrepancies were discussed and agreement was achieved. Extracted data from each study included (1) publication details (including authors, date and country of research), (2) research design, (3) participant characteristics (e.g., sample size, age, sex/gender, comparison groups, diagnosis, diagnostic method), (4) behaviours investigated, (5) measure of outcome variable, and (6) relevant findings.

Quality Assessment

A critical evaluation, assessing the quality of each included article, was conducted using the Appraisal Tool for Cross-Sectional Studies (AXIS; Downes et al., 2016). The AXIS tool consists of 20 items that evaluate study rationale, methods,

results, discussion, and other aspects of study design, as well as risk of bias. Each criterion was rated as either 'yes' = 1, 'no' = 0, and 'do not know' = 0. Scores ranged from 0 to 20, with higher overall scores reflecting higher quality and lower risk of bias. Papers were classified as low (0–7), moderate (8–14), or high (15–20) quality, based on classifications used in previous literature (Moor & Anderson, 2019; Musetti et al., 2021). The quality assessment of eight out of the nine included articles was completed by two independent authors, TC and NF. To minimise any bias, the quality assessment of Barbaro and Freeman (2021) was independently completed by TC and a fellow student researcher, as JB and NF were authors of this paper. Any discrepancies (< 1%) were discussed and consensus was achieved on all items.

Results

As detailed in the PRISMA flow diagram in Fig. 1, a total of 1760 articles were screened for eligibility, and the full text of 52 articles were reviewed. Nine articles met the inclusion criteria and are summarised in Table 2. Articles were categorised into three broad themes, including SCI (five studies), RRBIs (five studies), and additional behaviours such as play (five studies). Four articles appear in multiple broad themes, as there were two or more unique components to each study (Barbaro & Freeman, 2021; Harrop et al., 2015b; Hiller et al., 2016; Øien et al., 2017).

Study Characteristics

The characteristics of eligible studies are presented in Table 2. A total of 1844 participants were involved across the nine studies, including 592 autistic females. All nine articles compared autistic females and males, and four articles also compared autistic females to other peers,

Fig. 1 PRISMA flow diagram of study selection
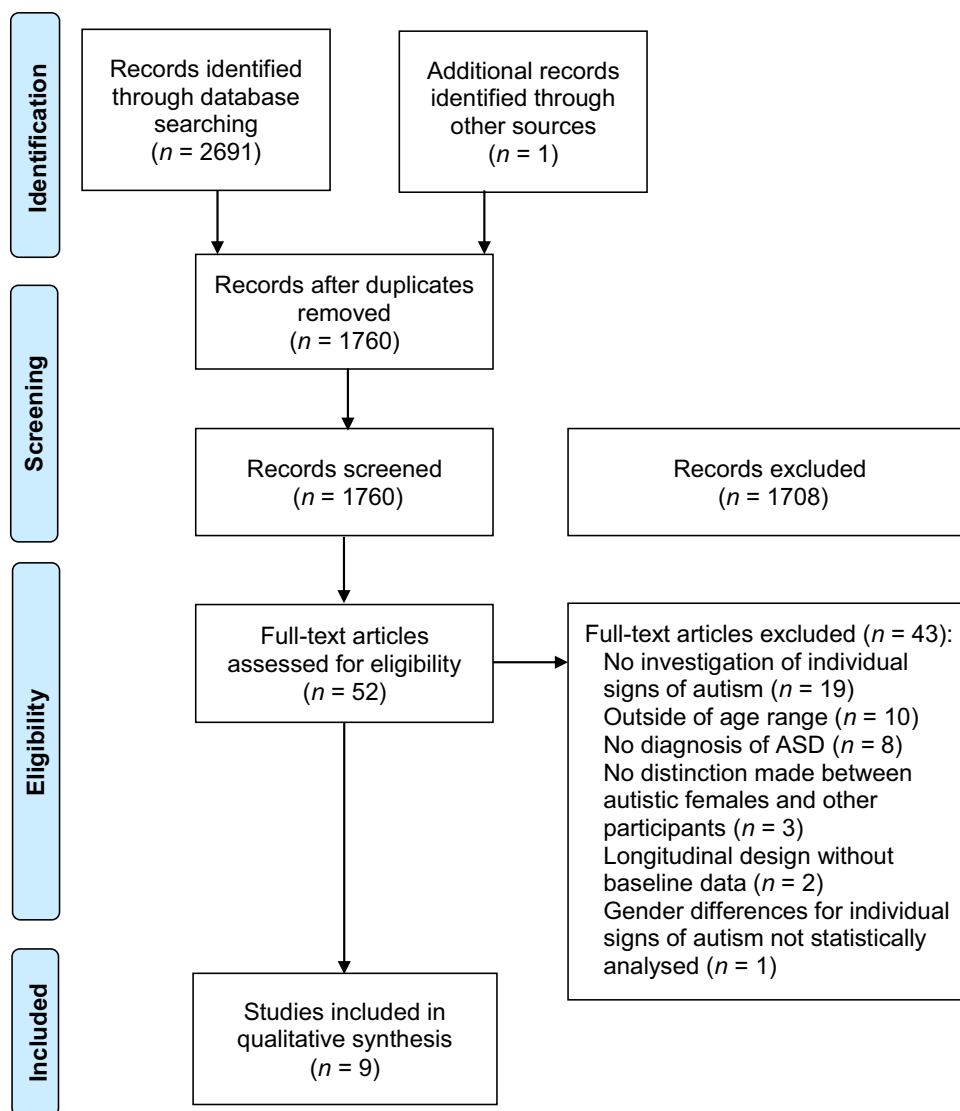
including TD (Bacon et al., 1998; Harrop et al., 2017; Knutsen et al., 2019), and those with an intellectual disability (ID; Bacon et al., 1998; Goldman et al., 2009; Knutsen et al., 2019), developmental and/or language delay (DD/LD; Bacon et al., 1998; Goldman et al., 2009). Two studies used parent-reported questionnaires to measure outcomes (Hiller et al., 2016; Øien et al., 2017), while seven studies used clinician observations (Bacon et al., 1998; Barbaro & Freeman, 2021; Goldman et al., 2009; Harrop et al., 2017, 2015a, b; Knutsen et al., 2019). Of the nine included studies, one involved retrospective analysis, whereby parents of older children reported on their child's behaviours before the age of 6 (Hiller et al., 2016).

