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A Systematic Review and Meta-analysis of Narrative Language Abilities in Children with Attention-Deficit/Hyperactivity Disorder

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Abstract

While children with ADHD are reported to have language problems, it is less clear if their ability to use language to tell a story (i.e., form a narrative) is impaired. Therefore, a systematic review and meta-analysis of studies examining the oral production of fictional stories in children with ADHD was conducted. Databases were systematically searched in January 2019 and December 2020 (follow-up). Studies comparing children (≤ 18 years) with ADHD to a control group of typically developing children were included. The meta-analysis adhered to PRISMA guidelines and was preregistered with PROSPERO [CRD42019122040]. Sixteen studies were retained. Results indicated that compared to typically developing children, children with ADHD produced less coherent narratives (Hedges' $g = 0.58$ $p < .001$), gave more ambiguous references (Hedges' $g = 0.52$, $p < .001$), made more disruptive errors (Hedges' $g = 0.41$, $p < .001$), and produced language that was less syntactically complex (Hedges' $g = 0.39$, $p < .05$). Children with ADHD also produced less language overall (Hedges' $g = 0.27$, $p < .05$), although this result appeared to be an artefact of publication bias. Two studies investigated internal state language and both found children with ADHD to produce narratives with less internal state language. Children with ADHD did not produce less fluent narratives (Hedges' $g = 0.23$, $p = .47$), although a scarcity of studies [K = 4] preclude firm conclusions. In conclusion, children with ADHD were impaired in several areas of oral narrative production and screening for narrative language problems should be considered when assessing language and communicative abilities in children with ADHD.

Keywords ADHD · Language · Narrative · Story-telling · Meta-analysis · Communication

Introduction

Attention-deficit/hyperactivity disorder (ADHD) is a common neurodevelopmental disorder characterized by a persistent pattern of inattention and/or hyperactivity/impulsivity that interferes with development and functioning across contexts (e.g. at home and in school; American Psychiatric Association; APA, 2013). From an early age, children with ADHD are reported to have problems with language (Tannock, 2018) and population-based studies suggest that the risk of having language problems is at least three times higher in children with ADHD compared to community controls (see e.g. Sciberras et al., 2014). As language problems often persist after controlling for language disorders (Oram et al., 1999), these problems appear to be associated with ADHD per se rather than co-occurring language disorders. In a recent meta-analysis Korrel et al. (2017) found children with ADHD to perform significantly below their peers on standardized language measures assessing overall, expressive, receptive, and pragmatic language with large effect sizes (ES; Hedges' $g = 0.97–1.23$). This suggests that ADHD is associated with problems across several aspects of language.

The meta-analysis by Korrel et al. (2017) was based on standardized or structured language tasks, and did not include complex language measures or analyses of discourse and conversation skills. This may, as noted by the authors, have led to an underestimation of language problems in children with ADHD. For example, studies have found prominent difficulties on narrative language tasks (i.e., storytelling) in individuals with brain damage (Liles et al., 1989) and neurodevelopmental disorders such as autism or pragmatic language impairment (Norbury & Bishop, 2003), in spite of few or no problems on standardized language tasks.

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From a linguistic point of view, a narrative is a genre of discourse – a form of social communication used to derive meaning from experiences and to construct a shared understanding of events (Losh & Gordon, 2014). In other words, it is the fundamental ability of orally producing a coherent story. The investigation of narrative abilities involves transcribing, coding, and analyzing longer pieces of discourse that can be elicited in various ways (e.g., prompting the story with a wordless picture book or retelling a story previously heard or seen). In language research, narratives typically refer to the narration of fictitious events (Loveland & Tunali, 1994). This is because personal stories tend to rely more heavily on autobiographical memory and story content than the ability to use language to construct a story (Orsolini, 1990). Narratives are considered to provide an ecologically valid measurement of the communicative competence of children generally (Boudreau, 2008) and children with ADHD specifically (Botting, 2002). As narrative ability is positively associated with academic and cognitive abilities (Boudreau, 2008; Curenton, 2011; Klecan-Aker & Swank, 1987) as well as social competency (Capps et al., 2000; Norbury & Bishop, 2003; Staikova et al., 2013), the investigation of narratives provide information about aspects of communication that is particularly relevant in the context of ADHD.

There are a series of challenges to the study of narrative ability in childhood ADHD. First, narrative abilities appear to vary depending on the administration procedure, including what kind of story is prompted and how (Boudreau, 2008). These varying procedures make it difficult to directly compare studies of narratives. Second, because there is no agreed upon approach to define and categorize narrative abilities in the context of ADHD, methodological diversity precludes meaningful conclusions about which specific categories of narrative ability are problematic in children with ADHD. To date, no systematic review about narratives in childhood ADHD has been conducted. The current meta-analysis hopes to fill this gap in the literature by providing an overview of narrative production abilities in children with ADHD as well as evidence concerning whether ADHD is associated with problems producing narratives.

The Current Study

The primary aim of this meta-analysis was to examine if children with ADHD differ from typically developing children in their narrative language ability. This was examined by statistically pooling evidence from previous studies investigating oral narrative production abilities in children with ADHD compared to typically developing children.

Because production of stories and story comprehension are considered very different abilities (Boudreau, 2008), the current study focuses on oral narrative production. Consequently, in the current study narrative abilities refer to the general production of narratives, not comprehension. Narratives are traditionally investigated with online tasks (i.e., participants are instructed to tell a story while looking at pictures or a wordless picture book) or recall tasks (i.e., participants are instructed to recall a story they have just heard, read, or watched). While online tasks are considered to resemble story construction abilities more than recall tasks (Boudreau, 2008), it has not (to our knowledge) been investigated whether narrative abilities differ significantly in online versus recall tasks. Consequently, and because a scarcity of studies was expected, an a priori decision was made to include recall tasks if they investigated narrative production abilities. If two different studies investigated the same sample, but recall tasks in one study and online tasks in another, then results from the online task were included in the meta-analysis (per theoretical definition this should provide a better measure of story construction/production ability; Boudreau, 2008). If enough studies are located, subgroup analyses will be conducted to compare performance in online versus recall tasks.

