



How has DSM-5 Affected Autism Diagnosis? A 5-Year Follow-Up Systematic Literature Review and Meta-analysis

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Abstract

We conducted a 5-year follow-up systematic review and meta-analysis to determine change in frequency of autism spectrum disorder (ASD) diagnosis since diagnostic and statistical manual 5 (DSM-5) publication and explore the impact of Social Communication Disorder (SCD). For 33 included studies, use of DSM-5 criteria suggests decreases in diagnosis for ASD [20.8% (16.0–26.7), $p < 0.001$], DSM-IV-TR Autistic Disorder [10.1% (6.2–16.0), $p < 0.001$], and Asperger's [23.3% (12.9–38.5), $p = 0.001$]; pervasive developmental disorder-not otherwise specified decrease was not significant [46.1% (34.6–58.0), $p = 0.52$]. Less than one-third [28.8% (13.9–50.5), $p = 0.06$] of individuals diagnosed with DSM-IV-TR but not DSM-5 ASD would qualify for SCD. Findings suggest smaller decreases in ASD diagnoses compared to earlier reviews. Future research is needed as concerns remain for impaired individuals without a diagnosis.

Keywords Autism Spectrum Disorder · DSM-5 · Diagnosis · Asperger's Disorder · PDD-NOS · Social Communication Disorder

Introduction

Autism Spectrum Disorder (ASD) was first established as a unique diagnosis from schizophrenia in 1980 in the Third Edition of the Diagnostic and Statistical Manual of Mental

Disorders (DSM)—the clinical diagnostic standard for mental disorders, including development disorders. Prior to 1980, the prevalence of autism estimated both in the United States (US) and globally ranged from 0.07 to 0.31 (Treffert 1970) to 0.49 (Wing and Gould 1979) per 1000 children. When the DSM, Fourth Edition, Text-Revision (DSM-IV-TR) was published in 2000 (American Psychiatric Association 2000), data from the first surveillance year (2000) of the Autism and Developmental Disabilities Monitoring (ADDM) Network estimated an ASD prevalence rate of 6.7 per 1000 or 1 in 150 children aged 8 years (Rice and Autism and Developmental Disabilities Monitoring Network Surveillance Year 2000 Principal Investigators 2007), a finding similar to that reported by Mattila et al. in a study of Finnish children (Mattila et al. 2011). The most recent estimate from the ADDM Network (2014) illustrates a further increase in prevalence to 16.8 per 1000 or 1 in 59 American children (Baio et al. 2018) and is consistent with estimates of the increase in diagnosis rate obtained by parent self-report via national surveys (Kogan et al. 2018; Schieve et al. 2006). While estimates by country and the methods by which they are derived may vary, the increasing prevalence of autism as a global issue clear (Adak and Halder 2017; Elsabbagh et al. 2012; Fombonne et al. 2009; Levy et al. 2009). Collectively,

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this has prompted public health concerns, an expansion of research efforts, and a continued need for services (Baio et al. 2018).

Changes in the criteria for autism diagnosis published in the Fifth Edition of the DSM (DSM-5) (American Psychiatric Association 2013a) have stimulated much debate. First, the DSM-IV-TR contained ASD subtypes of Autistic Disorder (AD), Asperger's Disorder, and pervasive developmental disorder-not otherwise specified (PDD-NOS) that were omitted in the DSM-5; instead, subtypes were collapsed into a single diagnostic category—ASD. The DSM-5 also reduced the core domains of impairment from three to two: (1) social interaction and social communication (previously two distinct categories of “social interaction” and “communication”) and (2) restricted, repetitive patterns of behavior, interests, or activities. In addition, while the DSM-IV-TR contained 12 distinct diagnostic criteria, the DSM-5 outlines only seven which are more general principles and behaviors. Finally, the DSM-5 allows for inclusion of historical behaviors in the ASD criteria, with the caveat that these behaviors must have been present in the early developmental period, while the previous edition was limited to current behaviors. Overall, these changes have caused concern that a higher threshold of symptoms is required for DSM-5 ASD diagnosis, thereby failing to capture some individuals who would have previously been diagnosed with ASD under the DSM-IV-TR and who may benefit from access to treatment and services (Maenner et al. 2014). Notably, while ADDM Network data on autism rates released just prior to publication of the DSM-5 identified a prevalence of 1 in 88 children aged 8 years old (Centers for Disease Control and Prevention 2012), the most recent ADDM Network prevalence estimate since DSM-5 publication was 1 in 59 children (Baio et al. 2018). However, data for this latest report are from 2014, and children included in this analysis would have primarily been evaluated under DSM-IV-TR ASD criteria (Baio et al. 2018). Therefore, the impact of DSM-5 criteria on ASD diagnosis rates remains unknown.

To date, three systematic literature reviews (one with a meta-analysis) which examined the potential impact of DSM-5 on ASD diagnosis rates have been published; two were conducted just prior to DSM-5 publication (Kulage et al. 2014; Sturmey and Dalfern 2014), and one was conducted a year after (Smith et al. 2015). All three determined that ASD rates could decrease by at least one-third. While numerous studies have quantified potential changes in ASD rates in the last 5 years, no new systematic literature reviews with meta-analyses have been conducted to synthesize data from studies comparing DSM-IV-TR and DSM-5 ASD rates. In addition, the impact of a new DSM-5 diagnosis, Social Communication Disorder (SCD)—defined as a primary deficit in social communication and interaction (SCI) without restrictive, repetitive behaviors (RRB) (Ohashi et al. 2015;

Sumi et al. 2014; Swineford et al. 2014)—on ASD rates has not been specifically examined in a systematic review since DSM-5 publication. This is an important gap in the literature because not only must an ASD diagnosis be “ruled out” before an SCD diagnosis can be given, but SCD was also initially described by the American Psychiatric Association as potentially capturing individuals with symptoms of PDD-NOS but who would no longer meet criteria for ASD under DSM-5 (American Psychiatric Association 2013b).

To address these gaps on the impact of DSM-5 on ASD diagnosis rates, the aims of this follow-up systematic literature review and meta-analysis were to: (1) determine the change in frequency of ASD diagnosis in the first five years after publication of the revised DSM-5 ASD criteria; (2) identify the DSM-IV-TR autism subtypes most affected by the new criteria; and (3) assess the potential of an alternative diagnosis of SCD for individuals who meet DSM-IV-TR but not DSM-5 ASD diagnostic criteria.

Methods

Search Strategy and Inclusion Criteria

We followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al. 2009) in conducting this literature review and meta-analysis. An a priori protocol was registered (PROSPERO 2017 CRD42017077533) in November 2017 and updated in October 2018; the protocol can be accessed from http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42017077533. We used Covidence (<http://covidence.org>), the web-based production platform for Cochrane Reviews, to manage our work flow. On October 26, 2017, we searched MEDLINE (PubMed), the Cumulative Index to Nursing and Allied Health Literature (EBSCO), Education Resources Information Center (ProQuest), and PsycInfo (Ovid) for original studies published from April 1, 2013, the end of coverage of the first literature review on this topic, through December 31, 2017. Subsequently, we re-ran the search on July 11, 2018 for studies published between January 1, 2018 and June 30, 2018. For search terms, two main domains were combined with the AND operator: one relating to DSM-5 and the other to autism diagnoses (e.g., Asperger's) or other related diagnoses (e.g., SCD). The full search strategy by database is available online in Appendix 1. Both subject headings and free text were used. No language requirement was placed on the text. To supplement the database search, we hand-searched issues of the *Review Journal of Autism and Developmental Disorders* and conference proceedings of the International Society for Autism Research from 2013 to 2017. We conducted a grey literature search for conference proceedings in both BIOSIS and

Embase and examined .gov and .org sites for seven pages of search results on Google.com.

All items found in the literature during the identification phase were screened by at least two authors who examined titles and abstracts for two inclusion criteria: studies needed to (1) present original data and (2) compare application of DSM-IV-TR and DSM-5 ASD diagnostic criteria to populations at risk for or previously diagnosed with ASD and/or one of three DSM-IV-TR ASD subtypes (AD, Asperger's disorder, or PDD-NOS). If it was unclear whether a study met these criteria based on abstract review, we conservatively included the study for full-text review. During full-text review, at least two authors assessed each study and came to a consensus for inclusion based on the following criteria: studies needed to (1) report results as raw data or percentages of individuals meeting diagnostic criteria using both DSM-IV-TR and DSM-5 criteria separately or (2) provide sufficient information so that percentages could be calculated (for example, present DSM-5 sensitivity and specificity with DSM-IV-TR as the reference standard). We excluded studies if they (1) did not compare DSM-IV-TR and DSM-5 diagnostic criteria applied to the same population; (2) did not provide sufficient information for extracting raw data on changes in rates of ASD diagnoses under DSM-IV-TR as compared to DSM-5; (3) had been included in the first literature review and meta-analysis on this topic (Kulage et al. 2014); (4) examined a duplicate study sample; or (5) used an inappropriate study design/article type for purposes of this review (i.e., editorials, letters to the editor, case reports, review articles, qualitative studies, or summaries or press releases of another article). We then hand-searched reference lists of included studies to locate other studies that may not have been identified in the electronic search.

Data Extraction

Two authors independently extracted data from each study and four authors compared results to arrive at a consensus. We extracted the following study characteristics: continent; study design; data sources; funding information; sample size; sample demographics including gender, race, and ethnicity; number diagnosed with ASD and/or its subtypes under DSM-IV-TR criteria; the version of DSM-5 ASD diagnostic criteria used in the study (i.e., draft or final); the discipline of the rater(s) responsible for making the autism diagnosis; and the instruments used by raters. The change in frequency of ASD diagnosis when DSM-5 criteria were applied to the same sample and/or subsamples was then calculated, including number and percent reduction in diagnosis. For studies which examined SCD,

we extracted information on the number of individuals with ASD and its subtypes under DSM-IV-TR criteria who did not meet DSM-5 criteria but would qualify for an alternative diagnosis of SCD. Finally, we collected data from studies which reported specificity and sensitivity of DSM-5 diagnostic criteria.

Quality Appraisal

To rate the scientific rigor of individual studies, we used the quality appraisal of reliability studies (QAREL) (Lucas et al. 2010) which was developed for use in systematic reviews and meta-analyses to assess the quality of studies which explore diagnostic reliability. This 11-item checklist examines seven principles including the appropriateness of subjects, qualification of examiners, examiner blinding, ordering of examination, suitability of the time interval between repeated measurements, appropriate test application and interpretation, and statistical analysis of intra or inter-rater agreement. Each QAREL item can be answered with "yes," "no," or "unclear," with five items also including "not applicable" as an option. When raters agree upon the interpretation of criteria for each item, the QAREL has been demonstrated to be a reliable assessment tool for studies of diagnostic reliability (Lucas et al. 2013). In this study, two authors independently rated each study using QAREL, and then four authors collectively reviewed results and came to a consensus on each item.