Quality Assessment

There was excellent agreement between appraisers (ICC = 0.971, $p < 0.001$) for AXIS ratings. The quality scores for the included studies ranged from 14 to 17 out of 20 (see Table 2). Six out of the nine studies were rated as high quality, with three studies rated as moderate quality. Overall, weaknesses in study designs were apparent on AXIS items three and seven, as no study reported a power analysis to justify sample size, and eight out of nine studies did not address and/or categorise non-responders.

Social Communication and Interaction

Verbal Communication

One out of the three studies examining verbal communication differences in autistic females and males reported a significant difference across sex/gender (Hiller et al., 2016). Specifically, parent-report indicated that females had more advanced vocabulary than males (Hiller et al., 2016). In contrast, no significant differences were found for 2-word

Table 2 Summary of Included Studies

Study: author (year), country, design	Sample characteristics		ASD diagnosis confirmation	ASD behaviour(s) assessed	Outcome measure(s)	Relevant Findings	Quality rating
	N (F:M)	Groups of interest (mean age)					
Bacon et al. (1998), US, cross-sectional observations	193 (45:148)	32 ASD NVIQ≥80; 6 females; 26 males (55.84 months) 51 ASD NVIQ<80; 4 females; 47 males (62.04 months) 42 DLD; 13 females; 29 males; (53.93 months) 39 intellectual disability; 11 females; 28 males (55.85 months) 29 TD; 11 females; 18 males (55.72 months)	Clinician assessment and brief Wing interview, according to DSM-III-R criteria	Response to distress	Response to non-social orienting stimulus (i.e., unfamiliar noise) and social simulated distress from examiner during semi-structured play session	No significant sex*diagnosis effects found for response to orienting and simulated distress	15
Barbaro and Freeman (2021)[a], Australia, cross-sectional observations	145 (33:112)	*18-months:* 12 ASD females (19.8 months) 45 ASD males (19.2 months) *24-months:* 21 ASD females (25.9 months) 67 ASD males (26.1 months)	Clinician assessment, ADOS-2 Toddler and ADI-R. Specific diagnostic criteria not reported	Pointing; eye contact; waving; showing; pretend play; imitation; response to name; follows point; social smile; uses 5–10 or 20–50 words; understands words; obeys simple instructions; pointing to facial features; loss of skills; 2-word utterances; parallel play; interest in other children	Clinician report SACS-R at 18 and 24 months of age	No significant differences found between females and males on any of the SACS-R 18-month and 24-month behaviours	17
Goldman et al. (2009), US, cross-sectional observations	277 (68:209)	44 ASD NVIQ≥80; 7 females; 37 males (59 months) 85 ASD NVIQ<80; 16 females; 69 males (59 months) 93 DLD; 24 females; 69 males (51 months) 55 Non-ASD NVIQ<80; 21 females; 34 males (54 months)	Wing Autistic Disorder Interview Checklist, according to DSM-III criteria	Motor stereotypies including face; head, trunk, shoulders; arm/leg; hand/finger; hand/finger with object; gait; self-directed; visual	Characteristics and number of repetitive movements (i.e., rhythmic; unilateral; bilateral; axial) observed at least twice during standardised play session with examiner	Autistic females (NVIQ<80) had significantly more rhythmic movements, including head/trunk (e.g. bending and scrunching, tilting the head from side to side), and arm/hand (e.g. flapping, clapping, and tapping), than autistic males	14

