



# Age of Diagnosis for Co-occurring Autism and Attention Deficit Hyperactivity Disorder During Childhood and Adolescence: a Systematic Review

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## Abstract

Early identification and intervention are recognised as important elements of the clinical pathway for autism spectrum disorder (ASD). Children with ASD and attention deficit hyperactivity disorder (ADHD) may be diagnosed at a different age than children who only have one of these diagnoses. This systematic review aimed to identify the age at which children were diagnosed with both ASD and ADHD. Of the 9552 articles screened, 12 were included in the review. The findings suggest that ASD is typically diagnosed later when ADHD is present, and ADHD is typically diagnosed earlier when ASD is present. Further research is needed to understand the factors impacting a delayed ASD diagnosis and an earlier ADHD diagnosis when the two conditions co-occur.

**Keywords** Co-occurring conditions · Diagnosis · Autism · Attention deficit hyperactivity disorder · Age of diagnosis

ASD is characterised by impairments in social communication and the presence of fixed, rigid behaviours (American Psychiatric Association, 2013). The number of individuals with ASD who also have an ADHD diagnosis ranges from 59 to 83% (Joshi et al., 2020).

Evidence suggests that children with ASD and co-occurring ADHD are a high-need population (Hong et al., 2020; Zablotzky et al., 2020). They appear to have poorer

outcomes overall and reduced response to social skills intervention than children with either diagnosis alone (Elwin et al., 2020; Fleming et al., 2020; McDougal et al., 2020). Co-occurring ADHD is associated with more severe ASD symptoms, lower adaptive functioning, and greater cognitive impairment compared to an ASD diagnosis alone (Yerys et al., 2019; Zachor & Ben-Itzhak, 2020). While emotional and conduct challenges for children with ASD appear to reduce overtime, these challenges appear to increase for children with ASD + ADHD (Flouri et al., 2015). Co-occurring ADHD also appears to further exacerbate stress, financial, and time burdens for families (Dovgan & Mazurek, 2019).

There is preliminary evidence that obtaining an ASD diagnosis may be delayed when ADHD is co-occurring (Davidovitch et al., 2015; Frenette et al., 2013; Jónsdóttir et al., 2011; Miodovnik et al., 2015). This delay may have implications for longer-term outcomes, given the known importance of prompt intervention in early childhood (Towle et al., 2020). This delay in diagnosis seems counterintuitive given that outcomes for children with ASD and co-occurring ADHD (hereafter referred to as ASD + ADHD) are more severe, and severity typically leads to earlier health-seeking behaviour and, ultimately, diagnoses (Miller et al., 2021). Data provided by the Centre for Disease Control and Prevention (2014) states the average age of diagnosis for ASD is 4 years and 4 months, and the average age for diagnosis of

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ADHD is 7 years (Visser et al., 2014) which suggests that autism is generally diagnosed prior to ADHD. The DSM-5 specifies that the symptoms must be present in childhood for both autism and ADHD (under the age of 12 years) (Kooij et al., 2019). Late diagnosis of autism is usually defined as occurring during adolescence (Hosozawa et al., 2020). Delayed diagnosis of adults is beyond the scope of this review. The missed diagnosis of an adult group has been described as a “lost generation” and provides a number of challenges, including retrospectively collecting a developmental history of symptoms present in early childhood (Lai and Baron-Cohen 2015). There is also controversy over whether adult-onset of ADHD can occur (Mucci et al., 2018).

It has been hypothesised that the presence of ADHD symptoms may make identifying ASD behaviours more challenging in the diagnostic process (Soke et al., 2018). Indeed, studies that have examined missed diagnoses of ASD suggest that co-occurring ADHD may be a contributing factor (Davidovitch et al., 2015; Fusar-Poli et al., 2020; Hosozawa et al., 2020). In a study of the objectivity of a widely used clinical tool that informs ASD diagnosis, Autism Diagnostic Observation Schedule (ADOS), only 25% of children with ASD + ADHD were correctly identified using this measure (Zander et al., 2016). Park et al. (2014) found that the distinguishing factor between those referred for ASD and diagnosed with ASD, and those referred for other reasons, but subsequently diagnosed, was that the former group had higher support needs with significantly lower externalising and hyperactive behaviour. They hypothesise that externalising behaviours and hyperactivity may mask ASD symptoms, and that externalising and hyperactive behaviours are more likely to occur if the individual with ASD has higher adaptive functioning (Park et al., 2014).

The influence of ASD on the timing of the ADHD diagnosis, when ASD and ADHD are co-occurring, has received less attention in the literature (Miller et al., 2018). There is also less emphasis on the importance of early identification of ADHD. This may be related to multiple factors, such as the fact that (a) ADHD symptoms may be seen as developmentally typical in early childhood, (b) the impact on functioning may not be evident until the child reaches school, and (c) assessments for identification require teacher input from school age (Miller et al., 2018; Silva et al., 2015; Visser et al., 2014). Nonetheless, there is increasing interest in earlier diagnosis and non-medical intervention for younger children with ADHD (Hatakenaka et al., 2016; Miller et al., 2020).