Method

Protocol and Registration

The study protocol was registered with PROSPERO [CRD42019122040] and the study conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Liberati et al., 2009) and Meta-Analysis of Observational Studies in Epidemiology (MOOSE; Stroup et al., 2000).

Eligibility Criteria

Due to scarcity of studies, broad inclusion criteria were applied. Studies were included if they had a minimum of 10 participants in total (i.e., ADHD plus controls) and compared aspects of oral narrative production in children ($\leq$ 18 years) with a formal ADHD (or equivalent) diagnosis or a score above a clinical cut-off on a validated ADHD rating scale, and no confirmed autism spectrum disorder (ASD) or language impairment diagnosis, to a control group of typically developing children. No constraints regarding IQ were imposed. If the study was part of an intervention study or additional experimental manipulation (e.g., telling a story on and off medicine) outcomes from baseline were included. As noted above, if two or more studies were conducted with the same sample, the study investigating online narrative production was included. If both studies used an online task, the study with the highest number of participants was included.

All English language studies with statistical information necessary for calculation of effect sizes were included. Theses or dissertations, regardless of publications status, were also included.

Search Strategy

The databases ERIC, PsycINFO, Education Database, Linguistics and language behavior abstracts, PsycArticles, PubMed and Embase were searched on the 17th and 18th of January 2019 with follow-up searches on the 18th of December 2020 using a predetermined strategy and search string: adhd or hkd or addh or hyperkine* or attention deficit* or hyperactiv* or hyperactiv* or overactive or inattentive or impulsiv* AND narrative OR story*. The search was not restricted by year. In PsycINFO, PsycArticles, and PubMed, "empirical studies" or "clinical trials" were included as search criteria. Abstracts, titles, and key words of eligible studies were screened by two independent reviewers (the first and the third author) and any disagreements resolved unanimously through discussion ($n = 51$). Full text screening was performed by two independent reviewers (the first and the third author) and disagreements were resolved though discussion ($n = 28$). Finally, the first authors of all the included studies were contacted and queried about unpublished or upcoming studies on the subject and reference lists of included studies were screened for additional studies.

Data Extraction and Categorization

Two independent reviewers (the first and the third author) independently extracted the following information about the characteristics of each study: ADHD assessment, IQ, number of participants (including gender and age distribution), matching and/or covariates in analysis, relevant inclusion/exclusion criteria (language impairment, ASD), medication status (including whether medication was discontinued prior to assessment), type of narrative task, as well as all relevant statistics. There were no disagreements among reviewers. See Table 1 for characteristics of included studies.

An a priori categorization scheme was developed based on the literature on narratives in children with ADHD, children with autism spectrum disorders, and typically developing children (see below). This resulted in seven categories, each measuring an aspect of the ability to orally produce a story. Two reviewers (the first and the third author) independently sorted the narrative outcomes from the eligible studies into the categories. Of the 122 outcomes extracted from the 16 studies, the two independent reviewers initially agreed on the coding of 51 outcomes, disagreed on 28, and were inconclusive about 43. Agreement was reached between the two reviewers (the first and third author) through discussion on the majority of outcomes ($n = 68$), while the remaining were resolved through discussion with a third reviewer (the fourth author, $n = 3$). The seven categories are described below (for ease of readability, the categories have been shortened and slightly reworded relative to the original protocol).

Oral Narrative Production Categories

Coherence: Coherence refers to the overarching structure of a story (Baixauli et al., 2016). It is the ability to create a coherent story structure that is easy to follow and has a logical order temporally and causally (Norbury & Bishop, 2003). Coherence is typically operationalized as inclusion of goals, attempts and outcomes, or establishment of a structure with a beginning, a middle, and an end (Norbury & Bishop, 2003).

Cohesion: Cohesion relates to linguistic structures that link sentences together (Norbury & Bishop, 2003) and has primarily been operationalized as ambiguous references (also referred to as within-clause errors). Adequate referencing of events and characters clarify to the listener how characters, events, and ideas in a story are related (Liles & Purcell, 1987; Wigglesworth, 1990). Though cohesion and coherence (see above) are related, the two are traditionally viewed as distinct categories (Karmiloff-Smith, 1985).

Disruptions: This category encompasses narrative errors that create disruptions in the storytelling. In studies of narrative abilities in children with ADHD, disruptions are frequently operationalized as sequence errors, misinterpretations, confabulations, and/or embellishments (Flory et al., 2006; Tannock et al., 1993).

Fluency: This category refers to errors in spoken narratives that make a story less fluent (Kuijper et al., 2017). Fluency of speech has been argued to reflect the cognitive processes underlying language production (Guo et al., 2008). Consequently, these outcomes measure how fluent the child's verbal production is. Examples of operationalizations of fluency includes for instance repeating sentences/words, abandoning sentences and false starts (Kuijper et al., 2017).

Production [category]: This category includes outcomes that pertain to how much language the child produces. Frequent operationalizations of production include the overall length of the story, number of words number of sentences etc. (Baixauli et al., 2016).

Syntactical complexity: All outcomes relating to the complexity of language will be extracted in a "syntactical complexity" category. Examples of operationalizations of the complexity of a child's language include lexical diversity (semantics), grammar and morpho-syntactic errors (Baixauli et al., 2016).

Internal state language: Internal state language (ISL) is operationalized as references to characters' internal states such as emotions or cognitive states, emotion verbs, or references to other mental states and activities (see e.g.