Table 2 (continued)

Study: author (year), country, design	Sample characteristics		ASD diagnosis confirmation	ASD behaviour(s) assessed	Outcome measure(s)	Relevant Findings	Quality rating
	N (F:M)	Groups of interest (mean age)					
Harrop et al., (2017), UK, cross-sectional observations	54 (26:28)	14 ASD females (46.07 months) 14 ASD males (44.79 months) 12 TD females (26.17 months) 14 TD males (22.21 months)	ADI-R and ADOS-G. Specific diagnostic criteria not reported	Play behaviour examining play complexity and toy engagement	*Play complexity:* percentage of time spent engaged in play and each play category; and highest level of play reached, during semi-structured play session with caregiver *Toy engagement:* portion of time spent engaged in toys; and number of different toys engaged with during semi-structured play session with caregiver	No significant group*sex effects found for play complexity, including time spent engaged in the different play complexity types; the time spent not engaged in play; the time spent in person engagement; or the highest level of play reached A significant group*sex effect found for engagement with the garage and cars, as TD males spent more time engaged with this set of toys than all other groups. No other significant group*sex effects found in toy engagement	17
Harrop et al., (2015a), US, cross-sectional observations	58 (29:29)	29 ASD females (38.81 months) 29 ASD males (35.83 months)	ADOS-2. Specific diagnostic criteria not reported	RRB categories: Object; motor/ mannerisms; sensory seeking; sensory aversion; sensory-visual; verbal	Frequency of RRBs (total and across categories) observed during standardised caregiver-child play interaction	No significant differences found between females and males on any of the RRBs within the caregiver-child play interaction	14

Table 2 (continued)

| Study: author (year), country, design | Sample characteristics | | ASD diagnosis confirmation | ASD behaviour(s) assessed | Outcome measure(s) | Relevant Findings | Quality rating |
	N (F:M)	Groups of interest (mean age)					
Harrop et al., (2015b), US, cross-sectional observations	80 (40:40)	40 ASD females (40.64 months) 40 ASD males (40.05 months)	ADOS-2. Specific diagnostic criteria not reported	Play behaviour, including play complexity and type Initiation of joint attention; response to joint attention; initiation of behavioural requesting; response to behavioural requesting	*Play complexity:* Percentages of total acts by play level (drawn from DPA) during structured play assessment with examiner *Play type:* Frequency of novel, child-initiated play types during structured play assessment with examiner *Joint attention and behavioural requesting:* ESCS	No significant differences found between females and males in play type or complexity No significant differences found between females and males in initiates joint attention or response to joint attention No significant differences found between females and males in initiation of behavioural requesting or response to behavioural requesting	14
Hiller et al. (2016), AUS, cross-sectional survey. Retrospective analysis	152 (60:92)	60 ASD females (10.27 years) 92 ASD males (11.27 years)	Carer report. Specific diagnostic criteria not reported	First social concern; imitation; interest in parts of mechanical objects; desire to be liked; vocabulary; response to light; response to being held; health; withdrawn presentation; lining up objects; destructive; managing change; aggression; providing comfort; friends; preparation to be picked up; play style	Parent-report on 17 items from a questionnaire pertaining to child's functioning when under 6 years of age, based on items found in standardised diagnostic instruments (e.g., ADI-R; Diagnostic E-2 Checklist; and Autism Detection in Early Childhood)	Females were significantly more likely to engage in complex imitation, have a strong desire to be liked by peers, and have more advanced vocabulary than males Females were significantly less likely to have interests in parts of mechanical objects than males No significant differences found for any other items	16

Table 2 (continued)