A systematic literature review was conducted with the aim of elucidating whether there is a significant difference in the age of ASD diagnosis for children and adolescents (0–21 years of age) with ASD only and co-occurring ASD + ADHD. The review also sought to compare the age

of ADHD diagnosis for children and adolescents with ASD only and those with co-occurring ASD + ADHD. This will allow an examination of the size and variation of any differences. If differences in age of diagnosis are found, this review will also examine quantifiable factors which may contribute to this.

## Methods

This systematic review followed the procedures outlined in the Preferred Reporting Items for Systematic Review and Meta-Analyses (PRISMA) statement (Moher et al., 2009).

## Protocol

The study protocol was registered on the 31st of December, 2020 (PROSPERO 2020 CRD42020222984). [https://www.crd.york.ac.uk/prospero/display\\_record.php?ID=CRD42020222984](https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42020222984). The protocol was subsequently updated (May 13, 2021) to state that all included studies must make a comparison between the age of diagnosis or onset between an ASD + ADHD group with an ASD only and an ADHD only group or a whole sample.

## Eligibility

Studies were eligible for inclusion in the systematic review if they met all of the following criteria:

1. They included children and adolescents up to the age of 21. If individuals over the age of 21 were included, individuals who were 21 and under were analysed separately. This was to distinguish between a delay in acquiring an autism diagnosis or ADHD diagnosis when a child or adolescent has entered a diagnosis pathway from an adult diagnosis where no diagnosis might have been sought during childhood or adolescence, or a misdiagnosis might have occurred.
2. There was a defined co-occurring group of individuals who were diagnosed with ASD + ADHD and a group of individuals diagnosed with ASD only and/or ADHD only.
3. There was a comparison between the age of diagnosis or onset between an ASD + ADHD group with an ASD only and an ADHD only group or a whole sample.
4. The study was peer-reviewed, written in English, and published after 2014.
5. The methodology used included a comparison of age of diagnosis across more than one participant (e.g., brief report, review, and case studies were excluded).

## Search Strategy

The literature search was conducted on October 30, 2020 using PsycINFO, MEDLINE, Scopus, and Pubmed. The search was limited to peer-reviewed studies published in English after 2014. The search terms were (diagnos\* OR misdiagnos\*) AND (autis\* OR ASD\* OR “autistic disorder\*” OR Asperger\* OR “pervasive developmental disorder\*” OR PDD-NOS) AND (“Attention Deficit Hyperactivity Disorder” OR “Hyperkinetic Disorder” OR ADHD OR “Attention Deficit Disorder” OR ADD). In each database, the search was limited to titles, abstracts, and keywords. A grey literature search was also conducted during the week of February 15, 2021, using the Google advanced search engine (limited to the first 100 results per search) and abstracts submitted to the International Society for Autism Research (INSAR) conferences in the past 5 years. All searches were limited to articles.

## Study Selection

One reviewer (WJS) conducted all database searches and imported the results into Covidence reference management software, where duplicates were removed. The same reviewer screened all the titles and abstracts of the studies against the inclusion/exclusion criteria and a second reviewer (LM) screened 20% of the total studies against the same criteria. Then the first reviewer screened full-texts of all potentially relevant articles, and the second reviewer screened 20%. Following each stage of screening, the reviewers discussed and resolved disagreements. If a consensus could not be reached, an additional reviewer (HW) was consulted. The percentage of agreements was calculated using the formula:  $\text{agreements}/(\text{disagreement} + \text{agreements}) \times 100$ . Agreement on the title/abstract screen was 98.5%, and agreement on the full-text screen was 92.5%

## Data Extraction

Data from each study were extracted and summarised in three tables in Microsoft® Word. The following data were extracted for eligible original studies: (a) the total number of participants, (b) the number of participants in each diagnostic group, (c) participant gender, (d) geographical location, (e) the date range of the participant data set, (f) how the diagnosis was reported (diagnostic determination), (g) description of the participant population (h) the age range of the participants, (i) the age of ASD and ADHD diagnoses for each diagnostic group, (j) statistical significance of any differences in age of diagnosis, (k) the age of parental first concern, (l) diagnostic wait times for each diagnostic group, and (m) quantitative factors related to the age of diagnosis. The data for all studies were independently extracted by one

reviewer (WJS). Reviewers (LM and HW) screened 40% of the total studies for extraction data resulting in an agreement rate of 94.1%.

## Risk of Bias

The risk of bias of included studies was assessed using the Hoy et al. (2012) guidelines. The indicators of high-quality studies included in this review were: (1) target population was representational of a national population; (2) sampling frame was representative of the target population; (3) random selection was used to select a sample; (4) non-response bias minimal; (5) data collected directly from subjects; (6) acceptable case-definition of ADHD or ASD; (7) same mode of data collection used for all subjects; (8) prevalence period; (9) no errors in reporting. One item, “study instruments have reliability and validity,” was not included in this study because the primary outcome, age of diagnosis, did not require a validated measure. The item related to the prevalence period (9) was modified to reflect the time that the study allowed for a co-occurring diagnosis to occur, whereby low risk was considered greater than a 2-year window. A score of “yes”/0 indicated a low risk for that item and “no”/1, indicating a high risk for that item. Studies with scores of 0–3 were deemed “low risk,” those with scores of 4–6 were “moderate risk,” and those with scores of 7–9 were “high risk.” If there was insufficient information in the article, but the in-depth protocol was referenced elsewhere, such as a National database, then this was sought. When more information was needed, however, and this was not clearly stated in the article, by default, the indicator was noted as high risk. Two reviewers independently evaluated 20% of the articles for the quality indicators. All discrepancies were discussed and resolved, and the overall agreement was 90%.