Data Analysis

We conducted three meta-analyses. In the first pooled analysis, all included studies were examined to determine the change in frequency of ASD diagnosis based on DSM-5 criteria. For the second pooled analysis, we included studies that explored differences in ASD diagnosis by DSM-IV-TR subtype. For each, data were extracted as the number of individuals meeting DSM-IV-TR ASD diagnostic criteria and the number no longer meeting ASD diagnostic criteria under DSM-5; we then computed the proportion of those who would not retain an ASD diagnosis. Pooled effects were estimated for the proportion of individuals who no longer met criteria for ASD diagnosis using a random effects meta-analysis model. For the third meta-analysis, we pooled data from studies that examined application of DSM-5 SCD criteria to ASD samples. Specifically, we extracted the number of individuals who met DSM-IV-TR ASD criteria but no longer met criteria for an ASD diagnosis under DSM-5 and, of those, the number who would alternatively meet criteria for SCD. Because of the small number of studies, and to obtain a more comprehensive assessment of the impact of the SCD diagnosis and its potential to capture these individuals, we also extracted the same data from the four studies

that examined SCD that were included in the first review on this topic (Kulage et al. 2014). A pooled effect was estimated for the proportion of individuals who would meet criteria for SCD. Results are presented as forest plots using random effects meta-analysis models.

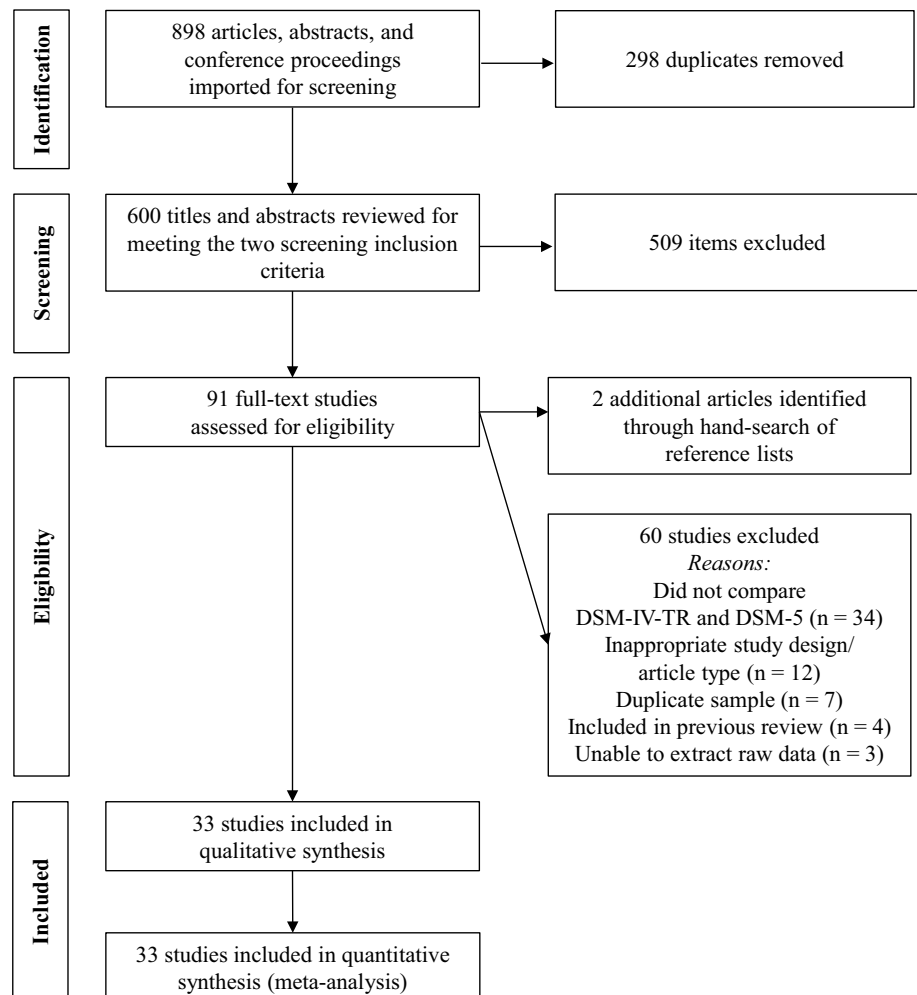
For pooled effects indicative of a statistically significant reduction ($p < 0.05$) in diagnoses when DSM-5 criteria were applied, we examined heterogeneity and publication bias. Heterogeneity was assessed using Cochran's Q and I^2 statistics and was considered to be present if the Cochran's Q p -value was < 0.05 or I^2 was $> 50\%$ (Higgins et al. 2003). To examine differences between studies that might explain heterogeneity, we conducted subgroup analyses by sample age; continent where the study was conducted; study design; instrument used to make an ASD diagnosis; discipline of the rater (MD, PhD, or both) responsible for making the diagnosis; version of DSM-5 ASD diagnosis criteria used (draft or final); study funding source; and three risk of bias domains: whether order of examination varied, measurement of intra and/or interrater agreement, and whether raters making the diagnosis were blinded to the results of the reference

standard (i.e., DSM-IV-TR diagnosis). To examine the risk of publication bias, we constructed a funnel plot, examined it visually, and conducted a Classic fail-safe N test, which is used to determine the number of additional studies needed to change interpretation of publication bias (Persaud 1996). Data were analyzed using Comprehensive Meta-Analysis statistical software (Biostat, Inc., Englewood, NJ).

Results

Figure 1 presents details of the literature search. A total of 898 records were initially identified from the database and supplemental search phases; following removal of duplicates, 600 articles were deemed eligible for screening. After screening titles and abstracts, 509 items were excluded, leaving an initial group of 91 studies for full-text assessment. However, prior to full-text assessment, the reference lists of the 91 studies were hand-searched, and two additional publications were identified, creating a total of 93 for full-text review. Sixty studies were subsequently

Fig. 1 PRISMA flow diagram for the systematic literature review



excluded after the full-text review, including seven which used the same sample as a study (Matson et al. 2012) that was included in the first review on this topic (See Appendix 2 online for list of excluded references and rationale for exclusion). Therefore, a total of 33 studies were included in the systematic review and meta-analysis; of these, 19 studies that examined ASD subtypes and nine studies that examined SCD (five studies identified in this review and four studies from the previous review) were eligible for the additional analyses.

Study Quality

Figure 2 summarizes the results of the quality appraisal of the 33 studies. All but one study (Kim et al. 2014) used an appropriate sample of subjects. In the majority of studies, appropriately credentialed raters provided diagnoses, correctly applied and interpreted the instruments or criteria for diagnoses, and employed an appropriate time-interval between DSM-IV-TR and DSM-5 measurement. However, only eight studies (Baio et al. 2018; Helles et al. 2015; Hiller et al. 2014; Kim et al. 2014; Mazurek et al. 2017; Mugzach et al. 2015; Taheri et al. 2014; Young and Rodi 2014) reported inter and/or intra-rater reliability, and variation in the order of examination could only be verified in three studies (Mazurek et al. 2017; Mugzach et al. 2015; Young and Rodi 2014). The risk for bias in relation to study blinding

across studies was largely unclear. Only one study specified that raters were blinded to the findings of other raters (Wong and Koh 2016), and no studies definitively indicated that raters were blinded to their own prior findings. In addition, in only four studies could we determine that the raters were blinded to both clinical information and additional cues not part of the diagnosing process (Mazurek et al. 2017; Taheri et al. 2014; Turygin et al. 2013; Wong and Koh 2016), and only three studies reported that raters were blinded to the results of DSM-IV-TR when applying DSM-5 criteria (Magana and Vanegas 2017; Sung et al. 2018; Wong and Koh 2016).

Characteristics of the Included Studies

Study Year, Type, and Continent

Table 1 provides a descriptive summary of each study. Publication years for articles ranged from 2013 to 2018 with the majority (61%) published in the 2 years immediately following the release of the DSM-5. Fifty-five percent ($n = 18$) of studies were prospective (Barton et al. 2013; Beighley et al. 2014; Dawkins et al. 2016; Helles et al. 2015; Jashar et al. 2016; Konst et al. 2014; Magana and Vanegas 2017; Mazurek et al. 2017; Ocakoglu et al. 2015; Romero et al. 2016; Signorelli et al. 2015; Sumi et al. 2014; Sung et al. 2018; Tartaglia et al. 2017; van Steensel et al. 2015; Wheeler