Study: author (year), country, design	Sample characteristics		ASD diagnosis confirmation	ASD behaviour(s) assessed	Outcome measure(s)	Relevant Findings	Quality rating
	N (F:M)	Groups of interest (mean age)					
Knutsen et al. (2019)[a], US and Canada, cross-sectional observations	702 (351: 351)	*Less than 6-years old:* 151 ASD females IQ≥70 (mean age NR) 151 ASD males IQ≥70 (mean age NR) 200 ASD females IQ<70 (mean age NR) 200 ASD males IQ<70 (mean age NR)	Clinician assessment and ADOS-2, according to DSM-IV or DSM-5 criteria	Stereotyped/idiosyncratic language; sensory interests in play material/person; hand and finger and other complex mannerisms; repetitive interest or stereotyped behaviours	ADOS-2 RRB domain and subcategory item scores	Females (IQ≥70) had significantly less repetitive interest or stereotyped behaviours than males (IQ≥70) No significant differences observed for any other behaviours	16
Øien et al. (2017)[a], Norway, cross-sectional survey	183 (32: 153)	32 ASD females (18.48 months) 153 ASD males (18.57 months)	Clinical records from the Autism Birth Cohort or the Norwegian Patient registry. Specific diagnostic criteria not reported	Enjoying being swung/ bounced; interest in other children; enjoy climbing on things; enjoy peek-a-boo; pretend play; point to ask for something; point to indicate interest; functional play with objects; show objects to others; eye contact; oversensitive to noise; response to smile; imitation; response to name; follow to point; walk unaided; look at things you are looking at; unusual finger/hand movements; attract attention to play/activity; suspected deafness; understand speech; stare at nothing; check parents reaction	Parent-report M-CHAT	Females had a higher number of failed items than males Females were significantly more likely to fail item 13 (imitation) compared to males Females were significantly less likely to fail item 15 (follow to point) than males No significant differences found for any other items	16

Note. ADOS-2, Autism Diagnostic Observation Schedule-Second Edition; *ADOS-G*, Autism Diagnostic Observation Schedule – Second Edition, toddler module; *ADOS-G*, autism diagnostic observation schedule-generic; *ADI-R*, autism diagnostic interview-revised; *ASD*, autism spectrum disorder; *DLD*, developmental language disorder; *DPA*, developmental play assessment; *DSM-III*, Diagnostic And Statistical Manual Of Mental Disorders (3rd edition); *DSM-III-R*, Diagnostic and Statistical Manual of Mental Disorders (3rd edition, revised); *DSM-IV*, Diagnostic and Statistical Manual of Mental Disorders (4th edition); *DSM-5*, Diagnostic and Statistical Manual of Mental Disorders (5th edition); *ESCS*, Early Social Communication Scales; *M-CHAT*, Modified Checklist for Autism in Toddlers; *NR*, not reported; *NVIQ*, non-verbal intelligence quotient; *RRB*, restricted and repetitive behaviours; *TD*, typically developing; *SACS-R*, Social Attention and Communication Surveillance-Revised

[a]Sample characteristics reported are subsamples pertaining to the groups and/or analysis of interest to this review

utterances, 5–10 words, and 20–50 words (Barbaro & Freeman, 2021), as well as in understanding speech/instructions (Barbaro & Freeman, 2021; Øien et al., 2017).

Nonverbal Communication

Nonverbal communicative behaviours used for social interactions were explored in four studies, with no significant sex/gender differences noted (Barbaro & Freeman, 2021; Harrop et al., 2015b; Hiller et al., 2016; Øien et al., 2017). Autistic females did not differ from autistic males regarding the use of conventional gestures, such as protodeclarative pointing (Barbaro & Freeman, 2021; Øien et al., 2017), protoimperative pointing (Øien et al., 2017), waving (Barbaro & Freeman, 2021), and preparation to being picked up (Hiller et al., 2016). Eye contact (Barbaro & Freeman, 2021; Øien et al., 2017), initiating behavioural requests (e.g., eye contact and/or pointing), and responding to behavioural requests, was also similar across autistic females and males (Harrop et al., 2015b).

Social-Emotional Reciprocity

Mixed results were found across three studies investigating imitation. Øien et al. (2017) found that autistic females showed less parent-reported imitation than autistic males. In contrast, Hiller et al. (2016) found that autistic females were more likely to engage in parent-reported complex imitation, such as imitation games or multiple actions than autistic males. Findings from Barbaro and Freeman (2021) found no significant differences in imitation abilities via professional observations.