## Community Participation

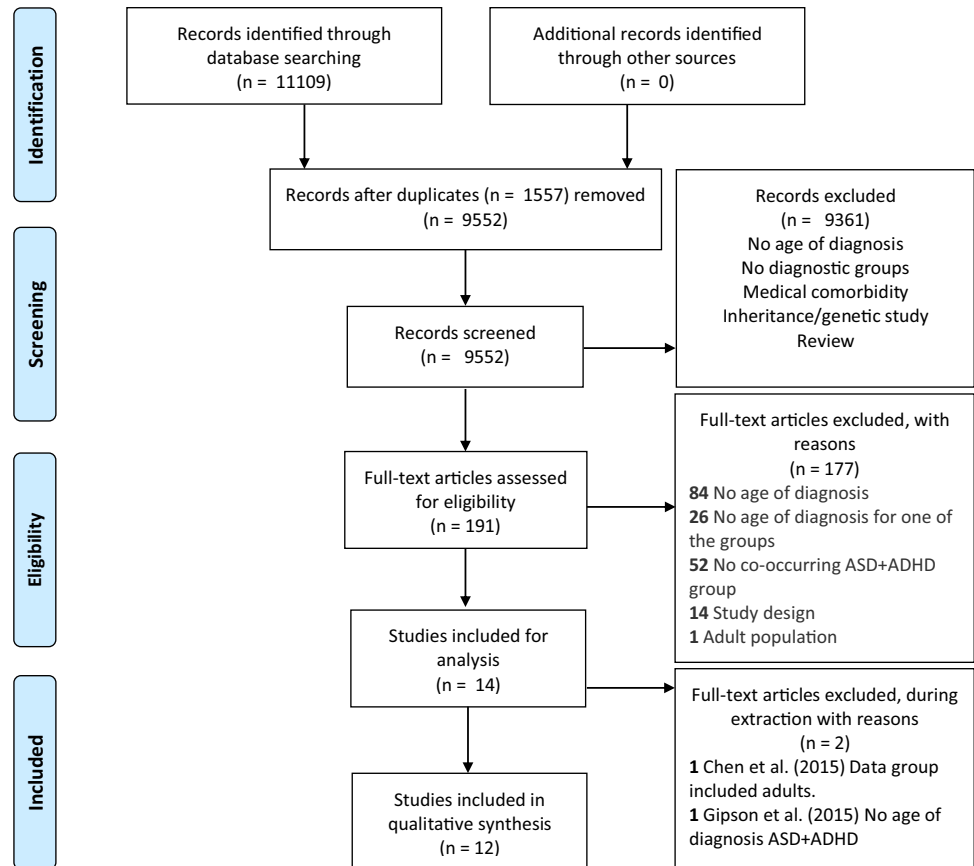
Community members were not involved in this study.

## Results

### Study Selection

The PRISMA flow diagram in Fig. 1 represents the study selection process (Moher et al., 2009). The initial screen yielded 11,109 results. After the duplicates were removed, 9552 articles were screened, and 9361 records were excluded. One hundred and ninety-one full-text articles were assessed for eligibility at the full-text stage, and 177 were excluded due to: (1) not providing quantitative data on the age of diagnosis, (2) not including a co-occurring ASD + ADHD group, (3) not having an age of diagnosis for

Fig. 1 PRISMA flow diagram



at least two groups, (4) no comparison with ASD or ADHD only group or whole group, (5) a review or a brief report, or a single case study or (6) including an adult population. Two studies that were originally included in the extraction were then excluded due to closer examination revealing that an adult population had been included (Chen et al., 2015) and that age of diagnosis for ASD + ADHD had not been reported (Gipson et al., 2015).

## Participants

Table 1 summarises the demographic characteristics, location and diagnostic determination for the 12 included studies. These studies included a total of 41,382 children with ASD, ADHD, or ASD + ADHD diagnoses, of which 14,584 were diagnosed with ASD only, 12,689 were diagnosed with ADHD only, and 14,109 were diagnosed with ASD + ADHD. All 12 studies reported participant gender, and the percentage of male participants ranged from 78.5 to 85.8%. Most studies were conducted in the USA, and studies were also conducted in Denmark, the Netherlands, Taiwan, Japan, and China. The two studies each from Taiwan and The Netherlands used the same respective national databases (Kentrou et al., 2019; Lin et al., 2014; Wang et al., 2018; Wei et al., 2021). The participant data range was from 1995

until 2020 across all studies. The main method of diagnostic determination, or how the study ascertained the diagnoses of the respective children, was through medical records. The exceptions included Miodovnik et al. (2015), Stevens et al. (2016), and Wang et al. (2018), where their diagnostic determinations were done through surveys or interviews with parents. The participants were drawn from three categories described as population, clinical and database. Population included a regional sampling of a population through general medical records across multiple health care settings, which was not specific to psychiatric hospitals or clinics specialising in the diagnosis of either ASD or ADHD. Clinical settings included any specific population drawn from a diagnosing clinic or specific psychiatric hospital. A database population was described as an elected database for parents to join, although in some cases, these databases were linked with access to nationally funded support.