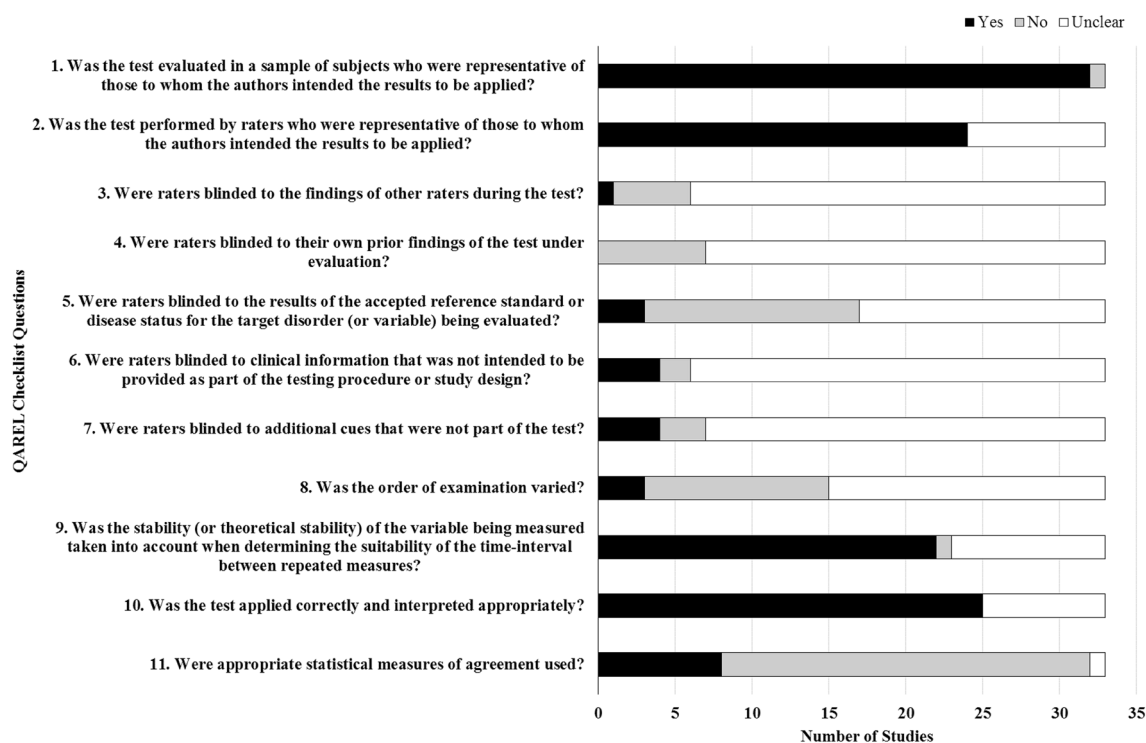


Fig. 2 Study quality appraisal results using the QAREL checklist

Table 1 Characteristics of 33 included studies

Author, location, study type, data sources, and funding sources	Sample characteristics (i.e., number, age, gender, race, ethnicity)	Clinician type and diagnostic instruments	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	Reduction in diagnoses using DSM-5 criteria
Baio et al. (2018)	N = 4920 Age 8 years	MD and PhD/PsyD ASD case determination criteria for DSM-IV-TR and DSM-5	4658 ASD ^a	4236 ASD	9.1% ASD
US					
Retrospective					
State vital records for 11 ADDM network sites					
Federal funding					
Barton et al. (2013)	N = 422 Ages 16.79–39.36 months	MD and PhD/PsyD	284 ASD	239 ASD ^b	15.8% ASD
US	76.1% male	ADI-R			
Prospective, cross sectional	69.9% white; 10.4% black	ADOS			
Toddlers with siblings with ASD from pediatric offices in Atlanta, Connecticut, and bordering states; Connecticut early intervention program		M-CHAT-R			
Federal funding					
Beighley et al. (2014)	8.3% Hispanic; 6.4% not reported; 3.1% Asian/Pacific Islander; 1.9% multi-racial	Not reported	135 ASD	51 ASD	62.2% ASD
US	N = 261	DSM-IV-TR/ICD-10 Checklist			
Prospective, cross sectional	Ages 16–87 years				
Louisiana state developmental center	53.6% male				
No funding reported	76.6% white, 23% black, 0.4% Hispanic				
Christiansz et al. (2016)	N = 185	PhD/PsyD	126 ASD	106 ASD	15.9% ASD
Australia	Ages 20–55 months and 1.7–4.6 years	ADI-R	103 AD	90 AD	12.6%
Retrospective	83.2% male	ADOS	23 PDD-NOS	16 PDD-NOS	30.4%
Early childhood services, pediatricians, and public regional autism assessment programme					
Federal funding					
Dawkins et al. (2016)	N = 183	PhD/PsyD	142 ASD	134 ASD	5.6% ASD
US	Ages 1–62 years	ADOS-2			
Prospective, cross sectional	78.6% male	CARS2-HF			
TEACCH Autism Program Centers in North Carolina	61.2% white, 20.9% black, 7.7% Hispanic	CARS2-ST			
No funding reported					
Foley-Niepon et al. (2017)	N = 45	Not reported	45 ASD	45 ASD	0% ASD
US	Ages 5.5–17.8 years	ADI-R	16 AD	16 AD	0% AD
Retrospective	75.6% male	ADOS	17 Asperger's	17 Asperger's	0% Asperger's
University center psychology clinic for gifted and talented students			12 PDD-NOS	12 PDD-NOS	0% PDD-NOS
Federal funding					
Harstad et al. (2015)	N = 227	MD and PhD/PsyD	156 ASD	120 ASD ^b	23.1% ASD

Table 1 (continued)

Author, location, study type, data sources, and funding sources	Sample characteristics (i.e., number, age, gender, race, ethnicity)	Clinician type and diagnostic instruments	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	Reduction in diagnoses using DSM-5 criteria
US	Ages 1.3–18 years	ADOS-2	114 AD	103 AD	9.6% AD
Retrospective	83.7% male	Bayley Scales, 3rd edition	5 Asperger's	3 Asperger's	40.0% Asperger's
Multidisciplinary developmental behavioral pediatric clinic		DAS-II	37 PDD-NOS	14 PDD-NOS	62.2% PDD-NOS
No funding reported					
Helles et al. (2015)	N = 50	MD and PhD/PsyD	39 ASD	31 ASD	20.5% ASD
Sweden	Ages 23–43 years	ASDI	9 AD	9 AD	0% AD
Prospective	100% male	DISCO-II	22 Asperger's	20 Asperger's	9.1% Asperger's
Gothenburg Child Neuropsychiatric Clinic		Gillberg's Criteria for Asperger's Syndrome	8 PDD-NOS	2 PDD-NOS	75.0% PDD-NOS
Non-federal funding					
Hillier et al. (2014)	N = 114	Not reported	114 ASD	87 ASD	23.7% ASD
Australia	Mean ages: Male = 8.76 ± 3.91 years Female = 8.06 ± 4.03 years	ADI-R			
Retrospective	55.3% male	ADOS			
Private practice specializing in diagnostic assessments for PDDs		ADEC			
Non-federal funding		CARS			
Jashar et al. (2016)	N = 281	MD or PhD/PsyD	203 ASD	146 ASD	28.1% ASD
US	Ages 16–39 months	M-CHAT	134 AD	116 AD	13.4% AD
Prospective	77.9% male	VABS	69 PDD-NOS	30 PDD-NOS	56.5% PDD-NOS
Pediatrician offices	76.5% white, 6.4% black, 10.3% Hispanic				
Federal funding					
Kim et al. (2014)	N = 292	MD and PhD/PsyD	206 ASD	184 ASD	10.7% ASD
South Korea	Ages 7–12 years	ADI-R	114 AD	112 AD	1.8% AD
Retrospective file review		ADOS	34 Asperger's	31 Asperger's	8.8% Asperger's
All children born from 1993 to 1999 in a suburb of Seoul, South Korea		BASC II-PRS	58 PDD-NOS	41 PDD-NOS	29.3% PDD-NOS
Federal and non-federal funding					
Konst et al. (2014)	N = 1722	PhD/PsyD	1104 ASD	605 ASD	45.2% ASD
US	Ages 17–37 months	BISCUIT – Part I			
Prospective, cross sectional	71.5% male	BISCUIT – Part II			
State-funded early intervention program for children at-risk for developmental disability	49.2% white, 38.2% black				
No funding reported	10.3% other, 2.3% Hispanic				
Maenner et al. (2014)	N = 7597	Not reported	6577 ASD	5339 ASD	18.8% ASD
US	Age 8 years	Not reported			
Retrospective	82.3% male				
ADDM Network, 11 sites in 2006 and 14 sites in 2008	55.8% white, 22.5% black; 12.2% Hispanic, 2.9% Asian/Pacific Islander				

Table 1 (continued)

Author, location, study type, data sources, and funding sources	Sample characteristics (i.e., number, age, gender, race, ethnicity)	Clinician type and diagnostic instruments	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	Reduction in diagnoses using DSM-5 criteria
Federal and non-federal funding	0.6% other				
Magana and Vanegas (2017)	N = 29	Not reported	20 ASD	23 ASD ^c	0% ASD
US	Ages 4–16 years	ADI-R (Spanish)			
Prospective	100% Hispanic				
Clinics and parent support groups across two Midwestern cities					
Federal and non-federal funding					
Mazurek et al. (2017)	N = 439	MD and PhD/PsyD	278 ASD	249 ASD ^d	10.4% ASD
US	Ages 2–17.7 years	ABC	229 AD	222 AD	3.0% AD
Prospective	79% male	ADOS-2	25 Asperger's	20 Asperger's	20.0% Asperger's
Six autism centers affiliated with the Autism Treatment Network	78% white	CBCL	24 PDD-NOS	6 PDD-NOS	75.0% PDD-NOS
Federal and non-federal funding					
Mugzach et al. (2015)	N = 2642	Not reported	2642 ASD	2485 ASD	5.9% ASD
Country not reported		ADI-R			
Retrospective					
Data source not reported					
Federal and non-federal funding					
Ocakoglu et al. (2015)	N = 28	Not reported	28 ASD	18 ASD	37.5% ASD
Turkey	Ages birth to 6 years	ABC	28 PDD-NOS	18 PDD-NOS	37.5% PDD-NOS
Prospective	82.1% male	CARS			
Children diagnosed with PDD-NOS by Ege University Disabled Health Committee in 2010–2011					
No funding reported					
Ohashi et al. (2015)	N = 68	MD	40 ASD	27 ASD	32.5% ASD
Japan	Ages 6.2–14.9 years	PARS	3 AD	3 AD	0% AD
Retrospective	63.2% male		16 Asperger's	13 Asperger's	18.8% Asperger's
Department of Psychology and Development, Nagoya City University Hospital			21 PDD-NOS	11 PDD-NOS	47.6% PDD-NOS
No funding reported					
Rieske et al. (2015)	N = 424	Not reported	300 ASD	192 ASD ^b	36.0% ASD
US	Ages 2–18 years	DSM-IV-TR/ICD-10 Checklists			
Retrospective database review	72.9% male	ASD-CC			
Outpatient clinics, schools, and community organizations	69.1% white, 17.4% unknown; 8.3% black				
No funding reported					
Romero et al. (2016)	5.2% other	MD	123 ASD	57 ASD	53.6% ASD
Spain	N = 123	DSM-IV-TR/ICD-10 Checklists	34 AD	17 AD	50.0% AD
	Ages 5–15 years				

Table 1 (continued)

Author, location, study type, data sources, and funding sources	Sample characteristics (i.e., number, age, gender, race, ethnicity)	Clinician type and diagnostic instruments	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	Reduction in diagnoses using DSM-5 criteria
Prospective Schools in Magala, Spain	82% male 100% white	IDEA	27 Asperger's 62 PDD-NOS	14 Asperger's 26 PDD-NOS	48.1% Asperger's 58.0% PDD-NOS
No funding reported Signorelli et al. (2015)	N=15 Adults; Ages not provided	Not reported	15 ASD	3 ASD	80.0% ASD
Italy Prospective Clinic-based sample		ADI-R ADOS VABS	15 Asperger's	3 Asperger's	80.0% Asperger's
No funding reported Solerdecoll Arimany et al. (2017)	N=118 Ages 3–17 years 86.5% male	Not reported	88 ASD	77 ASD ^b	12.5% ASD
Spain Retrospective chart review		ADI-R	18 AD	16 AD	11.1% AD
Department of Child and Adolescent Psychiatry and Psychology, Institute of Neurosciences, Hospital clinic, Barcelona, Spain			47 Asperger's 23 PDD-NOS	44 Asperger's 17 PDD-NOS	6.4% Asperger's 26.0% PDD-NOS
No funding reported Sumi et al. (2014)	N=180 Ages 2–5 years 75% male	MD and PhD/PsyD PARS	64 ASD 8 AD	62 ASD 8 AD	3.1% ASD 0% AD
Japan Prospective Nagoya West District Care Center for disabled children			27 Asperger's 29 PDD-NOS	27 Asperger's 27 PDD-NOS	0% Asperger's 6.9% PDD-NOS
No funding reported Sung et al. (2018)	N=110 Ages 5.1–19.6 years 80.9% male	Not reported	92 ASD	77 ASD	16.3% ASD
Singapore Prospective The Child Guidance Clinic under the Institute of Mental Health		ADI-R ADOS-2	30 AD 32 Asperger's 30 PDD-NOS	30 AD 29 Asperger's 18 PDD-NOS	0% AD 3.3% Asperger's 40.0% PDD-NOS
No funding reported Taheri et al. (2014)	N=22 Ages 5–19 years 95.5% male	PhD/PsyD CARS VABS-II	22 ASD 16 AD 6 PDD-NOS	12 ASD ^b 11 AD 1 PDD-NOS	45.5% ASD 31.3% AD 83.3% PDD-NOS
Canada Retrospective chart review The TRE-ADD Program					
No funding reported Tartaglia et al. (2017)	N=98 Ages 3–22 years 100% male 88.8% white	MD and PhD/PsyD ADI-R ADOS	29 ASD	29 ASD	0% ASD
US Prospective Hospital-based outpatient clinics and national SCA support organizations Federal and non-federal funding					

Table 1 (continued)