Table 1 Characteristics of included studies

	Study	N(ADHD/NC) %boys (ADHD/NC)	Age Range M age (ADHD/NC)	M IQ (ADHD/NC)	Diagnosis	Narrative task	Narrative category
1	Baixuali Fortea et al., 2018	35/37 91.4/62.1	7–11 9.1/8.5	99/101	DSM-5 ADHD diagnosis	Online (picture book)	Production Syntactic complexity Cohesion Disruptions Fluency
2	Derefinko et al., 2009	17/25 82.4/80	9–14 11.8/11.4	> 80	DSM-IV ADHD diagnosis	Online (picture book)	Production Coherence Cohesion
3	Flory et al., 2006	49/67 80/64	7–9 8.5/8.4	n/a	DSM-IV ADHD diagnosis	Online (picture book)	Production Coherence Cohesion Disruptions
4	Freer et al., 2011	89/99 78/58	5–11.5 9/8.7	n/a but "low IQ" excl. criterion	DSM-IV ADHD diagnosis	Online (one story with no pictures, one story with four pictures)	Coherence
5	Houghton et al., 2008	24/24 100/100	7–12 10.2/10.3	VIQ: 109,5/115,2 PIQ: 105,2/114,3	DSM-IV-TR ADHD diagnosis	Recall (Televised episode)	Coherence Production
6	Koltun, 2004	22/19 n/a	8–14 n/a	> 80	DSM-IV ADHD Diagnosis	Recall (audiotaped folk tales)	Production, Coherence Disruptions Cohesion
7	Kuijper et al., 2017	34/36 82/69	6–12 8.9/8.9	93.4/110	DSM-IV-TR ADHD diagnosis	Online (Picture book)	Production Syntactic complexity Fluency Internal state language Cohesion
8	Lee, 2017	15/15 73/60	6–9 8/7.9	> 85	DSM-IV ADHD diagnosis	Recall (stories read by participants)	Fluency
9	Lorch et al., 2010	57/98 77/61	4–9 7.2/7.2	> 80	DSM-IV ADHD diagnosis	Recall (televised episodes)	Coherence
10	Miniscalco et al., 2007	8/8 88/63	7–8 7.9/7.9	85–115	DSM-IV ADHD diagnosis	Recall (stories read aloud by experimenter)	Production Syntactic Complexity
11	Purvis & Tannock, 1997	17/17 100/100	7–11 8.7/9.4	> 80 105/110	DSM-III-R ADHD diagnosis	Recall (Audiotaped story)	Cohesion Disruptions
12	Renz et al., 2003	22/44 100/100	9–11 12/11.6	Reported to be within the normal range	DSM- IV ADHD diagnosis	Online (picture book)	Production Coherence Cohesion Disruptions
13	Rumpf et al., 2012	9/11 89/90	8–12 9.9/9.1	104/n/a for NC	ICD-10 ADHD diagnosis	Online (picture book)	Production Syntactic complexity Internal state language
14	Tannock et al., 1993	30/30 100/100	7–11 9.1/9.4	110/104	DSM-III-R ADHD diagnosis	Recall (audiotaped folk tales)	Production Disruptions Cohesion
15	Parigger, 2012	22/26 81/73	7–8 8.1/8.1	109/113	DSM-IV ADHD diagnosis	Online (picture book)	Production Syntactic complexity Fluency Coherence
16	Bergman & Hallin, 2021	15/31 46.7/32.3	11–16 13.7/13.7	Reported to be within the normal range	DSM-IV-TR ADHD diagnosis	Recall (with and without picture support)	Production Coherence Syntactic complexity

NC non-clinical, *M* mean, *n/a* not available, *DSM* Diagnostic and Statistical manual of Mental disorders, *ICD* International Statistical Classification of Diseases and Related Health Problems

Bamberg & Damrad-Frye, 1991; Capps et al., 2000). Consequently, this final category includes references to perceptions, thoughts, beliefs, and feelings (Rumpf et al., 2012).

Study Quality

Study quality and overall risk of bias was assessed by two independent reviewers (the first and the third author) using the Newcastle–Ottawa Scale for Assessing the Quality of Nonrandomized Studies in Meta-Analysis (NOS; Wells et al., 2000). Studies receive stars (*) based on whether they live up to quality criteria pertaining to selection, comparability, and ascertainment of exposure (Wells et al., 2000). In the selection and exposure categories, studies receive a star, if they are categorized as "a", which is the highest quality rating. Any rating below "a" (i.e., b or c) are not awarded stars. Under "Comparability" (see online resource 2 or Table 2 below), one star is awarded if the study controlled for age and two stars if the study controlled for other relevant factors (e.g., IQ) in addition to age. See Online Resource 2 for detailed description of the coding manual.

Statistical Strategy

The extracted data were statistically pooled in the program Comprehensive Meta-Analysis, version 3 (CMA; Borenstein et al., 2013). Random meta-analysis was used as if sampling was from a universe of possible studies as opposed to fixed meta-analysis, where one single effect size is assumed to underlie all studies (Borenstein et al., 2010).

Effect sizes (ESs) were calculated as Hedges' *g*, a measure of the standardized mean difference (Borenstein et al., 2009). It is interpreted similarly to Cohen's *d* with ESs of 0.2, 0.5 and 0.8 representing small, medium and large ESs, respectively (Hedges & Olkin, 1985). The calculation of *g*-values was performed in CMA. A summary effect was calculated for each outcome category, that is, whenever a study contributed with more than one outcome to a category, a standardized average was calculated using Cohen's *d* and the pooled variance (as is recommended in Borenstein et al., 2009). Thus, each study only contributed with one effect size in each narrative category.