## Risk of Bias

The risk of bias of each study was assessed using the indicators outlined in Hoy et al. (2012), and the values are presented in Table 2.

Nine studies were deemed low risk, and four were deemed at moderate risk of study bias. The most common

**Table 1** Demographic characteristic of participants in the included studies

Included studies	Total <i>n</i> <sup>a</sup>	Total <i>n</i> ASD only	Total <i>n</i> ADHD only	Total <i>n</i> ASD + ADHD	Male %	Geographic location	Date range of data set	Diagnostic determination	Participant description
1. Engelhard et al., 2020	1658	343	1175	140	79	USA	2013–2020	Medical records	Population
2. Hatakenaka et al., 2016	65	42	14	9	84	Japan	2012–2013	Medical records	Clinic
3. Jenson and Steinhaus, 2015	8958	NA	7116	1842	79.4	Denmark	1995–2010	Medical records	Clinic
4. Joshi et al., 2017	181	NA	74	107	82.5	USA	2005–2012	Medical records	Clinic
5. Kentrou et al., 2019	449	392	NA	57	80.2	The Netherlands	Not given	Parent/self-report	Database
6. Lin et al., 2014	5130	498	4237	395	82	Taiwan	1995–2013	Medical records	Population
7. Miodovnik et al., 2015	1451	746	NA	705	82	USA	2011–2012	Parent report	Population
8. Soke et al., 2018	1091	785 (8 yrs.)	NA	306	79.5	USA	2010	Medical records	Population
9. Stevens et al., 2016	1317	717	NA	600	78.8	USA	2011	Parent report	Population
10. Wallisch et al. 2018	363	259	73	31	80.7	USA	2000–2014	Medical records/survey	Clinic
11. Wang et al., 2018 (China)	433	186	NA	247	82.4	China	2013–2014	Parent report	Database
Wang et al., 2018 (Netherlands)	492	361	NA	131	78.5	The Netherlands	2013–2014	Parent report	Database
12. Wei et al., 2021	19,794	10,255	NA	9539	84.8	Taiwan	2001–2011	Medical records	Population

<sup>a</sup> Reflects the total of the three groups, it does not include controls or other group totals in the studies

area of higher risk was in ensuring that the participant pool was representative of a national sample. Studies that used parent reports for both diagnosis and age of diagnosis had a higher risk around case definition because data were considered collected via proxy as opposed to medical records. All articles used the same mode of data collection, so were all low risk in this area.

### Age of Diagnosis

Table 3 presents the differences in age of ASD and ADHD diagnosis depending on whether individuals were diagnosed with ASD, ADHD, or ASD + ADHD. In Wang et al. (2018), the age of ASD diagnosis for the ASD + ADHD only group was compared to the age of ASD diagnosis across the whole sample, rather than an ASD only group. The age ranges for each study differed and varied from 0 to 18 years across the studies. The studies of Hatakenaka et al. (2016) and Wallisch et al. (2020) were restricted to participants under the age of 6 years old. Soke et al. (2018) provided the mean difference

between the age of diagnosis for 8-year-old children with ASD + ADHD and ASD only. Joshi et al. (2017) provided the age of onset between groups.

### Age of Diagnosis for ASD

The average age of ASD diagnosis ranged from 3.5 years (Hatakenaka et al., 2016) to 6.2 years (Lin et al., 2014). The average age of ASD diagnosis when children had co-occurring ADHD ranged from 3.3 years (Wang et al., 2018) to 7.5 years (Lin et al., 2014). In the ASD + ADHD group, the average age of ASD diagnosis was between 0.7 (Wallisch et al., 2020) and 1.8 years (Kentrou et al., 2019; Miodovnik et al., 2015; Wei et al., 2021) later compared to the ASD only group.

### Age of Diagnosis for ADHD

The average age of ADHD diagnosis ranged from 4.9 years (Wallisch et al., 2020) to 9.8 years (Jensen & Steinhausen,



**Table 2** Risk of bias assessment results adapted from Hoy et al. (2012)

Risk of bias item <sup>a</sup>	1. Target pop	2. Sample of target	3. Random selection	4. Non-response bias minimal	5. Data direct	6. Case definition	7. Study instrument	8. Mode of data collection	9. Prevalence period	10. No errors	Total high risk items	Summary of overall risk <sup>b</sup>
1. Engelhard et al., 2020	No	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	1	Low
2. Hatakenaka et al., 2016	No	No	No	Yes	Yes	Yes	N/A	Yes	No	Yes	4	Moderate
3. Jenson and Steinhilber 2015	Yes	Yes	Yes	Yes	Yes	Yes	N/A	Yes	No	Yes	1	Low
4. Joshi et al., 2017	No	No	Yes	No	No	No	N/A	Yes	Yes	Yes	5	Moderate
5. Kentrou et al., 2019	No	No	No	No	No	No	N/A	Yes	Yes	Yes	6	Moderate
6. Lin et al., 2014	Yes	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	0	Low
7. Miodownik et al., 2015	Yes	Yes	Yes	Yes	No	No	N/A	Yes	Yes	Yes	2	Low
8. Soke et al., 2018	No	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	1	Low
9. Stevens et al., 2016	Yes	Yes	Yes	No	No	No	N/A	Yes	Yes	Yes	3	Low
10. Wallisch et al. 2018	No	No	Yes	No	Yes	Yes	N/A	Yes	No	Yes	3	Low
11. Wang et al., 2018 (China)	Yes	Yes	Yes	Yes	No	No	N/A	Yes	Yes	Yes	2	Low
Wang et al., 2018 (Netherlands)	No	No	No	Yes	No	No	N/A	Yes	Yes	Yes	5	Moderate
12. Wei et al., 2021	Yes	Yes	Yes	Yes	Yes	Yes	N/A	Yes	Yes	Yes	0	Low