Variations of sharing interest were evaluated by three studies (Barbaro & Freeman, 2021; Harrop et al., 2015b; Øien et al., 2017). Based on parent report, Øien et al. (2017) found that autistic females were more likely to follow a finger point than autistic males. When using clinician observations, however, no differences between autistic females and males on following a point were observed (Barbaro & Freeman, 2021). Furthermore, no sex/gender differences were found for joint attention, including initiating and responding (Harrop et al., 2015b), and looking where others are looking (Øien et al., 2017). Shared interests, including showing (Barbaro & Freeman, 2021; Øien et al., 2017), pointing to indicate interest, and attracting attention to play activities (Øien et al., 2017), were also similar across females and males. Additionally, there was no sex/gender difference in response to name (Barbaro & Freeman, 2021; Øien et al., 2017).

Four studies considered sharing of emotions, with no differences between sex/gender found for providing comfort (Hiller et al., 2016), responding to distress (Bacon et al., 1998), response to being held (Hiller et al., 2016), and response to social smile (Barbaro & Freeman, 2021; Øien et al., 2017).

Friendship Development and Maintenance

Social motivation was explored via parent-report in three out of five studies looking at SCI (Barbaro & Freeman, 2021; Hiller et al., 2016; Øien et al., 2017). Hiller et al. (2016) found that autistic females were more likely to desire to be liked by peers than autistic males. Barbaro and Freeman (2021) and Øien et al (2017), however, concluded that there were no sex/gender differences when it came to interest in other children. Similarly, there was no difference found for first concern about socialising behaviours (Hiller et al., 2016).

Restricted and Repetitive Behaviours and Interest

Stereotyped or Repetitive Speech, Motor Movements, and/or Use of Objects

Five studies investigated items related to stereotyped or repetitive speech, motor movements, and/or use of objects, with mixed results (Goldman et al., 2009; Harrop et al., 2015a; Hiller et al., 2016; Knutsen et al., 2019; Øien et al., 2017). When separating participants based on their intellectual functioning, Goldman et al (2009) identified significantly more rhythmic movements, including head/trunk and arm/hand, in autistic females (non-verbal IQ ≤ 80) than autistic males (non-verbal IQ ≤ 80). Knutsen et al (2019) also found that autistic females (IQ ≥ 70) had significantly less repetitive interests or stereotyped behaviours than autistic males (IQ ≥ 70).

In contrast, three studies found no sex/gender differences pertaining to hand, finger, and other complex mannerisms (Harrop et al., 2015a; Knutsen et al., 2019; Øien et al., 2017), while two studies found no sex/gender differences related to repetitive behaviours, including lining up objects (Harrop et al., 2015a; Hiller et al., 2016). Additionally, the two studies examining stereotyped/idiosyncratic language found no differences between autistic females and males (Harrop et al., 2015a; Knutsen et al., 2019).

Sensory Interests and Sensitivities

Sensory interests and sensitivities were explored in four of the five studies investigating RRBIs (Harrop et al., 2015a; Hiller et al., 2016; Knutsen et al., 2019; Øien et al., 2017). No differences were found between autistic females and males on sensory interests (Harrop et al., 2015a; Knutsen et al., 2019), sensory-visual (i.e., visual inspection), and sensory aversions/sensitivities (Harrop et al., 2015a; Hiller et al., 2016; Øien et al., 2017).

Management of Change and Fixated Interests

One study investigated fixated interests via parent-report, finding that autistic females were less likely to have interests in parts of mechanical objects than autistic males (Hiller et al., 2016). Hiller et al. (2016) also investigated the management of change to routine and reported no significant differences between autistic females and males.

Additional Behaviours

Five articles examined whether autistic females and males differed in behaviours not typically recognised within diagnostic manuals (e.g., DSM-5-TR; American Psychiatric Association, 2022) but are still recognised as being characteristic of some children with this diagnosis.

Play Behaviours

Play behaviours were investigated in five studies (Barbaro & Freeman, 2021; Harrop et al., 2017; 2015b; Hiller et al., 2016; Øien et al., 2017); all studies found that autistic females and males were equivalent in their play. Play behaviours included complex (i.e., simple object, combination, presymbolic, and symbolic; Harrop et al., 2017, 2015b), pretend (Barbaro & Freeman, 2021; Øien et al., 2017), parallel (Barbaro & Freeman, 2021), and functional play (Øien et al., 2017), as well as play style (Hiller et al., 2016). Additionally, autistic females did not differ from autistic males and TD females in type of toy engagement (Harrop et al., 2017).