Author, location, study type, data sources, and funding sources	Sample characteristics (i.e., number, age, gender, race, ethnicity)	Clinician type and diagnostic instruments	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	Reduction in diagnoses using DSM-5 criteria
Turygin et al. (2013)	N = 142 Ages 2–16 years	PhD/PsyD BASC-2	66 ASD	44 ASD ^b	33.3% ASD
US Retrospective database review	62.7% male				
University-affiliated clinic in Louisiana	76.1% white, 12% black				
No funding reported	7% Hispanic				
Van Steensel et al. (2015)	N = 90 Ages 7–17 years	Not reported ADI-R CSBQ	88 ASD	63 ASD	28.4% ASD
Netherlands Prospective	76.7% male				
Outpatient mental health centers					
No funding reported					
Wheeler et al. (2015)	N = 758 Ages 2–67 years	Not reported Survey questions; no standardized instrument	276 ASD	191 ASD	30.8% ASD
US Prospective (survey)	84.3% male				
Registry of fragile X syndrome	89.8% white, 3.8% Hispanic, 2.6% black				
Federal and non-federal funding					
Wong and Koh (2016)	N = 206 Mean age = 3 years, 10 months	MD and PhD/PsyD ADOS ADOS-2	202 ASD 174 AD 2 Asperger's 4 PDD-NOS	184 ASD 165 AD 1 Asperger's 1 PDD-NOS	8.9% ASD 5.2% AD 50.0% Asperger's 75.0% PDD-NOS
Singapore Retrospective	85.9% male				
Specialist multidisciplinary clinic for developmental concerns	67% Chinese, 18% Malay, 10% Indian, 5% other				
No funding reported					
Yaylaci and Miral (2017)	N = 150 Ages 3–15 years	MD ABC CARS	22 Non-specified 149 ASD 139 AD 4 Asperger's 6 PDD-NOS	17 Non-specified 120 ASD 120 AD 0 Asperger's 0 PDD-NOS	22.7% Non-specified 19.5% ASD 13.6% AD 100% Asperger's 100% PDD-NOS
Turkey Prospective, cross sectional	77.3% male				
Data source not reported					
No funding reported					
Young and Rodi (2014)	N = 233 Ages 1–54 years	PhD/PsyD ADI-R AQ CARS	210 ASD 76 AD 114 Asperger's 20 PDD-NOS	120 ASD ^b 56 AD 64 Asperger's 0 PDD-NOS	42.9% ASD 26.3% AD 43.9% Asperger's 100% PDD-NOS
Australia Prospective	72.5% male				
Private practice offering services by psychologists and pathologists					
No funding reported		CAST SCQ			
Zander and Bolte (2015)	N = 171 Ages 20–47 months	Not reported ABC ADI-R	127 ASD 68 AD 59 PDD-NOS	115 ASD 66 AD 49 PDD-NOS	9.4% ASD 2.9% AD 16.9% PDD-NOS
Sweden Retrospective chart review					

Table 1 (continued)

Author, location, study type, data sources, and funding sources	Sample characteristics (i.e., number, age, gender, race, ethnicity)	Clinician type and diagnostic instruments	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	Reduction in diagnoses using DSM-5 criteria
Subset from previous study sample from the Neuropsychiatric Resource Team Southeast, Division of Child and Adolescent Psychiatry, Stockholm County Council		ADOS-2			
Federal and non-federal funding		VABS-II			

^aThe abbreviation of “ASD” under DSM-IV-TR refers to group of three diagnoses under the autism spectrum: Autistic Disorder (AD), Asperger’s Disorder, and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), and “ASD” under DSM-5 refers to a diagnosis of Autism Spectrum Disorder

^bStudy used draft instead of final published DSM-5 criteria to diagnose ASD

^cThree individuals met DSM-5 but not DSM-IV-TR ASD criteria

^dOne individual met DSM-5 but not DSM-IV-TR ASD criteria

ADDM Autism and Developmental Disabilities Monitoring; *ADI-R* Autism Diagnostic Interview—revised; *ADOS* Autism Diagnosis Observation Schedule; *M-CHAT-R* Modified Checklist for Autism in Toddlers—revised; *ICD-10* international statistical classification of diseases and related health problems, 10th edition; *ADOS-2* Autism Diagnosis Observation Schedule, 2nd edition; *CARS2-HF* Childhood Autism Rating Scale, 2nd edition (high-functioning clinical tool); *CARS2-ST* Childhood Autism Rating Scale, 2nd edition (standard clinical tool); *TEACCH* Treatment and Education of Autistic and Communication related handicapped Children; *DAS-II* Differential Ability Scale, 2nd edition; *ASDI* Autism Spectrum Disorder Interview; *DISCO-II* Diagnostic Interview for Social and Communication Disorders, 2nd edition; *ADEC* Autism Detection in Early Childhood; *CARS* Childhood Autism Rating Scale; *M-CHAT* Modified Checklist for Autism in Toddlers; *VABS* Vineland Adaptive Behavior Scale; *BASC II-PRS* Behavior Assessment System for Children II—Parent Report Scale; *BISCUIT-Part I* Baby and Infant Screen for Children with a Utism Traits-Part I; *BISCUIT-Part II* Baby and Infant Screen for Children with a Utism Traits-Part II; *ABC* Autism Behavior Checklist; *CBCL* Child Behavior Checklist; *PARS* Pervasive Developmental Disorder-Autism Society Japan Rating Scale; *ASD-CC* Autism Spectrum Disorders-Comorbidity for Children; *IDEA* Autism Spectrum Disorder Inventory; *TRE-ADD* Treatment, Research, and Education for Autism and Developmental Disorders; *VABS-II* Vineland Adaptive Behavior Scale, 2nd edition; *SCA* sex chromosome aneuploidy; *BASC-2* Behavior Assessment System for Children, 2nd edition; *CSBQ* Children’s Social Behavior Questionnaire; *AQ* Autism Spectrum Quotient; *CAST* Childhood Asperger Syndrome Test; *SCQ* Social Communication Questionnaire

et al. 2015; Yaylaci and Miral 2017; Young and Rodi 2014), and the remaining were retrospective. While 16 studies were conducted in North America (15 in the US and one in Canada), the majority of studies were conducted globally, including seven in Asia (Kim et al. 2014; Ocakoglu et al. 2015; Ohashi et al. 2015; Sumi et al. 2014; Sung et al. 2018; Wong and Koh 2016; Yaylaci and Miral 2017), six in Europe (Helles et al. 2015; Romero et al. 2016; Signorelli et al. 2015; Solerdelcoll Arimany et al. 2017; van Steensel et al. 2015; Zander and Bolte 2015); and three in Australia (Christiansz et al. 2016; Hiller et al. 2014; Young and Rodi 2014); one was unreported (Mugzach et al. 2015).

Demographics

Samples were heterogeneous in terms of size, age, and data sources. Sample sizes ranged from 15 (Signorelli et al. 2015) to 7597 (Maenner et al. 2014) individuals. The majority of studies ($n = 24$) restricted their samples to pediatric populations (i.e., ages ≤ 19 years). There were six studies limited to young children under the age of five (Barton et al. 2013; Christiansz et al. 2016; Jashar et al. 2016; Konst et al. 2014; Sumi et al. 2014; Zander and Bolte 2015); eight that included all children ages ≤ 19 years (Harstad et al. 2015; Mazurek et al. 2017; Ocakoglu et al. 2015; Rieske et al. 2015; Solerdelcoll Arimany et al. 2017; Turygin et al. 2013; Wong and Koh 2016; Yaylaci and Miral 2017); and 10 studies with older children ages 5–19 years (Baio et al. 2018; Foley-Nicpon et al. 2017; Hiller et al. 2014; Kim et al. 2014; Maenner et al. 2014; Magana and Vanegas 2017; Ohashi et al. 2015; Romero et al. 2016; Taheri et al. 2014; van Steensel et al. 2015). Two studies included samples of children and adults ages ≥ 5 years (Beighley et al. 2014; Sung et al. 2018); two restricted inclusion to adults ≥ 20 years (Helles et al. 2015; Signorelli et al. 2015); four included all ages (Dawkins et al. 2016; Tartaglia et al. 2017; Wheeler et al. 2015; Young and Rodi 2014); and one did not report ages (Mugzach et al. 2015). Twenty-eight studies provided data on gender, race, and/or ethnicity of their samples. In the 27 studies which reported gender, 79.6% of the cumulative sample population was male (11,367 of 14,276). For the 16 studies which reported figures on race and/or ethnicity, 61% of the cumulative sample population was white (7926 of 12,975). Nine studies specifically indicated their populations included individuals of Hispanic ethnicity; out of a total sample population of 11,395 individuals, only 1113 (9.8%) were Hispanic.

Data Sources and Funding Sources

Studies used a wide variety of data sources; for example, prospective studies included sources such as early intervention programs and centers; pediatric offices; developmental

clinics; support groups; and organizational registries. For retrospective studies, data sources included state records (e.g., ADDM Network site records); hospital, university, and clinic records; private practices; public schools; community organizations; census records; and previous study samples. Fifteen studies reported receiving financial support from a variety of funding sources including federal (e.g., National Institutes of Health, Centers for Disease Control and Prevention) and non-federal (e.g., Autism Speaks, Simons Foundation Autism Research Initiative) entities (Baio et al. 2018; Barton et al. 2013; Christiansz et al. 2016; Foley-Nicpon et al. 2017; Helles et al. 2015; Hiller et al. 2014; Jashar et al. 2016; Kim et al. 2014; Maenner et al. 2014; Magana and Vanegas 2017; Mazurek et al. 2017; Mugzach et al. 2015; Tartaglia et al. 2017; Wheeler et al. 2015; Zander and Bolte 2015).

Diagnostic Instruments, Raters, and DSM-5 Criteria Version

The most common screening instruments used in combination with clinical impressions to diagnose ASD were the Autism Diagnostic Interview—Revised (ADI-R) and the Autism Diagnosis Observation Schedule (ADOS) with more than half of studies (55%) using either one of these or both. Other objective tools coupled with clinical impressions included a wide variety of checklists, scales, and diagnostic instruments focused on identifying and measuring autism characteristics, developmental delays, and social behavior deficiencies. Clinicians who interpreted findings of the instruments to make the diagnosis of ASD included physicians (e.g., child psychiatrists, behavioral pediatricians), psychologists (e.g., PhD and/or PsyD), and teams of physicians and psychologists. The majority of studies (78.8%) used the final published version of the DSM-5 (American Psychiatric Association 2013a) to diagnose ASD, and the 2011 draft version of the criteria (You et al. 2011) was used to diagnose ASD in the remaining studies (Barton et al. 2013; Harstad et al. 2015; Rieske et al. 2015; Solerdelcoll Arimany et al. 2017; Taheri et al. 2014; Turygin et al. 2013; Young and Rodi 2014).