<sup>a</sup> The following questions guided the assessment: 1. Was the study's target population a close representation of the national population in relation to relevant variables?; 2. Was the sampling frame a true or close representation of the target population?; 3. Was some form of random selection used to select the sample, OR was a census undertaken? 4. Was the likelihood of non-response bias minimal?; 5. Were data collected directly from the subjects (as opposed to a proxy)?; 6. Was an acceptable case definition used in the study? e.g. authors specify the method by which ADHD or ASD was diagnosed, such as ADOS using DSM V criteria; 7. Was the study instrument that measured the parameter of interest shown to have validity and reliability?; 9. Was the length of the shortest prevalence period for the parameter of interest appropriate?; 10. Were the numerator(s) and denominator(s) for the parameter of interest appropriate?

<sup>b</sup> Low risk 0–3; Moderate risk 4–6; High risk 7–9

**Table 3** Mean age of ASD and ADHD diagnosis and difference in age of diagnosis for ASD only, ADHD only, and ASD + ADHD groups

Included studies	Age range (years)	Mean age of ASD diagnosis (years)		Mean <sup>a</sup> difference	Sig	Mean age of ADHD diagnosis (years)		Mean <sup>a</sup> difference	Sig
		Whole sample				ADHD only			
		ASD only	ASD + ADHD			ASD only	ASD + ADHD		
1. Engelhard et al., 2020	1–13	4.1 (1.8)	5.5 (2.4)	-1.4		7.2 (1.8)	5.9 (1.8)	+1.3	
2. Hatakenaka et al., 2016	1.9–6	3.5 (1.1)	4.8 (0.8)	-1.3		5.1 (1.1)	4.8 (0.8)	+0.3	
3. Jenson and Steinhaus 2015	4–17					9.8 (3.5) <sup>b</sup>	8	+1.8	
4. Joshi et al. 2017 <sup>c</sup>	6–17					4.0 (1.9) <sup>c</sup>	3.5 (1.7) <sup>c</sup>	+0.5	<i>p</i> = 0.12
5. Kentrou et al., 2019	4–18	5.7 (2.5)	7.14 (2.4) <sup>b</sup>	-1.8	<i>p</i> < 0.001				
6. Lin et al., 2014	0–18	6.2 (4.4)	7.5 (3.7)	-1.3	<i>p</i> < 0.001	8.4 (3.1)	7.4 (3.3)	+1.0	<i>p</i> < 0.001
7. Miodovnik et al., 2015	2–17	4.6 (0.2)	6.4 (0.3) <sup>b</sup>	-1.8	<i>p</i> < 0.001		5.9 (0.2) <sup>b</sup>		
8. Soke et al. 2018 <sup>d</sup>	8			-1.0	<i>p</i> ≤ 0.05				
9. Steven et al. 2016 <sup>e</sup>	6–17	4.7 (2.7)	6.3 (3.2)	-1.6	<i>p</i> < 0.001				
10. Wallisch et al. 2018 <sup>e</sup>	3–6	4.5	5.0	-0.7	<i>p</i> < 0.001	4.9	5.0	-0.1	
11. Wang et al., 2018 (The Netherlands)	6–14	5.0 (2.2)	5.2 (2.0)	-0.2					
Wang et al. (China)	6–14	3.3 (1.1)	3.3 (1.2)	0.0					
12. Wei et al., 2021	0–17	5.9	6.9 (3.0) <sup>b</sup>	-1.8	<i>p</i> < 0.001		6.7 (2.8) <sup>b</sup>		

Note: The whole sample included all children diagnosed with autism including ASD only and ASD + ADHD. <sup>a</sup> Mean difference for the age of ASD and ADHD diagnosis was calculated by subtracting the age of diagnosis for the ASD + ADHD group from the age of diagnosis of the ASD/ADHD only group, or the whole sample. <sup>b</sup> Two data points combined using weighting for the sample because data in the original study was split by gender or order of diagnosis (e.g. ADHD before ASD/ADHD same/after ASD). <sup>c</sup> Mean age of onset for the diagnoses. <sup>d</sup> Only the mean difference was provided. <sup>e</sup> Months converted to years

2015). The average age of ADHD diagnosis when children had co-occurring ASD ranged from 4.8 years (Hatakenaka et al., 2016) to 8 years (Jensen & Steinhausen, 2015). In the ASD + ADHD group, the average age of ADHD diagnosis was between 0.1 (Wallisch et al., 2020) and 1.8 years (Jensen & Steinhausen, 2015) earlier compared to the ADHD only group.