Other

No significant sex/gender differences were found for aggressive behaviours, internalising problems (Hiller et al., 2016), health (Hiller et al., 2016; Øien et al., 2017), or loss of skills (Barbaro & Freeman, 2021).

Discussion

There is a growing acknowledgement of a sex/gender bias that impacts autistic females receiving an appropriate and timely diagnosis. The aim of this review was to therefore gain a better understanding of the female autism presentation in early childhood by systematically reviewing studies that investigated sex/gender differences in the early signs of autism in young children (i.e., under the age of 6). Overall, there were few differences in relation to sex/gender, suggesting that autistic females and males may be more similar than different in early childhood. There was evidence for some differences between autistic females and males on a few individual behaviours associated with SCI and RRBI; however, consistent with much of the autism literature, results were often conflicting. For example, some studies indicated that autistic females have particular strengths in individual behaviours related to following a finger point (Øien et al., 2017), vocabulary, social desire, and imitation (Hiller et al., 2016). This was, however, contradicted by studies that found greater imitation difficulties in females (Øien et al., 2017) and/or an absence of sex/gender differences (Barbaro & Freeman, 2021; Øien et al., 2017). Similarly, some studies found significant differences between females and males for repetitive or stereotyped behaviours (Knutsen et al., 2019) and motor movements (Goldman et al., 2009), while others did not (Harrop et al., 2015b; Hiller et al., 2016; Knutsen et al., 2019; Øien et al., 2017). Implications and possible explanations for these discrepancies are discussed below.

Sample Heterogeneity

While the cause of mixed findings is likely multifactorial, the heterogeneity of this population and factors that may influence autistic traits in different individuals should be considered. Studies included in the review consisted of children with varying cognitive, verbal, and non-verbal abilities, all of which could be factors contributing to the different manifestations of autistic traits (Mayes & Calhoun, 2011; Rommelse et al., 2015; Venker et al., 2014). It was only when participants were separated based on their intellectual functioning that significant differences between autistic females and males related to RRBIs were found (Goldman et al., 2009; Hiller et al., 2016; Knutsen et al., 2019). For example, autistic females without ID had reduced repetitive interests or stereotyped behaviours (Knutsen et al., 2019), and less fixated interests in parts of mechanical objects (Hiller et al., 2016) than autistic males without ID. For females with a non-verbal IQ of less than 80, however, more stereotyped *rhythmic* movements were observed (Goldman et al., 2009). When IQ was not stratified, females and males did not differ (Harrop et al., 2015a; Øien et al., 2017). This suggests that some sex/gender differences may be contingent on the level of intellectual functioning, particularly as repetitive and stereotyped motor movements are also pronounced in those with ID alone (Bodfish et al., 2000). Therefore, it is currently unclear whether it is autistic females or females with a certain IQ who may present with differing individual RRBIs.

Measurement Considerations

There was also heterogeneity in the measurement of early signs of autism across studies, including the use of various parent-report surveys and observational-based assessments. Measures that had not been previously trialled and/or validated were also utilised (Bacon et al., 1998; Goldman et al., 2009; Hiller et al., 2016), as well as retrospective reporting (Hiller et al., 2016).

Retrospective Analysis

Retrospective research by Hiller et al. (2016) that involved children diagnosed with autism after the age of 5 accounts for a large portion of the significant sex/gender differences found within this review. Although not evident in autistic children identified during early childhood (Barbaro & Freeman, 2021; Øien et al., 2017), results of the retrospective analysis indicated that autistic females who received a diagnosis of autism later in life demonstrated more advanced individual SCI behaviours (i.e., vocabulary, imitation, and social desire) and fewer individual RRBIs (i.e., fixated interests in parts of mechanical objects) in early childhood than males (Hiller et al., 2016). When similar results have been reported in previous research, they were attributed to the notion that females mask or camouflage their difficulties in social settings (Attwood, 2007; Wing, 1981). As this review focused on early childhood, however, it is unlikely that the ability to engage in such complex practices, including understanding others' behaviour, recognising one's strengths and difficulties, and developing strategies to compensate and reduce the appearance of autistic behaviours, would have yet been developed (Banerjee & Yuill, 1999; Knutsen et al., 2019; Pellicano et al., 2014). Rather, these results support the theory of a different expression of autism in some females (Hull et al., 2017; Kirkovski et al., 2013; Lai et al., 2015) and can provide insight into why it is more difficult to detect autism in females who may be missed at an early age. For reasons not yet known, it appears that some SCI difficulties in this subgroup of females are not as overt/recognisable as those who are diagnosed in early childhood.