Changes in ASD Diagnosis Rates since DSM-5 Publication

The percent reduction in DSM-IV-TR ASD diagnoses using DSM-5 criteria ranged from 0% (Foley-Nicpon et al. 2017; Magana and Vanegas 2017; Tartaglia et al. 2017) to 80% (Signorelli et al. 2015). Overall, 91% of studies reported ASD diagnosis reduction rates between 0 and 50% when applying DSM-5 criteria, with the majority of studies (60.6%) reporting reduction rates of 0–25% and 30.3% demonstrating reduction rates of 26–50%. Only three studies (9.1%) reported ASD diagnosis rates $> 50\%$ (Beighley et al. 2014; Romero et al. 2016; Signorelli et al. 2015); of

Table 2 Impact of Social Communication Disorder on Individuals who do not retain an ASD diagnosis under DSM-5

Study and country	DSM-IV-TR diagnoses (including subtypes)	DSM-5 diagnoses	SCD diagnoses N (% captured)
Dickerson Mayes (2013) ^a	25 ASD ^b	7 ASD ^c	5/18 (28%) ASD
US	25 PDD-NOS	7 PDD-NOS	5/18 (28%) PDD-NOS
Huerta et al. (2012) ^a	4,453 ASD	4,058 ASD ^c	75/395 (19%) ASD
US and Canada			
Kim et al. (2014)	206 ASD	184 ASD	17/22 (77%) ASD
South Korea	114 AD	112 AD	2/2 (100%) AD
	34 Asperger's	31 Asperger's	2/3 (67%) Asperger's
	58 PDD-NOS	41 PDD-NOS	13/17 (76%) PDD-NOS
Mazurek et al. (2017)	278 ASD	249 ASD ^d	2/30 (7%) ASD
US			
Ocakoglu et al. (2015)	28 ASD	18 ASD	0/10 (0%) ASD;
Turkey	28 PDD-NOS	18 PDD-NOS	0/10 (0%) PDD-NOS
Ohashi et al. (2015)	40 ASD	27 ASD	5/13 (38%) ASD
Japan	3 AD	3 AD	AD = N/A
	16 Asperger's	13 Asperger's	2/3 (67%) Asperger's
	21 PDD-NOS	11 PDD-NOS	3/10 (30%) PDD-NOS
Sumi et al. (2014)	64 ASD	62 ASD	2/2 (100%) ASD
Japan	8 AD	8 AD	AD = N/A
	27 Asperger's	27 Asperger's	Asperger's = N/A
	29 PDD-NOS	27 PDD-NOS	2/2 (100%) PDD-NOS
Taheri and Perry (2012) ^a	129 ASD	82 ASD ^c	2/47 (4%) ASD
Canada			
Wilson et al. (2013) ^a	80 ASD	61 ASD ^c	12/19 (63%) ASD
Europe			

SCD social communication disorder; N/A not applicable

^aStudy included in previous literature review

^bThe abbreviation of "ASD" under DSM-IV-TR refers to group of three diagnoses under the autism spectrum: Autistic Disorder (AD), Asperger's Disorder, and Pervasive Developmental Disorder-not otherwise specified (PDD-NOS), and "ASD" under DSM-5 refers to a diagnosis of Autism Spectrum Disorder

^cStudy used draft instead of final published DSM-5 criteria to diagnose ASD

^dOne participant met DSM-5 but not DSM-IV-TR ASD criteria

note, the highest reduction rate of 80% was in a sample of 15 individuals, all of whom were adults (Signorelli et al. 2015).

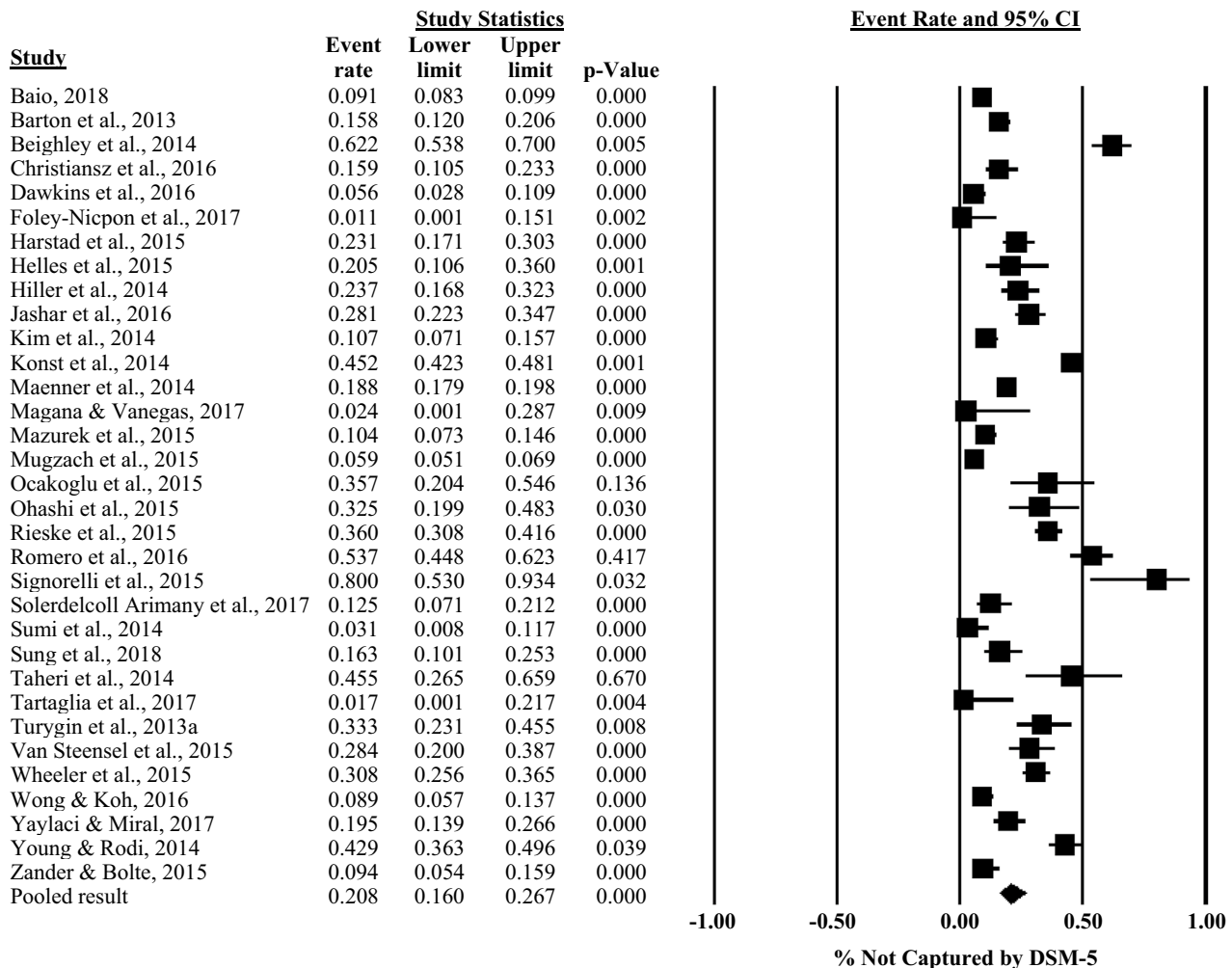
DSM-IV-TR Subtypes most affected by DSM-5 ASD Criteria

Nineteen studies (57.5%) reported data on changes in ASD diagnosis under DSM-5 criteria according to one or more of the DSM-IV-TR ASD subtypes, and the reduction rates in ASD diagnosis varied widely by subtype. In the 17 studies that examined AD, reduction rates of $\leq 25\%$ were demonstrated in the vast majority of studies (82.4%) with the remaining reporting reduction rates of 26–50%. For the 14 studies that looked at Asperger's, the reduction rates were more equally spread with 57.1% of studies reporting reduction rates of ≤ 25 and 42.9% of studies reporting reduction rates $\geq 26\%$. Of note, Signorelli et al. (2015) reported a reduction rate in Asperger's of 80% and Yaylaci and Miral (2017) reported a reduction rate of 100%. Highest overall

reduction rates were seen for the PDD-NOS subtype. Only 16.7% of the eight studies which examined PDD-NOS saw ASD diagnosis reduction rates of $\leq 25\%$. The majority of studies (66.6%) reported PDD-NOS reduction rates in the 26–75% range with the remaining three studies (16.7%) finding reduction rates $> 75\%$, two of which reported a 100% reduction rate (Yaylaci and Miral 2017; Young and Rodi 2014).

Impact of DSM-5 Social Communication Disorder (SCD) Diagnosis

Table 2 provides details on the five studies from the current review (Kim et al. 2014; Mazurek et al. 2017; Ocakoglu et al. 2015; Ohashi et al. 2015; Sumi et al. 2014) and four studies from the first review (Dickerson Mayes et al. 2013; Huerta et al. 2012; Taheri and Perry 2012; Wilson et al. 2013) that examined the proportion of individuals



Random effects model, $p < 0.001$; Cochran's $Q = 1454.9$, $p < 0.001$, $I^2 = 97.8$

Fig. 3 Forest plots of the 33 studies included studies representing the proportion of individuals who met criteria for an Autism Spectrum Disorder (ASD) diagnosis under DSM-IV-TR but not for DSM-5 ASD. Squares represent effect sizes of individual studies with

extended lines denoting 95% confidence intervals. Sizes of squares indicate the weight of each study based on sample size using random effects analysis. The diamond represents the estimated pooled effect size

with DSM-IV-TR ASD who did not retain an ASD diagnosis under DSM-5 but alternatively met SCD criteria. Only three studies utilized US populations (Dickerson Mayes et al. 2013; Huerta et al. 2012; Mazurek et al. 2017). Five studies examined the impact of SCD on DSM-IV-TR ASD subtypes (Dickerson Mayes et al. 2013; Kim et al. 2014; Ocakoglu et al. 2015; Ohashi et al. 2015; Sumi et al. 2014). Individuals qualifying for an alternative SCD diagnosis included 2/2 (100%) for the AD subtype; 4/6 (66.7%) for the Asperger's Disorder subtype; and 23/57 (40.4%) for the PDD-NOS subtype.

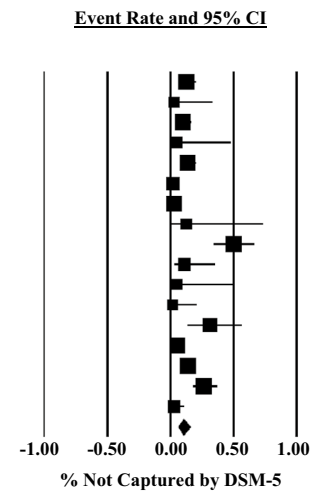
DSM-5 Sensitivity and Specificity

Seven studies reported the sensitivity and specificity of DSM-5 diagnostic criteria with ADI-R and/or ADOS. Of three studies that used both the ADI-R and ADOS (Barton et al. 2013; Christiansz et al. 2016; Sung et al. 2018), sensitivity and specificity values ranged from 0.84 to 0.93 and 0.54 to 0.83, respectively. For two studies that used the ADI-R alone (Magana and Vanegas 2017; Solderdelcoll Arimany et al. 2017), the sensitivity range was reported between 0.88 and 0.90 while the specificity range was between 0.57 and 0.86. The remaining two studies used the ADOS alone (Dawkins et al. 2016; Mazurek et al. 2017); the

Fig. 4 Forest plots of Autistic Disorder (top), Asperger's Disorder (middle), and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) (bottom) representing the proportion of individuals who met criteria for diagnosis under DSM-IV-TR criteria but not for DSM-5 Autism Spectrum Disorder. Squares represent effect sizes of individual studies with extended lines denoting 95% confidence intervals. Sizes of squares indicate the weight of each study based on sample size using random effects analysis. The diamond represents the estimated pooled effect size