### Results of Possible Contributing Factors to Age of Diagnosis

In the included studies, some factors were quantifiably measured in comparison to the co-occurring group and the single diagnosis. Some studies included the influence of these factors in general on the age of diagnosis but did not report the impact separately on a co-occurring group, so are not included in the results section. The factors that were quantifiably measured in comparison to a co-occurring group included age of parental first concerns, diagnostic wait times, order of co-occurring diagnosis, presence of intellectual disability (ID), and the effect of gender on the age of diagnosis.

#### Age of First Parental Concerns

Two studies compared the timing of when parents first had concerns for their children. Stevens et al. (2016) found that parents in the ASD + ADHD group first expressed concerns that their child might have ASD at 2.8 years compared to 2.4 years in the ASD only group, a statistically significant difference ( $p < 0.001$ ). Stevens et al. (2016) also investigated whether there was a significant difference between parents' first concern and when medical assistance was sought and found that help seeking was significantly later for the ASD + ADHD group than the ASD only group ( $p < 0.001$ ). (Wang et al., 2018) found that parents in the ASD + ADHD group first expressed concerns that their child might have ASD at 2.4 years in China and 3.4 years in the Netherlands, compared to 2.5 years in China and 3.2 years in the Netherlands in the whole sample ( $p < 0.05$ ). Joshi et al. (2017) was the only study that examined the age of parents' first ADHD concern. They found that parents in the ASD + ADHD group first expressed concern that their child might have ADHD at 3.5 years compared to 4.0 years for the ADHD only group, which was not statistically significant.

#### Diagnostic Wait Times

Two studies examined the difference in the time between the first visit to a practitioner to the time at which the child is diagnosed. Hatakenaka et al. (2016) found that the ASD + ADHD group waited 5 months longer than the ASD only group for a diagnosis of ASD after the first visit, and

the ADHD only group waited 8 months longer than the ASD group for a diagnosis (no statistical analysis performed). Stevens et al. (2016) found that the ASD + ADHD group waited 1.3 years longer than the ASD only group for a diagnosis after first seeking medical assistance, however, statistical analysis was carried out for the age of first concern to the age of diagnosis and age of first concern to age first medical assistance sought but not medical assistance sought to the age of diagnosis.

#### Order of Co-occurring Diagnosis

Miodovnik et al. (2015) and Wei et al. (2021) found that age of ASD diagnosis was significantly later in children with ASD + ADHD for whom ADHD was diagnosed first: 8.6 years (Miodovnik et al., 2015) and 8.5 years (Wei et al., 2021), in comparison to those who had their ADHD diagnosed at the same time or after their ASD diagnosis: 4.7 years (Miodovnik et al., 2015) and 5.5 years (Wei et al., 2021) ( $p < 0.001$  for both studies).

#### Presence of ID

Stevens et al. (2016) found that an additional diagnosis of intellectual disability (ID) led to a reduction in the age of diagnosis of ASD when co-occurring ADHD was present from 6.3 years to 5.4 years. The ASD diagnosis for the co-occurring with ID was still significantly later than an ASD diagnosis of a child with ASD + ID (3.7 years) or ASD only (4.7 years) ( $p < 0.001$ ). Hatakenaka et al. (2016), Jensen and Steinhausen (2015) Miodovnik et al. (2015), Soke et al. (2018), Wei et al. (2021), and Wallisch et al. (2020) discuss ID or developmental delays in relation to the age of diagnosis or diagnosis under 6 years, but not in relation to the co-occurring group. Joshi et al. (2017) exclude children with an IQ under 70.

#### Gender

Kentrou et al. (2019) found that being female-led to an increase in the age of ASD diagnosis compared to being male. These gender differences in the age of ASD diagnosis were exacerbated with the co-occurring group being diagnosed 1.5 years later in males and 2.6 later in females compared to gender differences in the ASD only group (Kentrou et al., 2019) ( $p < 0.001$ ).

### Discussion

The primary aim of this systematic literature review was to examine the difference in age of ASD and ADHD diagnosis for an ASD + ADHD group in comparison to children with



only one of these diagnoses. The 12 articles in this review supported the hypothesis that there is a significant delay in ASD diagnosis with co-occurring ADHD. The delay in age of ASD diagnosis ranged between 0.7 years and 1.8 years compared to an ASD only group. The reverse trend was also apparent with children over 6 years, with ADHD being diagnosed earlier when co-occurring ASD was present. The earlier age of ADHD diagnosis for children over 6 years ranged between 1 and 1.8 years compared with an ADHD only group. Quantifiable factors that may have impacted the timing of these diagnoses included slightly later parental first concerns for the ASD + ADHD group compared to ASD only and slightly earlier compared to an ADHD only group. Children with co-occurring ASD + ADHD and their families also waited longer for a diagnosis. Age of diagnosis may also be affected if they received their ADHD diagnosis before the ASD diagnosis or if the child is female. The impact of later parental concerns, gender, and order of diagnosis, must be interpreted with caution due to a scarcity of research looking at these factors in relation to the age of diagnosis across these diagnostic groups.