Heterogeneity was explored using Q, I^2, T, T^2, and prediction intervals. The Q statistic is the weighted sum of squared deviations of all effect sizes from the mean effect size and a significant Q-test indicates that the variance in effect size is due to true variance (Borenstein et al., 2009). The recommended alpha level for Q-test is 0.1 (Borenstein et al., 2009). I^2 is a measure of the proportion of true variation between studies compared to variation due to sampling error, *not* the total amount of heterogeneity, as I^2 is often interpreted as (Borenstein et al., 2017). A smaller I^2 indicates that a larger percentage of the variation in the forest plot is due to sampling error, not true variation in the synthesized effect sizes (Borenstein et al., 2017). T is an estimate of the standard deviation of true effect sizes, while T^2 is an estimate of the variance of true effect sizes (Borenstein et al., 2017). T^2 in log units and the mean effect was used to calculate prediction intervals (i.e., estimates of the range of true effects; Borenstein et al., 2017). Prediction intervals provide a range within which the ES of 95% of all possible studies would fall (not to be confused with the CI that is a measure of the precision of the weighted mean ES; Borenstein et al., 2017). Prediction intervals were only calculated with 8 or more studies and only if T^2 was above 0 (Borenstein et al., 2009).

Not enough studies were identified in the literature search to perform sub-group analysis and compare recall and online tasks, and thus this aspect of the study protocol had to be abandoned.

Publication bias was investigated with Eggers test for small study effect and Duvall and Tweedie's trim and fill method when 10 or more studies were included in the meta-analysis. As the investigation of publication bias with statistical tests is not recommended with fewer than 10 studies (Borenstein et al., 2009) funnel plots were investigated visually to assess publication bias in categories with fewer than 10 studies.

Results

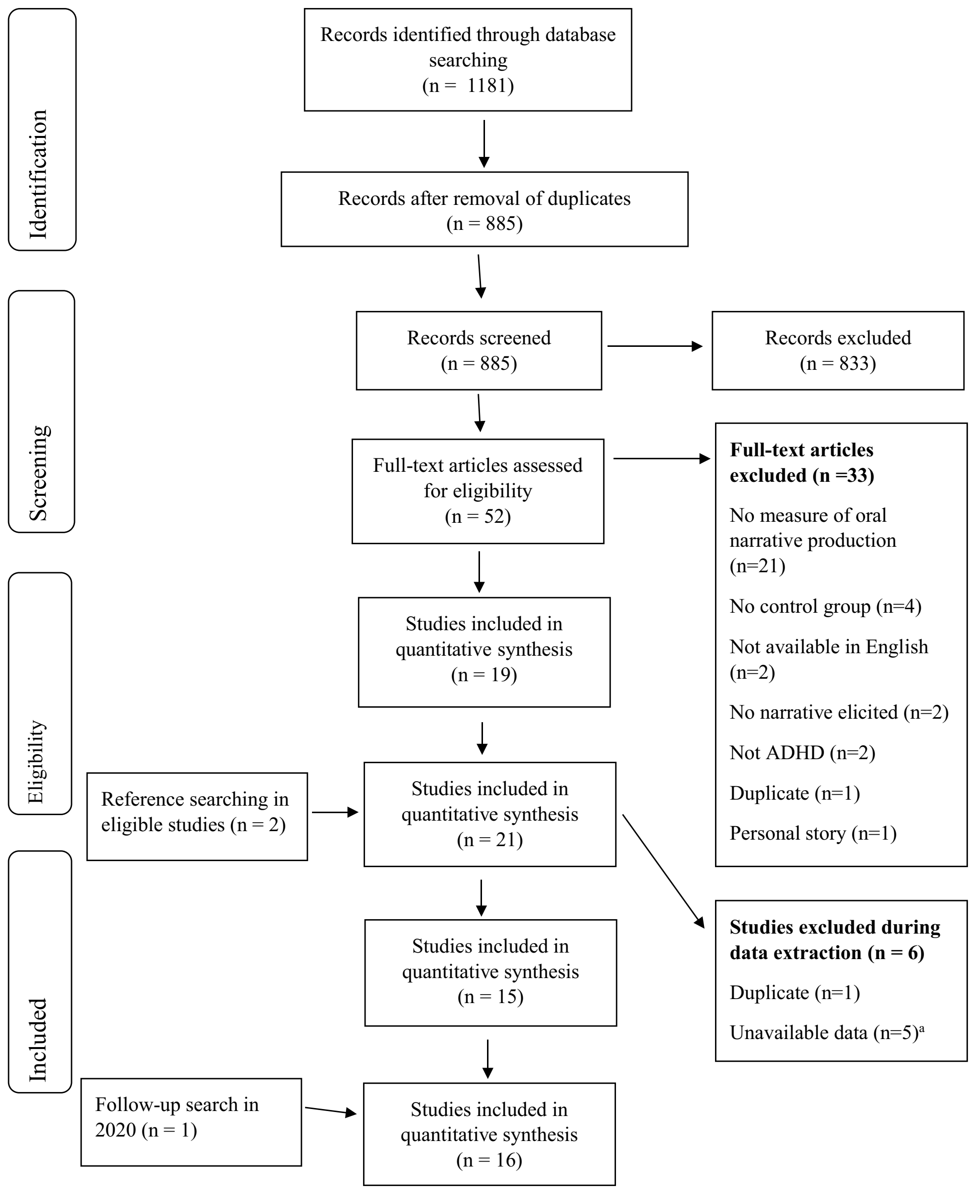
Inclusion of Studies

The search process is illustrated in the flow diagram in Fig. 1. After removal of duplicates, 885 studies were screened based on abstract, title, and keywords, and 52 studies went on to full-text screening. Of these, 33 were excluded; the majority due to outcomes being unrelated to narrative production (21 of 33 studies, see flow diagram below). Reasons for exclusion are detailed in Fig. 1. Reference searching led to the identification of two additional studies, leaving 21 studies to be included in the final review and meta-analysis. However, during data extraction six studies had to be excluded (see reasons in flow diagram below). Of the initial 1181 studies, 15 studies were included in the meta-analysis. Follow-up searches in December 2020 resulted in one additional study, resulting in 16 studies being included in the final meta-analysis.