Autistic Disorder

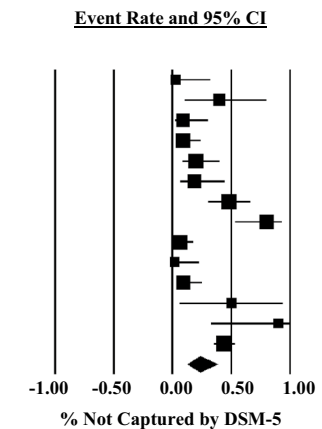
Study	Event rate	Study Statistics		p-Value
		Lower limit	Upper limit	
Christiansz et al., 2016	0.126	0.075	0.205	0.000
Foley-Nicpon et al., 2017	0.029	0.002	0.336	0.015
Harstad et al., 2015	0.096	0.054	0.166	0.000
Helles et al., 2015	0.050	0.003	0.475	0.042
Jashar et al., 2016	0.134	0.086	0.203	0.000
Kim et al., 2014	0.018	0.004	0.067	0.000
Mazurek et al., 2015	0.031	0.015	0.063	0.000
Ohashi et al., 2015	0.125	0.007	0.734	0.198
Romero et al., 2016	0.500	0.338	0.662	1.000
Solerdelcoll Arimany et al., 2017	0.111	0.028	0.352	0.006
Sumi et al., 2014	0.056	0.003	0.505	0.052
Sung et al., 2018	0.016	0.001	0.211	0.004
Taheri et al., 2014	0.313	0.136	0.567	0.144
Wong & Koh, 2016	0.052	0.027	0.096	0.000
Yaylaci & Miral, 2017	0.137	0.089	0.204	0.000
Young & Rodi, 2014	0.263	0.177	0.373	0.000
Zander & Bolte, 2015	0.029	0.007	0.110	0.000
Pooled result	0.101	0.062	0.160	0.000



Random effects model, $p < 0.001$; Cochran's $Q = 90.9$, $p < 0.001$, $I^2 = 82.4$

Asperger's Disorder

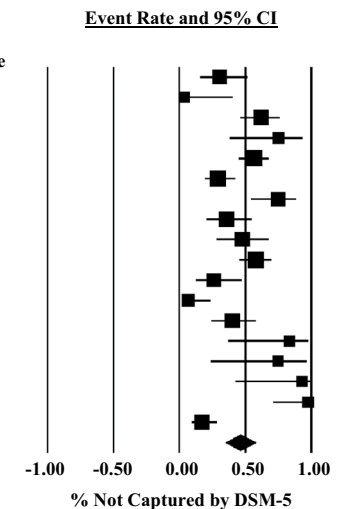
Study	Event rate	Study Statistics		p-Value
		Lower limit	Upper limit	
Foley-Nicpon et al., 2017	0.028	0.002	0.322	0.013
Harstad et al., 2015	0.400	0.100	0.800	0.657
Helles et al., 2015	0.091	0.023	0.300	0.002
Kim et al., 2014	0.088	0.029	0.240	0.000
Mazurek et al., 2015	0.200	0.086	0.400	0.006
Ohashi et al., 2015	0.188	0.062	0.447	0.022
Romero et al., 2016	0.481	0.304	0.664	0.847
Signorelli et al., 2015	0.800	0.530	0.934	0.032
Solerdelcoll Arimany et al., 2017	0.064	0.021	0.180	0.000
Sumi et al., 2014	0.018	0.001	0.230	0.005
Sung et al., 2018	0.094	0.031	0.254	0.000
Wong & Koh, 2016	0.500	0.059	0.941	1.000
Yaylaci & Miral, 2017	0.900	0.326	0.994	0.140
Young & Rodi, 2014	0.439	0.351	0.531	0.191
Pooled result	0.233	0.129	0.385	0.001



Random effects model, $p = 0.001$; Cochran's $Q = 65.4$, $p < 0.001$; $I^2 = 80.1$

PDD-NOS

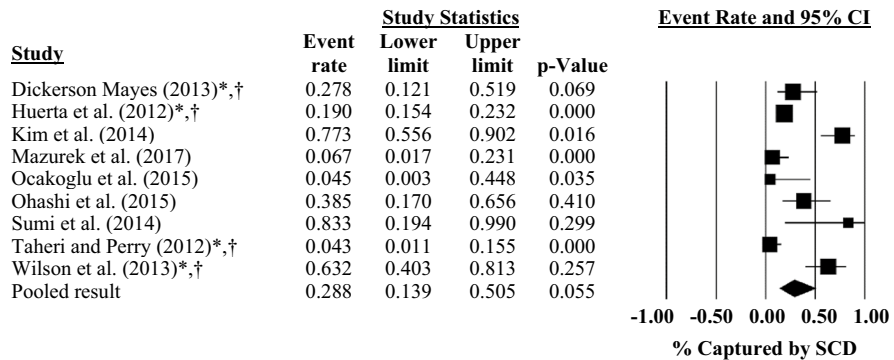
Study	Event rate	Study Statistics		p-Value
		Lower limit	Upper limit	
Christiansz et al., 2016	0.304	0.153	0.515	0.068
Foley-Nicpon et al., 2017	0.038	0.002	0.403	0.026
Harstad et al., 2015	0.622	0.458	0.761	0.143
Helles et al., 2015	0.750	0.377	0.937	0.178
Jashar et al., 2016	0.565	0.447	0.677	0.280
Kim et al., 2014	0.293	0.191	0.422	0.002
Mazurek et al., 2015	0.750	0.544	0.883	0.020
Ocakoglu et al., 2015	0.357	0.204	0.546	0.136
Ohashi et al., 2015	0.476	0.279	0.682	0.827
Romero et al., 2016	0.581	0.455	0.696	0.206
Solerdelcoll Arimany et al., 2017	0.261	0.122	0.472	0.028
Sumi et al., 2014	0.069	0.017	0.238	0.000
Sung et al., 2018	0.400	0.243	0.581	0.277
Taheri et al., 2014	0.833	0.369	0.977	0.142
Wong & Koh, 2016	0.750	0.238	0.966	0.341
Yaylaci & Miral, 2017	0.929	0.423	0.996	0.081
Young & Rodi, 2014	0.976	0.713	0.999	0.009
Zander & Bolte, 2015	0.169	0.094	0.287	0.000
Pooled result	0.461	0.346	0.580	0.521



Random effects model, $p = 0.52$; Cochran's $Q = 80.3$, $p < 0.001$; $I^2 = 78.8$

Fig. 5 Forest plot of Social Communication Disorder (SCD) representing the proportion of individuals who met criteria for an Autism Spectrum Disorder (ASD) diagnosis under DSM-IV-TR criteria but not for DSM-5 and instead met the criteria for an alternative diagnosis of SCD. Squares represent effect sizes of individual studies with extended lines denoting 95% confidence intervals. Sizes of squares indicate the weight of each study based on sample size using random effects analysis. The diamond represents the pooled effect size

Social Communication Disorder



Random effects model, $p = 0.06$; Cochran's $Q = 57.5$, $p < 0.001$; $I^2 = 86.1$

* Study included in previous literature review.

† Study used draft instead of final version of published DSM-5 criteria to diagnose ASD.

sensitivity range was 0.89 to 1.00 and the specificity range was 0.71 to 0.99.

Quantitative Synthesis

Results of the meta-analyses are provided in Figs. 3, 4, and 5. Data from 33 studies which examined changes in DSM-IV-TR ASD diagnosis when DSM-5 criteria were applied were pooled and represent data from 18,648 individuals. Using a random effects model, the pooled proportion suggests a 20.8% [95% confidence interval (CI) 16.0–26.7, $p < 0.001$] reduction in ASD diagnoses (Cochran's $Q = 1454.9$, $p < 0.001$; $I^2 = 97.8$) when DSM-5 criteria were applied (Fig. 3).

Figure 4 presents the pooled analyses that examined DSM-IV-TR diagnoses of AD, Asperger's Disorder, and PDD-NOS when DSM-5 criteria were applied. Nineteen of 33 studies examined these subtypes: AD was examined in 17 studies with data representing 1285 individuals; Asperger's Disorder was examined in 14 studies with data representing 387 individuals; and PDD-NOS was examined in 18 studies with data representing 519 individuals. Pooled effects suggest statistically significant reductions in ASD diagnoses of 10.1% (95% CI 6.2–16.0, $p < 0.001$) for those with AD (Cochran's $Q = 90.9$, $p < 0.001$, $I^2 = 82.4$) and 23.3% (95% CI 12.9–38.5, $p = 0.001$) for those with Asperger's Disorder (Cochran's $Q = 65.4$, $p < 0.001$, $I^2 = 80.1$) when DSM-5 criteria were applied. The reduction in diagnoses for PDD-NOS was not statistically significant [46.1% (95% CI 34.6–58.0), $p = 0.52$] (Cochran's $Q = 80.3$, $p < 0.001$; $I^2 = 78.8$). For all models, heterogeneity was greater than expected by chance alone.

Figure 5 provides the pooled analysis that examined the number of individuals who met DSM-IV-TR ASD diagnosis but would not meet DSM-5 criteria and instead would

qualify for an alternative diagnosis of SCD; these include data from nine studies representing 556 individuals. While the finding did not achieve statistical significance, the pooled effect suggests that less than one-third [28.8% (95% CI 13.9–50.5), $p = 0.06$] of those who met DSM-IV-TR ASD diagnostic criteria but not DSM-5 would meet SCD diagnostic criteria. Heterogeneity was greater than expected by chance alone (Cochran's $Q = 57.5$, $p < 0.001$, $I^2 = 86.1$). Although four of the studies that examined the impact of SCD used the draft version of DSM-5 ASD diagnostic criteria, there were no statistical differences between those and the five studies which used the final version of the criteria.

Subgroup Analyses

Table 3 presents results of subgroup analyses for ASD and the AD and Asperger's subtypes. Of 10 variables explored, six were found to contribute to heterogeneity: age group (all models); continent where study was conducted (ASD); instruments administered to make the diagnosis (AD); clinician who made the diagnosis (all models); study funding sources (ASD and AD); and one risk of bias criterion – measures of intra and inter-rater agreement (ASD).