Parental Reporting

Interestingly, most sex/gender differences found in this review were also the result of information provided by parents (Hiller et al., 2016; Øien et al., 2017). Such findings often lacked consistency across informants, as the same sex/gender differences were not observed by clinicians. Concerns regarding the accuracy of parent-reported data have been raised, as it is vulnerable to interpretation and recall bias (Ozonoff et al., 2008, 2018b). Correct recall is particularly challenging when

parents retrospectively report on their children's behaviour, as per Hiller et al. (2016), because there is a greater likelihood of errors in memory (Hus et al., 2011; Ozonoff et al., 2018b). Furthermore, the reliability of parent-reported data is often questioned because parents lack the specific knowledge that professionals possess when it comes to evaluating child development (Law & Roy, 2008; Nordahl-Hansen et al., 2014). Such limitations suggest that the parent-reported sex/gender differences discovered in this review may not be a true reflection of a child's behaviour/autistic traits.

On the other hand, some research has found that parents are usually reliable reporters (Larsen et al., 2018; Miller et al., 2017; Spikol et al., 2019). Parents possess the most knowledge about their child's everyday functioning and can raise concerns about behaviours that may be missed during a standardised assessment (Horovitz et al., 2012; Ozonoff et al., 2018a). Retrospective parent reporting can also help identify what behaviours were most memorable (and most likely challenging) to parents after time has passed (Tanner & Dounavi, 2021). Additionally, as mentioned by Øien et al. (2017), survey and interview questions can be open to interpretation depending on the wording and examples provided. Parents' interpretation of questions may differ from clinicians (given differences in background and training) and can therefore offer perspectives that may not have been considered. Differences in informants likely contribute to conflicting findings, although both parental reports and professional observations offer valuable information. Neither should be preferred nor discounted, but rather, used together to gain a greater understanding of early childhood signs of autism.

Individual Behaviours

The significant finding from Goldman et al. (2009) highlights the importance of breaking observed behaviours down into their individual form. In children with ID, Knutsen et al. (2019) found no significant sex/gender differences for stereotyped movements, although this item on the ADOS-2 (Lord et al., 2012) covers a range of movements, including hand and finger, and other complex body mannerisms. Females were only noted to differ from males once motor stereotypies were categorised into specific forms (i.e., rhythmic, head/trunk, and arm/hand; Goldman et al., 2009). Similarly, Hiller et al. (2016) found sex/gender differences regarding fixated/repetitive interests, possibly because the item pertained specifically to parts of mechanical objects, rather than a variety of behaviours (as per item D4 of the ADOS-2; Knutsen et al., 2019). It seems that when individual behaviours are grouped into one item, sex/gender differences may be missed. It is only when behaviours are broken down individually that differences can be identified.

Sample Size

As per the quality analysis, a key weakness of eight out of the nine studies included in this review was the lack of power analysis to justify their sample size. Not only were more males than females included in most study samples, but several studies also reported their small sample size to be a limitation (Barbaro & Freeman, 2021; Harrop et al., 2017, 2015a). All significant findings came from studies that had some of the largest sample sizes, while some of the studies with small samples noted that their results showed trends toward significant sex/gender differences in the early signs of autism (Harrop et al., 2015a, b). Although it cannot be said with certainty, it is possible that some of these conflicting findings and non-significant trends are a consequence of small sample sizes, and that more differences may have been reported if studies were adequately powered.

Implications

This systematic review revealed no consistent sex/gender differences in the early signs of autism — so why is it that females are at a much greater likelihood of going unidentified than males? While further research is needed, the findings indicate two possible reasons. Firstly, it may be that a female-specific manifestation of autism (i.e., phenotype) does exist, whereby some females present with an expression of autism in early childhood that does not fit with our current conceptualisation (Hull et al., 2017, 2020; Kirkovski et al., 2013; Lai et al., 2015). However, earlier studies were predominantly made up of individuals who had already come to the attention of clinicians/received a diagnosis of autism, meaning their presentations aligned with existing (male-influenced) diagnostic criteria (Kopp & Gillberg, 1992; Kreiser & White, 2014). Therefore, these studies may not have included those individuals who expressed their early characteristics of autism in an unfamiliar manner, limiting our understanding of this specific manifestation. Results from Hiller et al. (2016) provide insight into this hypothesis and support the proposition that females who are most likely to present with a different expression of autism are those who go un- or misdiagnosed early in life, not those that make up clinical samples. Furthermore, while results of this review suggest that play and aggressive behaviours, internalising problems, health, and loss of skills, are additional characteristics that do not appear to differ between females and males, more research is needed and there is still a plethora of behaviours to be investigated. For example, Tanner and Dounavi (2021) reported that delayed fine or gross motor skills can be an indicator of autism. Research also suggests that demand avoidance is worth exploring (Kopp & Gillberg, 2011). Additional behaviours that have not yet been considered by the current literature may also make up the female presentation of autism.