The inverse relationship, where an ADHD diagnosis is made 12 to 20 months earlier when ASD is present over the age of 6 years, has not been a focus of the research literature (Lin et al., 2014). The three studies that mentioned this earlier diagnosis of ADHD attributed this finding to greater symptom severity of ASD + ADHD and earlier developmental identification associated with ASD symptoms (Engelhard et al., 2020; Jensen & Steinhausen, 2015; Lin et al., 2014). Elwin et al. (2020) demonstrated that when ASD co-occurred with ADHD, ADHD symptoms were quantifiably more severe than in an ADHD only population. Therefore, this severity might indicate that the ADHD-symptom profile is more apparent to caregivers and leads to earlier diagnosis. In the study by Joshi et al. (2017), however, the ASD + ADHD group did not look different from an ADHD only group in terms of severity. In the two studies that included a population under 6 years, the difference in age of ADHD diagnosis was not apparent (Hatakenaka et al., 2016; Wallisch et al., 2020). This might be due to the ADHD diagnosis being given simultaneously with ASD and the inclusion of participants only under the age of 6 years when ADHD is usually diagnosed after the age of 6 (Visser et al., 2014). The effect of ID on the age of diagnosis of a co-occurring group also has mixed results. Stevens et al. (2016) was the only study to statistically report an earlier diagnosis effect on a co-occurring group with and without ID; however, there is some evidence that ID has the reverse or no statistically significant effect on the age of diagnosis across a combined ASD group (Miodovnik et al., 2015; Wei et al., 2021).

Delays in diagnoses can occur at various points along the clinical pathway, from identification of early atypical

development to health-seeking behaviour and a diagnostic assessment (Becerra-Culqui et al., 2018; Yamauchi et al., 2015). Two studies in the systematic review showed that the co-occurring group had slightly later parental concerns than the ASD only group (Joshi et al., 2017; Stevens et al., 2016; Wang et al., 2018). Stevens et al. (2016) also showed that there was a delay between parents' first concerns and when they sought medical assistance. Although beyond the scope of this literature review, one possible explanation is the nature of that parental concern. There was some evidence that the ASD + ADHD group have parental concerns which are more likely to be behavioural in nature rather than the key signs of early ASD diagnosis, such as language delay, social concerns, or fixed and rigid behaviour (Wallisch et al., 2020). Parents may not report a behavioural concern to their child's diagnostician until after the behaviours are seen as developmentally inappropriate, typically past the age of 6 years and when the child is in a school setting, which further delays health seeking (Yamauchi et al., 2015; Zablotzky et al., 2017). The parental concerns of ASD + ADHD children more closely resemble parental concerns of an ADHD group than an ASD only group (Wallisch et al., 2020). This fits with Joshi et al.'s (2017) results which suggest that ADHD with co-occurring ASD does not look significantly different in clinical manifestation from ADHD alone in measures such as type, number of symptoms, and diagnostic subtypes or even age of onset. An important future research direction would clarify patterns of parent referral concerns in relation to wait times to diagnosis.

After parental concerns and health-seeking comes the clinicians' role and subsequent diagnosis (Becerra-Culqui et al., 2018; Yamauchi et al., 2015). Greater wait times between first seeking medical assistance and wait time to diagnosis was reported between 0.4 and 1.3 years later than an ASD only group than an ASD + ADHD group (Hatakenaka et al., 2016; Stevens et al., 2016). There is evidence that children with ASD + ADHD see a higher number of professionals prior to diagnosis and spend longer on waitlists prior to getting a diagnosis (Eggleston et al., 2019). ADHD symptoms may also mask ASD symptoms during clinic assessments. (Gipson et al., 2015; Soke et al., 2018; Stevens et al., 2016). Nomura et al. (2014) studied children with ASD at age 5 years who had shown signs of ADHD and found that hyperactivity and inattentiveness were apparent during a short visit, but subsequent parental home reports and observations during school revealed more ASD symptoms (Nomura et al., 2014). It has also been suggested that "search satisfying" occurs, whereby a diagnostician finds one diagnosis that fits, and this leads to the delay in the recognition of ASD symptoms and diagnosis (Gipson et al., 2015; Soke et al., 2018). This fits with the findings that the delay in ASD diagnosis increases when the population of ASD + ADHD children is divided into children that

received their ADHD diagnosis first and those that received their ASD diagnosis first or at the same time as their ADHD diagnosis (Kentrou et al., 2019; Miodovnik et al., 2015; Wei et al., 2021). Kentrou et al. (2019) only included children in the ASD + ADHD group if they had been diagnosed with ADHD first. Therefore, it is suggested that once a diagnosis is given, diagnosticians and parents may not seek further diagnostic clarification. An important future research direction would examine the role of the diagnostician in possible delays, including the different professional groups of diagnosticians, which might use different diagnostic methodologies. Future research might also consider the impact of specialised and non-specialised professionals in diagnostic delays and possible amelioration of this delay with specific training in diagnosing autism and co-occurring conditions.

There is also a well-known delay in identifying females on the spectrum, perhaps due to better camouflaging of social skills (Lai et al., 2017; Wood-Downie et al., 2020), but this delay is compounded by the addition of ADHD as it could be argued that the presentation further complexifies. Evidence for this complexity was given by Kentrou et al. (2019), who showed that the delayed ASD diagnosis with co-occurring ADHD stretched out a further year when the child was female in comparison with male children with ASD + ADHD. The more complex a diagnostic picture, and the less typical symptom profile, the more difficult the diagnosis (Avlund et al., 2020; Smith et al., 2019).