Study Characteristics

A total of 16 studies were included in the systematic review, two of which were theses (6, 15). Study characteristics are detailed in Table 1. The studies included 1015 participants, 429 children with ADHD and 586 control children (range of sample size in ADHD group 8–99), aged 4–16 years. Based on the fifteen studies that provided information about gender distribution, 84.6% of the ADHD participants were male (control group 74.2%). Information about general cognitive ability was provided in 15 of 16 studies. Seven studies

Fig. 1 PRISMA flow diagram of study identification and selection



[a]Authors were contacted for unpublished data but no additional data was provided. Flow diagram from Moher et al., 2009

provided mean IQs (M ADHD groups = 104.3, M control groups = 110.6[1]; 1, 5, 7, 11, 13, 14, 15), two reported that all children had an IQ above 85 (8, 10), and three that IQ was above 80 (2, 6, 9). The three remaining studies reported that all children in the sample had an IQ "within the normal range" (4, 12, 16). Ten studies reported that children receiving medication (16.6–100%) were medication-free during assessment (2, 3, 4, 5, 6, 9, 11, 12, 14, 15). One study reported that 13.3% of the ADHD sample received medication which was not discontinued during assessment (16). One study reported that 71.4% received medication, but did not report whether it was discontinued during assessment (1). Four studies did not report the medication status of the children (7, 8, 10, 13).

Comparing aspects of narrative ability in children with ADHD to typically developing children was the primary aim in 15 of 16 (93.8%) studies. The tasks applied to measure narratives varied extensively. Eight studies included an online task (1, 2, 3, 4, 7, 12, 13, 15), and eight a recall task (5, 6, 8, 9, 10, 11, 14, 16). There were different types of online tasks; in seven studies children told stories based on a wordless picture book (1, 2, 3, 7, 12, 13, 15) and in one study children told two stories, one without cues and one prompted by four pictures (4). Of the recall tasks, four studies used recall of audiotaped stories (6, 11, 14, 16), two used retellings of televised stories (5, 9), one used recall of a story

[1] VIQ from Houghton et al. (2008) used in calculation of mean. The mean for the NC group was based on six studies, as the seventh study (13) only provided a mean IQ for the ADHD group.

read aloud by an experimenter (10), and one used recall of stories read by the participants (8).

A total 122 narrative production outcomes were extracted and sorted into the categorization system (details on the coding of outcomes are included in Online Resource 1, Supplemental Table 1).

Coherence

Nine studies (2, 3, 4, 5, 6, 9, 12, 15, 16) contributed with outcomes to this category. The weighted mean ES was moderate, Hedges' $g = 0.58$ $p < 0.001$, with a SE = 0.08 and CI [0.43; 0.74] indicating children with ADHD produce less coherent narratives than typically developing children (see Fig. 2A). The Q statistic was not significant at $Q(8) = 5.99$ $p = 0.65$, $I^2 = 0$, $T = 0$, $T^2 = 0$, indicating that all of the variance between studies in the size of effect was due to sampling error, not variance in true effects.

A visual inspection of the funnel plot (see Fig. 3A) indicated a rather symmetric distribution of the effect sizes of the studies around the mean.

Cohesion

Eight studies contributed with outcomes (1, 2, 3, 6, 7, 11, 12, 14). The weighted mean ES was moderate Hedges' $g = 0.52$, $p < 0.001$, SE = 0.10, CI [0.34; 0.70], indicating that children with ADHD produce more ambiguous references in their narratives than typically developing children (see Fig. 2B). The Q statistic was not significant $Q(7) = 7.11$, $p = 0.42$, $I^2 = 1.50$, T of 0.03 and $T^2 = 0.001$, suggesting that no significant proportion of the variance was due to variance in true effects. The 95% prediction interval was [0.28; 0.76], indicating that the true effect size in 95% of cases will fall somewhere in this range of a small to a large effect.

A visual inspection of the funnel plot (see Fig. 3B) suggested that the studies were relatively symmetrically distributed around the mean.

Disruptions

Six studies contributed to this category (1, 3, 6, 11, 12, 14). The weighted mean ES was small to moderate Hedges' $g = 0.41$, $p < 0.001$, SE = 0.10, CI [0.24; 0.58] (see Fig. 2C). This suggests that children with ADHD produce more disruptions in their narratives than typically developing children. The Q-statistic was not significant $Q(5) = 1.20$, $p = 0.95$, $I^2 = 0$, $T = 0$, $T^2 = 0$, indicating that all of the variance between studies in the size of effect was due to sampling error, not variance in true effects.

In the visual inspection of the funnel plot (Fig. 3C), 5 of the 6 studies were found to cluster quite near to the mean.

Production [Category]

Twelve studies contributed with outcomes (1, 2, 3, 5, 6, 7, 10, 12, 13, 14, 15, 16). The weighted mean ES was small but significant, Hedges' $g = 0.27$, $p < 0.05$, SE = 0.11, CI [0.05; 0.48], indicating that overall children with ADHD produce less narrative language than typically developing children (see Fig. 2D). The Q statistic was significant at alpha level 0.1 $Q(11) = 19.61$, $p < 0.05$, $I^2 = 43.91$, $T = 0.25$, $T^2 = 0.06$, indicating that a significant proportion of the variance between studies was due to variance in true effects. The 95% prediction interval was [-0.33; 0.87], indicating that in the universe of populations represented by these studies, the true effect size will in 95% of cases fall somewhere in this range.

Egger's test indicated a small study effect ($p < 0.05$). A visual inspection of the funnel plot (Fig. 3 D) revealed more studies to the right of the mean. The Duval and Tweedie's trim and fill method trimmed five studies and estimated that in the absence of publication bias there would be no effect; Hedges' $g = 0.03$, [-0.22; 0.28], $Q(6) = 48.71$.

Fluency

Four studies contributed to this category (1, 7, 8, 15). The weighted mean ES was small and not significant, Hedges' $g = 0.23$, $p = 0.47$, SE = 0.31, CI [-0.38; 0.83], suggesting there is no significant difference in the fluency of the narratives of children with ADHD compared to typically developing children (see Fig. 2E). The Q statistic was significant $Q(3) = 14.60$, $p < 0.01$, $I^2 = 79.45$, $T = 0.55$, $T^2 = 0.30$, indicating that a significant proportion of the variance between studies in the size of effect was due to variance in true effects.