Publication Bias

Figure 6 displays the funnel plot representing differences in the proportion of those diagnosed with ASD using DSM-IV-TR versus DSM-5 criteria for all studies. The open circles indicate each of the 33 individual studies. The upper portion of the funnel plot displays symmetry. The three circles on the lower left side represent studies with small sample sizes and do not represent a major concern. Findings of the Classic fail-safe N test suggests

Table 3 Subgroup analyses

Variable	All studies		Autistic Disorder		Asperger's Disorder	
	# Studies	Pooled result (%) (95% CI)	# Studies	Pooled result (%) (95% CI)	# Studies	Pooled result (%) (95% CI)
Study sample age^{a,b,c}						
Young children <5 years	6	17.2 (8.7, 31.3)	4	10.6 (6.3, 17.1)	1	1.8 (0.1, 23.0)
Young (<5 years) and older children (5–18 years)	8	20.3 (13.2, 30.0)	5	7.5 (4.1, 13.1)	5	29.2 (9.9, 61.0)
Children (5–19 years)	10	21.7 (14.5, 31.1)	5	13.2 (2.7, 45.5)	5	15.4 (5.3, 37.1)
Children and adults	2	36.4 (6.6, 82.3)	1	1.6 (0.1, 21.1)	--	--
Adults only	2	49.2 (6.2, 93.4)	1	5.0 (0.3, 47.5)	2	39.2 (1.7, 96.0)
All ages	4	18.7 (8.3, 36.9)	1	26.3 (17.7, 37.3)	1	43.9 (35.1, 53.1)
Age not reported	1	5.9 (5.1, 6.9)	--	--	--	--
Continent^a						
North America	16	21.6 (14.9, 30.2)	5	9.8 (4.5, 20.0)	3	19.2 (6.2, 46.1)
Europe	6	29.2 (13.9, 51.2)	4	11.9 (1.9, 49.1)	4	29.0 (6.7, 70.0)
Asia	7	15.9 (10.1, 24.2)	5	5.7 (2.3, 13.4)	6	16.9 (6.1, 39.0)
Australia	3	26.4 (13.3, 45.7)	2	18.7 (8.6, 35.8)	1	43.9 (35.1, 53.1)
Not reported	1	5.9 (5.1, 6.9)	--	--	--	--
Study design						
Prospective	18	25.5 (18.5, 33.9)	7	12.8 (6.0, 25.1)	8	31.2 (15.8, 52.2)
Retrospective	15	16.8 (12.2, 22.7)	9	8.1 (4.6, 13.8)	6	13.6 (6.3, 27.1)
Instruments^b						
ADI-R and ADOS	9	16.4 (10.8, 24.0)	5	3.9 (1.3, 11.5)	4	17.4 (2.6, 62.8)
ADI-R	5	14.9 (4.3, 40.9)	2	21.1 (9.6, 40.1)	2	19.8 (2.2, 72.8)
ADOS	4	11.0 (6.0, 19.4)	3	5.5 (2.8, 10.4)	3	25.8 (13.2, 44.1)
Other	14	29.8 (18.6, 44.2)	6	19.6 (10.1, 34.8)	5	24.1 (6.9, 57.4)
Not reported	1	18.8 (17.9, 19.8)	--	--	--	--
Clinician type^{a,b,c}						
MD	3	34.0 (15.2, 59.8)	3	24.8 (6.4, 61.2)	3	44.6 (15.9, 77.4)
PhD/PsyD	6	28.1 (17.6, 41.7)	3	21.3 (11.8, 35.3)	1	43.9 (35.1, 53.1)
MD or PhD/PsyD	1	28.1 (22.3, 34.7)	1	13.4 (8.6, 20.3)	--	--
Both MD and PhD/PsyD	9	11.9 (8.7, 16.1)	6	4.8 (2.8, 8.2)	6	15.0 (7.2, 28.9)
Not reported	14	22.7 (15.1, 32.7)	4	4.6 (1.9, 10.7)	4	16.0 (2.1, 62.4)
DSM-5 criteria version						
Draft	7	28.0 (19.6, 38.4)	4	18.1 (9.3, 32.5)	3	24.9 (5.8, 63.8)
Final	26	18.9 (13.8, 25.4)	13	7.8 (4.1, 14.1)	11	22.6 (10.4, 42.4)
Funding source^{a,b}						
Both federal and non-federal	8	11.3 (6.4, 19.0)	3	2.7 (1.5, 4.9)	2	14.2 (6.2, 29.4)
Federal only	5	14.3 (7.8, 24.8)	3	12.8 (9.1, 17.5)	1	2.8 (0.2, 32.2)
Non-federal only	2	22.9 (16.9, 30.2)	1	5.0 (0.3, 47.5)	1	9.1 (2.3, 30.0)
No funding reported	18	28.8 (21.9, 36.8)	10	15.1 (8.2, 26.1)	10	31.4 (16.5, 51.5)
Risk of bias						
<i>Blinded to reference standard</i>						
Low risk	3	11.1 (5.9, 19.7)	2	4.9 (2.6, 8.9)	2	18.3 (2.7, 64.9)
Unclear risk	16	22.2 (13.8, 33.7)	7	12.0 (7.1, 19.5)	5	26.3 (10.4, 52.4)
High risk	14	21.5 (15.2, 29.6)	8	9.1 (3.0, 24.6)	7	20.5 (6.5, 49.1)

Table 3 (continued)

Variable	All studies		Autistic Disorder		Asperger's Disorder	
	# Studies	Pooled result (%) (95% CI)	# Studies	Pooled result (%) (95% CI)	# Studies	Pooled result (%) (95% CI)
<i>Order of examination varied</i>						
Low risk	2	22.9 (4.6, 64.8)	2	9.7 (1.0, 53.8)	2	32.7 (13.9, 59.4)
Unclear risk	20	20.9 (15.0, 28.3)	8	10.6 (8.0, 14.1)	6	22.7 (8.8, 47.3)
High risk	11	19.5 (10.6, 33.0)	7	8.6 (2.0, 29.7)	6	17.7 (4.5, 49.4)
<i>Statistical measures of agreement^a</i>						
Low risk	8	17.1 (10.1, 27.4)	5	8.6 (2.2, 28.2)	4	18.9 (6.7, 42.8)
Unclear risk	1	36.0 (30.8, 41.6)	—	—	—	—
High risk	24	22.1 (16.7, 28.6)	12	10.8 (6.4, 17.6)	10	25.8 (10.5, 50.8)

MD physicians (e.g., child psychiatrists, behavioral pediatricians); *PhD/PsyD* psychologists; *Both MD and PhD/PsyD* teams of physicians and psychologists

^aVariable contributing to heterogeneity ($p < 0.05$) in all studies

^bVariable contributing to heterogeneity ($p < 0.05$) in Autistic disorder

^cVariable contributing to heterogeneity ($p < 0.05$) in Asperger's disorder

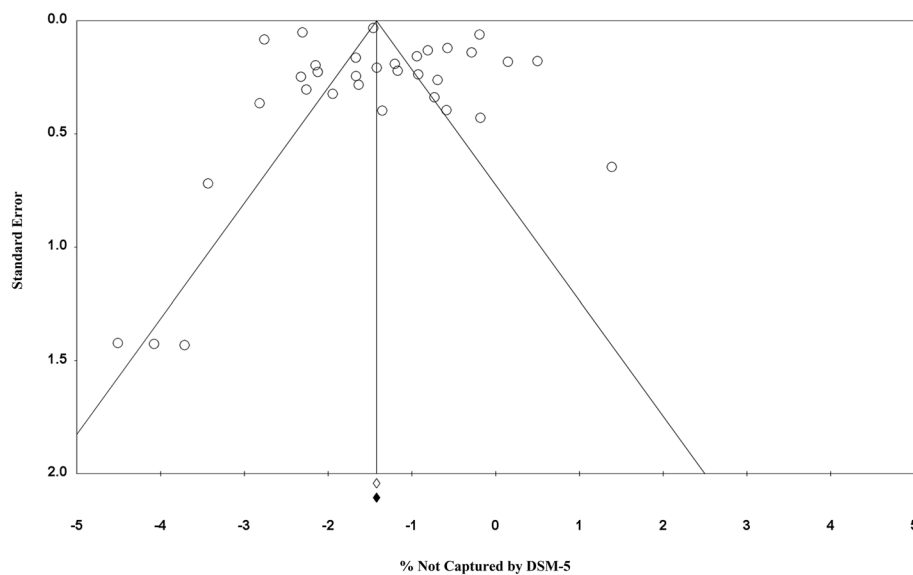


Fig. 6 Funnel plot represents differences in proportion of those diagnosed with ASD using DSM-5 versus DSM-IV-TR criteria. Plot shows the standard error of the difference in proportion (Y axis) versus the reported percent not captured by DSM-5 (X axis) using a random effects model. The vertical line indicates the pooled effect estimate. The open circles indicate each of the 33 individual studies

included in the meta-analysis. The open diamond indicates the pooled effect size and 95% confidence interval for meta-analysis, and the filled diamond indicates pooled effect size and 95% confidence interval when missing studies suggested by publication bias analysis are included

that an additional 7765 studies would need to be added to significantly change the pooled effect. Funnel plots for the subtypes AD and Asperger's Disorder are found online in Appendix 3; findings of the Classic fail-safe N test suggest that an additional 1455 and 135 studies, respectively, would need to be added to significantly change the pooled

effect. The funnel plot for SCD is found online in Appendix 4; findings of the Classic fail-safe N test suggests that an additional 89 studies would need to be added to significantly change the pooled effect. The filled circle represents a study estimated to be missing from the analysis.

Discussion

Current Study Findings

Despite advances in understanding pathophysiology in ASD, it remains a behaviorally defined clinical syndrome. As such, the diagnosis is often based on several variables including the parental historical presentation of concerns, demonstration of such behaviors during evaluations, clinical providers' experience, rating instruments, and final determination based on clinically agreed upon diagnostic guidelines set forth by the DSM. Revisions in updated DSM classification may change an individual's diagnosis. In reviewing studies published in the five years since publication of the DSM-5, which has more stringent criteria required for an ASD diagnosis, our study findings indicate that a significant number of individuals who qualified for a DSM-IV-TR ASD diagnosis would not meet DSM-5 criteria. With more than one-fifth of individuals with notable SCI difficulties coupled with disruptive RRBs who will no longer qualify for an ASD diagnosis, clinicians, researchers, and public health officials need to recognize that there are individuals lacking a diagnosis but remain in need of services. Early diagnosis and intensive treatment has been linked to improvement across many domains in autism (Reichow et al. 2018; Rogers 2016; Salomone et al. 2016; Schreibman et al. 2015); however, a recent study examining treatment patterns of ASD among children using nationally representative data found that nearly 30% of US children with ASD are not receiving behavioral or medication treatment (Xu et al. 2018). A variety of therapies provided by the board of education and insurance carriers are often limited based upon an ASD diagnosis and/or clearly defined developmental delays (Candon et al. 2018; Turcotte et al. 2016). Acknowledging their need for treatment, clinicians may be providing ASD diagnoses in addition to other comorbidities, which are common in children with ASD, notably attention-deficit hyperactivity disorder (ADHD), obsessive compulsive behaviors, mood disorders, sensory processing issues, or anxiety (Belardinelli et al. 2016; Ford 2014; Soke et al. 2018).

ADDM Network data also continue to demonstrate that ASD prevalence rates are rising even with tightened DSM-5 diagnostic criteria. If true positive diagnoses are actually increasing, parental awareness and acceptance, less stigmatization, better trained clinicians, more thorough data collection methods, and even increasing genetic tendencies could be contributing factors. In addition, comorbid diagnoses are now allowable for ASD under DSM-5, enabling clinicians to give multiple comorbid diagnoses of intellectual disability, ASD, and ADHD, which could also explain why ASD rates have continued to rise since publication of the DSM-5.

It is notable that findings from this current systematic literature review and meta-analysis indicate a smaller decrease in ASD diagnoses when comparing DSM-IV-TR to DSM-5 as compared to all earlier reviews. Additionally, all studies which examined DSM-IV-TR ASD subtypes were also found to have smaller decreases in ASD diagnosis when comparing DSM-IV-TR to DSM-5 as compared with the first review. This may be because clinicians now have a greater comfort level with interpreting DSM-5 criteria. It could also indicate that fewer individuals are failing to receive an ASD diagnosis than what previous studies anticipated. Nevertheless, these findings do show that approximately one in five individuals who would have received an ASD diagnosis under DSM-IV-TR would not receive a diagnosis under DSM-5 with only a minority being alternatively captured by SCD. Most recent ADDM Network data show a continued increase in prevalence of ASD; however, the majority of children included in the last data reported from surveillance year 2014 were diagnosed under DSM-IV-TR criteria (Baio et al. 2018). It will be important to examine the next release of ADDM Network data on autism rates, which is anticipated to be based solely on children diagnosed with DSM-5 criteria; considering the findings of our meta-analyses, we would predict there may be a decrease in autism rates reported. Regardless of whether ASD prevalence rates are on an upward or downward trend, the potential numbers of individuals who may have been previously eligible for a DSM-IV-TR diagnosis of ASD but would not qualify under DSM-5 as reported by this study remains alarming and points to a need for continued research on this topic.