There were methodological issues with the studies included within this review. The assessment of bias (Hoy et al., 2012) demonstrated the most common areas of risk were a nationally representative sample, direct data collection and case definition. High-risk in the latter two categories reflected a different methodology whereby parents were interviewed or surveyed. The age of diagnosis was recalled rather than contemporaneously noted in medical records, and parents defined the case definition by affirming that their child had been given a diagnosis. However, there is a potential limitation with medical records too, whereby different procedures for diagnosing ASD and ADHD within and between countries and different diagnosticians responsible for diagnosing might impact the age of diagnosis. The fallibility of memory might limit the comparison across studies that used medical records (Kentrou et al., 2019; Miodovnik et al., 2015; Stevens et al., 2016; Wang et al., 2018). Miodovnik et al. (2015) address this concern citing both ADHD research showing parent-reported survey data was similar in estimates to insurance data (Visser et al., 2013) and equally consistent in parent-reported autism diagnosis with two nationally representative surveys (Control and Prevention, 2006), concluding that there is convergent validity between parent-reported diagnosis of autism and ADHD and other forms of data collection. However, this remains a limitation even if the consistency across studies suggests that whether

recalled or collected contemporaneously in medical records, there is a delay in autism diagnosis when ADHD is present.

In addition, the two studies from the Netherlands and the two from Taiwan each drew from the same nationally representative database; the Netherlands Autism Registrar and Taiwan National Health Insurance database, respectively. It is possible that the same participant pools were used across these studies, thus reducing the true participation numbers. A further limitation was the lack of reporting around subtypes (inattentive, hyperactive, or combined) of ADHD present with ASD. Of the 12 studies, only Joshi et al. (2017) differentiated subtypes of ADHD co-occurring with ASD, and this might be an additional factor to consider in future research on the age of diagnosis. In addition, some studies controlled for differences in age of diagnosis, such as adjusting for child sex, race/ethnicity, maternal education and study site, which reduced the delayed age of diagnosis for the ASD + ADHD group by one month compared to unadjusted diagnosis delay of 1 year (Soke et al., 2018). Other studies did not control for these possible differences, which might have impacted the age of diagnosis.

The current review was limited as the inclusion criteria specified articles would be published after 2014 to minimise difficulties posed by the DSM restrictions on dual diagnosis of ASD + ADHD prior to 2013. Some articles published after 2014 included a data set range that was collected prior to 2013. However, diagnosis of co-occurring ADHD with autism was prevalent before 2014 because of the ethical responsibility of clinicians to provide treatment for ADHD. Due to research and specifically effective interventions for ADHD, clinicians have had the advice that they should make the comorbid diagnosis when ADHD symptoms reach clinical significance (Goldstein & Schwebach, 2004). It is difficult to speculate on how much impact the DSM-5 had on the restriction of a co-occurring diagnosis in these earlier populations. It is likely that increased acceptance of a co-occurring diagnosis will result in an increased diagnosis of more moderate ADHD symptoms with autism, but the same trends in diagnosis were seen pre- and post-2014.

Another consideration is that studies can report the age of diagnosis as an incidental finding, and although the literature review search terms were broad, it is possible that an article was missed. Furthermore, we used Hoy et al. (2012) as a risk of bias, and another indicator tool might have produced different results.

## Implications

The findings of this review suggest that professionals should be aware of the delay in ASD diagnosis with co-occurring ADHD. A short visit with clear ADHD symptoms and parental concerns about behaviour should not rule out the presence of ASD, and an ADHD diagnosis

should not preclude further ASD investigation. This review also highlights that children with ASD + ADHD with relatively equal “severity” of symptoms do not appear to present early to the clinic compared to children with ASD only. This diagnostic picture can be further complicated by the child being female or having been diagnosed with ADHD first, which might create diagnostic overshadowing. A delayed ASD diagnosis is not unique to a geographical location. Therefore, being aware of this phenomenon will help reduce delayed and missed diagnoses. The studies examining contributing factors to this delay are few, but greater evidence of these factors would mean implications for screening and assessment. Children showing factors associated with a delayed diagnosis might be subject to earlier screening and more in-depth assessment.

There are several important directions for ASD + ADHD research. Further examination of factors that may impact the age of diagnosis is warranted, such as which ASD symptoms might be more apparent in short clinic visits. A greater detailed study of the pathway from parental concerns to health-seeking and diagnosis would help to identify which, if any, of these steps contributes most to delayed ASD diagnosis when ADHD is present. Further research on the impact of factors such as later parental concerns, gender, order of diagnosis, and intellectual disability is also required to corroborate the results of the few studies which investigated these factors. More research into the effect of ASD on early ADHD diagnosis would also be illuminating, particularly as severity and earlier developmental ASD symptoms do not appear to be adequate explanations of an earlier diagnosis of ADHD. Another area for consideration in understanding delayed diagnoses is an examination of delayed or missed adult diagnoses.

The findings of this review, that ASD is typically diagnosed later when ADHD symptoms are present and ADHD is typically diagnosed earlier when ASD is present, are of concern. This is because an early diagnosis of ASD is essential in enabling the provision of early intervention. A more complex diagnostic picture with less obvious key symptoms for a diagnosis of ASD might lead to longer waiting periods between the first clinic visit and an eventual diagnosis. Further research is needed to understand the weight and importance of each factor in the delay of an ASD diagnosis and the earlier diagnosis of ADHD when the two conditions co-occur.

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## Declarations

**Conflict of Interest** The authors declare no competing interests.

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