Secondly, it could in fact be that females do not differ from males in the early childhood signs of autism. Although it remains unclear as to why some females are not recognised as being on the autism spectrum at a younger age, genetics, comorbidities, culture, and social and economic factors could instead be at play (Kreiser & White, 2014; Lai & Szatmari, 2020; Mazzone et al., 2012). It may also be that there is a subgroup of children — both female and male — who are missed very early on (potentially due to the aforementioned factors). However, further investigation would be required as there is very limited research and knowledge about later diagnosed males and their experiences, given that later diagnosed research focuses primarily on females and understanding the female autism phenotype (Bargiela et al., 2016; Lehnhardt et al., 2016).

Limitations

One limitation is the scarce amount of research in this specific field and consequently the small number of articles included in this review. Given this, the conclusions made are difficult to generalise. Conclusions drawn were also limited due to the heterogeneous nature of sample characteristics, methodology (e.g., measurement), and study findings. Additionally, as papers published in languages other than English were excluded from this review, relevant studies that provide different sociocultural perceptions of autistic behaviours were potentially missed (Harrison et al., 2017; Young et al., 2018).

While this paper set out to review the early childhood signs of autism in females, participants within and across samples spanned multiple developmental periods: infancy, toddlerhood, and preschool. Autistic traits can change across early development (Gotham et al., 2012; Venker et al., 2014), which may also correspond to sex/gender differences (Szatmari et al., 2015; Waizbard-Bartov et al., 2021). Thus, any potential differences pertaining to a specific developmental period could have been concealed by the wide-spanning age range of participants.

Future Directions

Ultimately, the contradictory findings, heterogeneous nature of autism, different study designs used, and the lack of replicated studies make it difficult to determine if any sex/gender differences in the early childhood signs of autism exist. Given this, large-scale research that utilises similar samples and methodologies is needed in this field. This will allow for comparisons across the literature and clearer conclusions to be made about autistic females. Future research should also examine IQ and age as possible factors influencing sex/

gender differences. Additionally, ongoing research exploring sex/gender differences in additional behaviours not currently included in the diagnostic criteria is in the very early stages and remains of great importance (as per Lai et al., 2015). Future research should consider behaviours not captured in this review, such as fine and gross motor difficulties (Tanner & Dounavi, 2021), demand avoidance (Kopp & Gillberg, 2011), and novel behaviours outside those typically targeted in the literature.

Our understanding of the earliest childhood signs of autism in females who are diagnosed later in life is not well understood, partially because much of the existing research consists of young children who have already received a diagnosis (Loomes et al., 2017). Therefore, to better understand why some autistic females are missed early on, investigating the early childhood signs of autism specific to these later diagnosed females is key (rather than focusing on young females who have already received a diagnosis of autism). This research should consider methods such as retrospective video analysis of females and males diagnosed later in life. This will allow for observation of the earliest-presented signs of autism in females as compared to males while removing memory errors and recall bias. Interviews with parents of these children, however, can also be of value, as it will allow for interpretation and differing perspectives, as well as individual behavioural exploration (rather than grouping of behaviours).

Conclusion

This is the first systematic review to synthesise studies that investigated sex/gender differences in the early childhood signs of autism, including those associated with SCI, RRBIs, and additional behaviours. While there is some evidence that females differ from males on a few individual signs of autism, this review indicates that there are more sex/gender similarities than differences in the early years of life. It is, however, difficult to draw accurate conclusions based on the information available in the current literature. Further large-scale research with more homogeneous study designs is needed to disentangle the current ambiguity. Specifically, it is important to focus closely on later diagnosed females to better determine whether their early signs of autism present differently to males, and if there are signs of autism not yet identified that are specific to them during early childhood. In doing so, important findings can be shared with health professionals to allow them to better detect autism in females in a timely manner, increasing opportunities for females to access early supports and services, and ultimately enhancing their development and well-being.

Declarations

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