Visual inspection of the funnel plot (Fig. 3E) suggested that the four studies were relatively far away from the mean, with two studies located below the mean and two above.

Syntactical Complexity

Six studies contributed with outcomes to this category (1, 7, 10, 13, 15, 16). The weighted mean ES was small to moderate and significant, Hedges' $g = 0.39$, CI [0.15; 0.63], $p < 0.05$, SE = 0.12, indicating children with ADHD have less syntactically complex narratives than typically developing children (see Fig. 2E). The Q statistic was not significant $Q(5) = 3.752$, $p = 0.59$, $I^2 = 0$, $T^2 = 0$, indicating no significant proportion of the variance was due to variance in true effects.

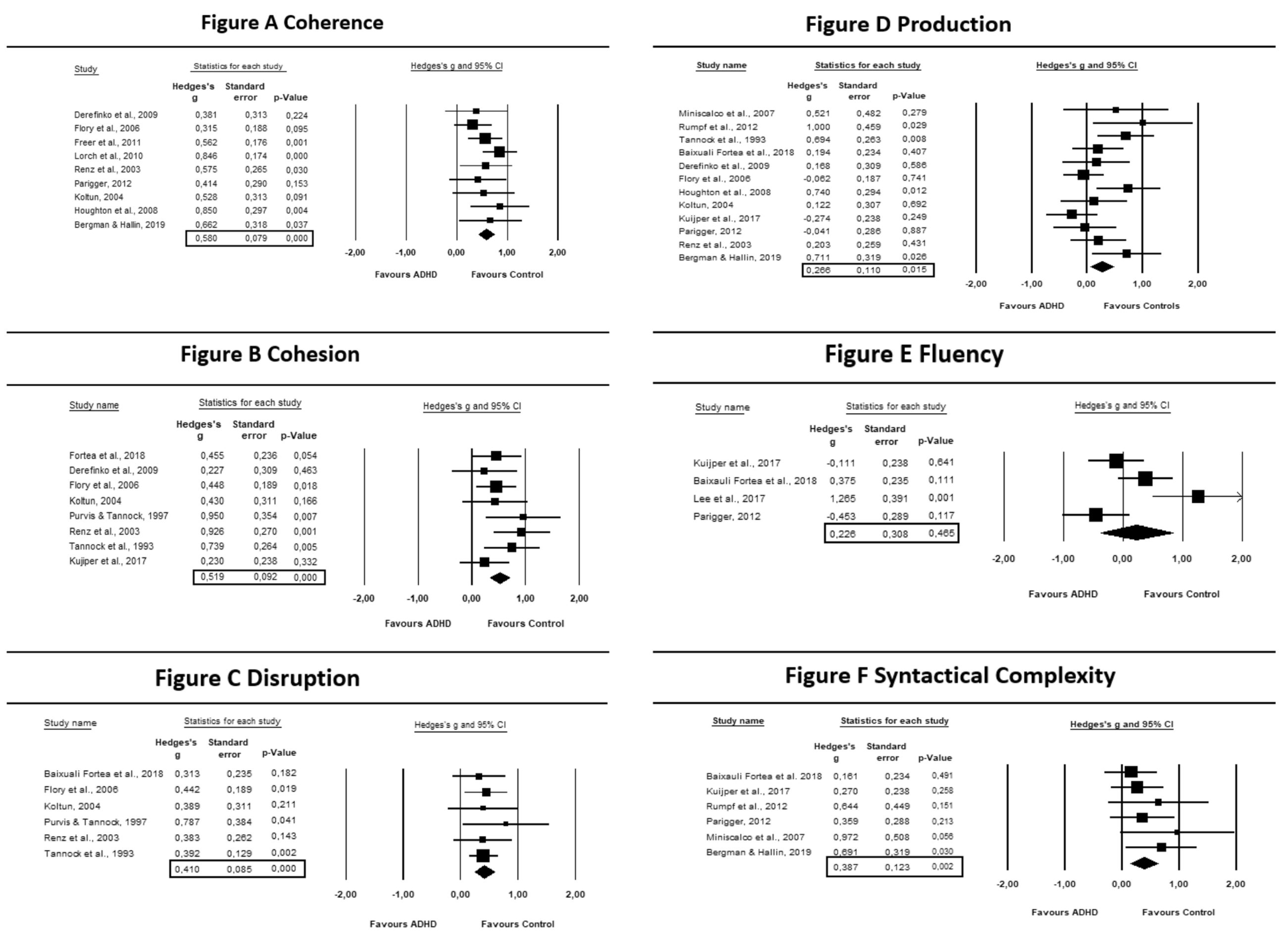


Fig. 2 **A-F** Forest plots by narrative category, Hedges' g, and 95% confidence intervals. The black squares refer to individual studies and the size of the squares represent their relative weights assigned in the random effects model. The black lines refer to each study's 95% CI. The black diamond refers to the overall mean ES for each narrative category

Visual inspection of the data (Fig. 3F) found three studies to the right as well as three studies to the left of the mean.

Internal State Language

Two studies contributed with outcomes to the internal state language category (7, 14). Meta-analysis was not considered feasible with only two studies, however, both studies found children with ADHD to use less internal state language in their narratives than typically developing controls. One study found a significant difference with a small effect size d = 0.25 (7) and the other with a large effect size $d = 1.10$ (14). Details can be found in Online Resource 1, Supplemental Table 2.