Autism remains a behaviorally defined clinical disorder set forth by a multitude of clinicians experienced in caring for this population. These clinical criteria remain diagnostic despite the emergence of biomarkers in blood (Smith et al. 2018) and saliva (Hicks et al. 2018) samples, in addition to neuroimaging (Bi et al. 2018; Li et al. 2018; Shen et al. 2018; Zhao et al. 2018) and electrophysiological (Levin et al. 2017; Muhle et al. 2018; Righi et al. 2014) profiles. Moreover, the use of diagnostic tools to support or refute ASD diagnosis are often created and validated in homogeneous autism cohorts, such as male-dominant groups (Halladay et al. 2015). There is increasing awareness that females are likely being under- or misdiagnosed with ASD for numerous reasons, including ascertainment bias, differential presentation with more SCI deficits and less RRBs, and a role for a female protective effect which may alter the endophenotype (Goldman 2013; Jacquemont et al. 2014; Lai et al. 2015; Volkmar et al. 1993). Moreover, autism is being recognized and accepted in black, Hispanic, and other non-Caucasian individuals (Baio et al. 2018; Singh and Bunyak 2018).

Another question remains regarding who should assign the autism diagnosis. An individual may see a medical doctor, including a psychiatrist, developmental pediatrician, or

neurologist, or they may see a psychologist. The use of different tools may aid in diagnosis. Interestingly, where both MDs and PhD/PsyDs were involved in the diagnosis there was the lowest decrease in ASD diagnosis rates between DSM-IV-TR and DSM-5. This would suggest a multidisciplinary evaluation may have more specificity in initial diagnosis than a single provider. An earlier diagnosis is crucial to identify the need for early intensive behavioral interventions which have been proven as the mainstay of ASD treatment (Dawson 2013; Orinstein et al. 2014; Weitlauf et al. 2014).

Findings of Other Prior Reviews and Meta-Analyses Versus Current Study Findings

The change in ASD diagnostic criteria with introduction of the DSM-5 has been of great interest to the public as well as clinicians and researchers. Three prior systematic literature reviews have studied the impact of the changes in DSM-5 ASD diagnosis criteria on autism rates (Kulage et al. 2014; Smith et al. 2015; Sturmey and Dalfern 2014). Table 4 summarizes the findings of these previous systematic reviews in comparison to the current study. All prior reviews were published within a period of less than two years after publication of the DSM-5 with 56% of the included studies being duplicative at the time of the third review (Smith et al. 2015). While general findings were consistent across studies, the estimated reduction in ASD rates under DSM-5 criteria varied widely across included studies, ranging from 7 to 62%. Only one previous study included a meta-analysis, reporting a pooled decrease of 31% in ASD across studies (Kulage et al. 2014). The current five-year follow-up study includes a large number of studies published since April 2013 with only nine being duplicative of articles included in previous reviews. Comparing current study findings for estimated ASD reduction to the first review, the pooled decrease is smaller (20.8% vs. 31%) but remains a concern.

The number of studies included in the three previous systematic literature reviews which examined the impact of the DSM-5 diagnostic criteria on DSM-IV-TR ASD subtypes ranged from five to 13 studies. Across reviews, findings were consistent that the most affected subtype would be PDD-NOS, followed closely by Asperger's Disorder, with AD being the least impacted. Comparing current study findings for estimated reductions in diagnoses by subtype with that of the first review, reductions are less for AD (10.1% vs. 22%) and Asperger's (23.3% vs. 70%); while statistical significance was not achieved, the reduction for PDD-NOS was also less than previously reported (46.1% vs. 70%) (Kulage et al. 2014). Again, this trend may be reflected in the next release of ADDM Network data (Baio et al. 2018).

Social Communication Disorder

In the first review on this topic, 4 of 14 studies (29%) examined the impact of SCD and its potential to capture individuals with a DSM-IV-TR ASD diagnosis but who would not receive a DSM-5 ASD diagnosis (Kulage et al. 2014). Based on its intended purpose, it is surprising that five years later only five studies captured in the current review examined the potential impact of SCD; we expected to find a substantially higher number of studies exploring the impact of this new DSM-5 diagnosis. Importantly, when examining all nine studies that looked at SCD diagnoses, less than one-third (28.8%) of individuals who did not retain their ASD diagnosis under DSM-5 criteria would qualify for an SCD diagnosis. This is concerning and provides the only data combining results from multiple studies in the literature to date that SCD does not seem to be fulfilling its purpose as a "catch all" or alternative diagnosis for individuals who would have had an ASD diagnosis under DSM-IV-TR but not under DSM-5 criteria. Surprisingly, the PDD-NOS subtype—which was originally targeted by the SCD diagnosis—seems to be the subtype least likely to obtain an alternative SCD diagnosis (only 40% captured); however, across studies that examined DSM-IV-TR subtypes, the subtype sample sizes were small, limiting the scope of this finding. Discussion points in the studies which examined SCD emphasized two themes. Aligning with the results of this study, although SCD was originally described as an alternative diagnosis for individuals with symptoms of PDD-NOS but who would no longer have an autism diagnosis under DSM-5 criteria, it does not seem to be capturing a significant number of these individuals (Dickerson Mayes et al. 2013; Huerta et al. 2012; Mazurek et al. 2017; Ocakoglu et al. 2015; Wilson et al. 2013). Second, the few individuals who would receive SCD as an alternative diagnosis did not meet DSM-5 ASD criteria because of insufficient deficiencies in the RRB domain required for an ASD diagnosis (Huerta et al. 2012; Kim et al. 2014; Ohashi et al. 2015; Sumi et al. 2014; Taheri and Perry 2012).

Considering these findings, although limited, further research is clearly needed to evaluate the impact of SCD as a diagnosis and the degree to which it captures individuals who fail to meet DSM-5 ASD criteria, particularly across DSM-IV-TR subtypes and for individuals with significant impairment imposed by RRBs. Currently, the need for SCD to function as an alternative diagnosis for ASD is unclear; while some studies have indicated that an SCD diagnosis could serve as another means of obtaining required treatment and services (Greaves-Lord et al. 2013; Kim et al. 2014; Ohashi et al. 2015), others have questioned this possibility (Dickerson Mayes et al. 2013; Smith et al. 2015). The inherent overlap in diagnostic criteria for ASD and SCD poses challenges for its recognition and use as a distinct disorder

Table 4 Summary of findings of systematic reviews examining the effects of DSM-5 criteria on the number of individuals diagnosed with ASD

Study	Study type	No. of articles included in prior reviews (%)	No. of duplicate articles included in prior reviews (%)	% Reduction in ASD diagnoses under DSM-5	No. of articles that included DSM-IV-TR ASD subtypes	% Reduction in ASD diagnoses by subtypes
Kulage et al. (2014)	Systematic Review with Meta-Analysis	14	N/A	31% (95% CI 20–44) pooled decrease across studies, $p = 0.006$	7	Pooled results: 22% AD, $p < 0.001$ 70% Asperger's, $p = 0.38$ 70% PDD-NOS, $p = 0.01$
Sturmeijer and Dalferen (2014)	Systematic Review	12	9 (75%)	36.97% median overall reduction across studies; range = 7% – 54% Pooled analysis not conducted	5	19.35% median reduction in more impaired group (i.e., AD); range = 0% – 26.3% 71.27% median reduction in less impaired group (i.e., Asperger's and PDD-NOS); range = 16.6% – 100% Pooled result not conducted
Smith et al. (2015)	Systematic Review	25	14 (56%)	Reduction ranged between 10% and 62% across studies; 8 studies (32%) reported ranges exceeding 40% Pooled analysis not conducted	13	1%–31% AD
Current follow-up study	Systematic review with meta-analysis	33	9 (27%)	20.8% (95% CI 16–27) pooled decrease across studies, $p < 0.001$	19	0%–44% Asperger's 0%–50% PDD-NOS Pooled analysis not conducted
						Pooled results: 10.1% AD, $p < 0.001$, 23.3% Asperger's, $p = 0.001$, 46.1% PDD-NOS, $p = 0.52$

ASD autism spectrum disorder; CI confidence interval; AD autistic disorder; PDD-NOS pervasive developmental disorder-not otherwise specified

from ASD (Visser and Tops 2017). It is essential to view SCD as an independent diagnosis and recognize where it overlaps with ASD before its usefulness can be ascertained and tailored treatments can be developed. Future studies which measure SCD prevalence beyond applying the diagnosis to individuals who do not meet DSM-5 ASD criteria are warranted (Swineford et al. 2014). Further complicating the applicability of the diagnosis, five years after DSM-5 publication research is still being conducted to design standardized screening and/or diagnostic instruments for SCD (Baird and Norbury 2016; Norbury 2014; Visser and Tops 2017; Yuan and Dollaghan 2018). Overall, these issues add to the “ongoing debate regarding the validity of SCD as a diagnostic entity” (Visser and Tops 2017). Indeed, examination of SCD as a diagnosis, relative to other developmental communication disorders, is in its infancy, leaving its impact unknown. Exploring whether SCD is a legitimate diagnosis independent of ASD, as well as its potential to serve as a gateway for eligibility for treatment and services, are important areas for future research.

Limitations

The findings of this systematic literature review and meta-analysis must be interpreted with some caution. Overall, risk of bias of the included studies was moderate with potential bias stemming from lack of blinding of raters to results of the references standard, DSM-IV-TR diagnosis, and failure to assess interrater agreement in classification of DSM-5 diagnoses. While we took measures to conduct a rigorous systematic review, it has some limitations. Heterogeneity greater than expected by chance alone was present in each meta-analytic model. Six variables were identified that explained some of the heterogeneity; however, it is likely that additional unidentified factors also contributed to heterogeneity both within and between studies but were not explored. Finally, importance of the findings on SCD, which are the products of two separate but related systematic reviews, is limited by the small sample sizes across studies.

Conclusions

The diagnosis of ASD and the potential impact of SCD for those who do not meet criteria for an ASD diagnosis using DSM-5 criteria is evolving. Findings of this systematic review and meta-analysis provide further insight regarding how DSM-5 is being used both nationally and internationally since the release of the new diagnostic criteria and point to areas of future research, particularly for SCD.

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Author Contributions KMK conceived of the study, participated in the design and coordination of the study, and drafted and revised the manuscript. JG conducted the initial literature search and JU updated the literature search; both coordinated the study workflow in Covidence, drafted part of the methods section, and revised the manuscript. DR participated in the design and coordination of the study, drafted the introduction, and revised the manuscript. JMB provided clinical context in interpretation of study findings, drafted part of the results and discussion sections, and revised the manuscript. AMS conceived of the study, participated in the design and coordination of the study, performed analyses, and drafted and revised the manuscript. KMK, JG, JU, DR, and AMS participated in the literature screening, quality appraisal, and data extraction. All authors read and approved the final manuscript.

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Compliance with Ethical Standards

Conflict of interest All authors declare they have no conflicts of interest.

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