Risk of Bias Assessment

Ratings on the NOS for each study are detailed in Online resource 2. Regarding selection, none of the ADHD samples were randomly sampled. Of the 15 studies, 13 recruited community controls, while the remaining three studies recruited children through the same hospital as the ADHD group (11, 14) or through outpatient clinics (7). Regarding comparability, 10/16 studies controlled for age, while only 5/16 controlled for additional variables (see Table 2 below). Only two studies controlled for general language ability (vocabulary and expressive language). Regarding exposure, the coder was blind to diagnostic status in 9/16 studies. A majority of studies (14/16) used the same procedure in the ADHD and the control group, and the majority of

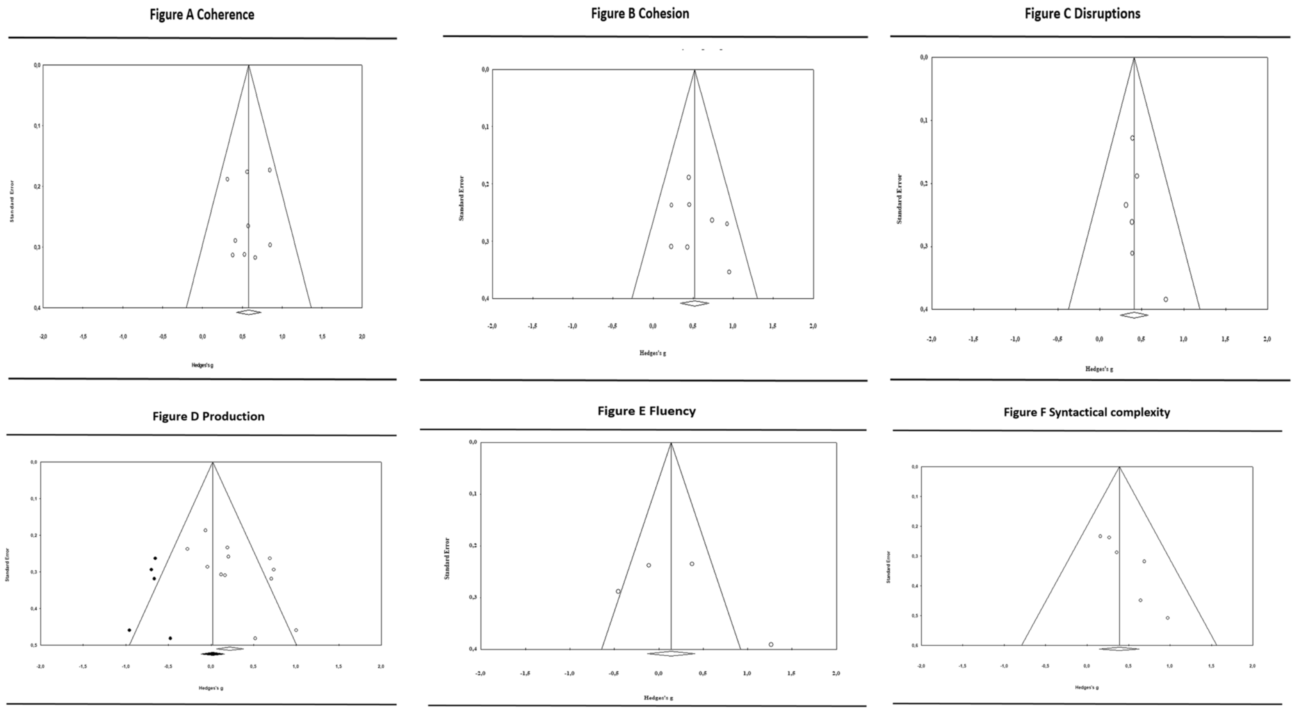


Fig. 3 **A-F** Funnel plot by narrative category of standard error by Hedges' g effect size. The white circles refer to the studies. In Fig. 3**D** the black studies refer to studies added by Duval and Tweedie's Trim and Fill approach to adjust for publication bias. In Fig **A-F**, the white diamonds refer to adjusted mean ES values. The black diamond in Fig. 3**D** refers to adjusted mean ES values corrected for publication bias

Table 2 Overview of the ratings on the Newcastle–Ottawa Scale for the included studies

	Study	Selection (max 4 stars)				Comparability (max 2 stars)	Exposure (max 3 stars)		
		1	2	3	4		1	2	3
1	Baixuali Fortea et al., 2018	*	b	*	*	**	*	*	*
2	Derefinko et al., 2009	*	b	*	*		*	*	*
3	Flory et al., 2006	*	b	*	*	*	*	*	*
4	Freer et al., 2011	*	b	*	*	**	*	*	c
5	Houghton et al., 2008	*	b	*	*	*	c	b	c
6	Koltun, 2004	*	b	*	*	**	b	b	*
7	Kuijper et al., 2017	*	b	b	*		*	*	*
8	Lee et al., 2017	*	b	*	*	*	c	*	*
9	Lorch et al., 2010	*	b	*	*	*	*	*	*
10	Miniscalco et al., 2007	*	b	*	*	*	*	*	*
11	Purvis & Tannock, 1997	*	b	b	*		c	*	*
12	Renz et al., 2003	*	b	*	*		c	*	*
13	Rumpf et al., 2012	*	b	*	*	**	c	*	c
14	Tannock et al., 1993	*	b	b	*	**	*	*	*
15	Parigger, 2012	*	b	*	*		c	*	*
16	Bergman & Hallin, 2021	*	b	*	*		c	*	*

Max maximum

Selection 1: * children with ADHD had clinical hospital diagnosis or clinical diagnostic interview performed to affirm ADHD diagnosis. Selection 2: b sampling of control group not random or not described. Selection 3: * control group consisted of community controls; b hospitalized controls. Selection 4: * no one in control group had previous or current ADHD diagnosis

Comparability: * matching/controlled for age in analysis, ** matching/controlled for age + other variables in analysis

Exposure 1: * coder of narrative was blind to diagnostic status of the children; b coders were not blinded; c = no description. Exposure 2: * administration of narrative task was the same for both groups (ADHD and controls), b different procedures for ADHD and control group. Exposure 3: * rates of completion of narrative task was the same for both groups (ADHD and controls), c rate different or no description studies had comparable response rates (i.e., percentage of children who completed the narrative task) for the two groups (13/16).

Discussion

The primary aim of this systematic review and meta-analysis was to determine whether children with ADHD differ from typically developing children in their narrative language ability. To that end, six categories of narrative production labeled coherence, cohesion, disruptions, production, fluency, and syntactical complexity were examined through meta-analysis. Overall, the results suggested that children with ADHD have deficits in several areas of narrative production compared to typically developing children. These areas may especially relate to the coherence of speech. This is, to the best of our knowledge, the first systematic review and meta-analysis of its kind within the field of ADHD, and the results add to the growing evidence for language problems in ADHD.

Children with ADHD were found to be impaired (relative to typically developing children) in the categories of narrative production labelled coherence, cohesion, and disruptions. Coherence is the ability to tell a story where the purpose, the major events, and the outcome are all conveyed in a temporally meaningful order, making the story easy to comprehend, whereas cohesion is the ability to correctly and unambiguously reference characters throughout a story, so the listener is able to follow the storyline. Coherence and cohesion both obtained medium mean ESs (0.58 and 0.52 respectfully), although the prediction interval for cohesion indicated a range of effects in the population from a small to a large effect (i.e., the effect is large in some populations and small in others). Disruptions (i.e., narrative errors that disrupt the story such as telling events out of order, confabulations, and misinterpretations) obtained a small to medium mean ES (0.41). Heterogeneity analyses indicated only a trivial amount of variation in true effects for these categories, suggesting that regardless of measures applied or populations investigated, children with ADHD have more problems in these areas of narrative production than typically developing children. Coherence, cohesion, and disruptions all pertain to coherency of speech, and problems in these categories may make children with ADHD come across as more linguistically incoherent, possibly affecting their social communication. Theoretically, narrative coherence, cohesion, and disruption are often argued to be associated with executive control (see e.g., Tannock & Schachar, 1996). Specifically, it has been argued that the ability to organize, plan, as well as hold multiple events and characters in working memory is necessary in order to produce coherent narrative language (Tannock & Schachar, 1996). An association between language and executive functions is also proposed in Barkley's neuropsychological model of ADHD (Barkley, 1997, 2015) where impairments in inhibition and working memory are proposed to affect internalization of speech and verbal working memory. Executive function deficits have consistently been associated with ADHD (Pievsky & McGrath, 2017), but studies also suggest a high degree of heterogeneity, with some but not all children with ADHD displaying these deficits (Karalunas & Nigg, 2020; Nigg et al., 2005). Possibly, children with ADHD and executive dysfunction have more problems with coherence, cohesion, and disruptions than children with ADHD and little or moderate executive function deficits. This could also help explain why the prediction interval in the cohesion category suggests that some ADHD populations have more difficulty with cohesion than others do. The specific association between narrative production and executive function has, however, rarely been examined in children with ADHD (Kuijper et al., 2017). Furthermore, ADHD has been associated with deficits in a neuropsychological domains other than executive function, including delay aversion (Marx et al., 2021; Sonuga-Barke et al., 1992) or temporal processing (Coghill et al., 2018). While speculative due to the lack of research, it is possible that these deficits could influence the production of language. For instance, a delay averse child might impulsively tell the different aspects of a story in a random order thus creating a less coherent story or a child with ADHD and problems in temporal processing might produce a less coherent story with more disruptions (e.g., telling events out of sequence) due to problems perceiving or representing time (Coghill et al., 2018). Moving forward, studies should examine whether and how neuropsychological deficits contribute to language problems in children with ADHD and widen the field to include other domains than executive function.

Syntactical complexity had a significant (albeit modest) mean ES (Hedges $g = 0.39$). In the syntactical complexity category, the traditional heterogeneity measures indicated that no amount of heterogeneity was due to true variation in effects. These findings suggest that children with ADHD have less complex language and make more grammatical and morpho-syntactical errors than their peers. Previous studies using standardized language tasks have also found that children with ADHD have problems with structural aspects of language (Korrel et al., 2017), and taken together, this suggests that children with ADHD have tangible problems with not only coherence of speech, but also the very basic domains of structural language such as vocabulary, morphology and syntax. Our ability to communicate and produce coherent language is dependent upon these basic language skills (Baker et al., 2012), and the current results therefore suggest that children with ADHD have problems with language that go beyond being able to concentrate and plan speech in the moment. Though requiring more research, this could mean that clinicians might be able to improve the social communicative skills of children with ADHD by providing language intervention targeted at structural language.

Production [category] refers to how much language is produced (while the remaining categories encompass the quality of the produced language). There was a small significant mean effect in the production category (Hedges' $g = 0.27$), indicating that overall, children with ADHD produce less narrative language than typically developing children. The Duvall and Tweedie's trim and fill method indicated that the mean effect was an artefact of publication bias, suggesting children with ADHD do not produce less language than typically developing children. The large amount of heterogeneity should, however, be taken into account when interpreting results. In addition to a significant Q-statistic indicating significant variance in true effects, (i.e. one or more unknown variables likely affect how much language children with ADHD produce), the prediction interval suggested that the effect sizes varied substantially across studies. In other words, the effect is positive (and large) in some populations (meaning that children with ADHD produce significantly less narrative language than controls); whereas the effect is negative in other populations (meaning children with ADHD produce more language than typically developing children). This heterogeneity might also have affected the precision of the publication bias analysis. Given that "excessive talking" is a core symptom of ADHD in DSM-5 (APA, 2013) and ICD-11 (WHO, 2018), one might expect children with ADHD to produce *more* language than controls. However, while children with ADHD may talk excessively, hog conversations etc. in everyday interactions, they may have difficulty producing language when specifically asked to do so (e.g., prompted to describe a photo or create a sentence using a specific word; Green et al., 2014) or even asked by parents how their day went. Therefore, though the current meta-analysis indicates that children with ADHD do not have problems in this area of narrative production, moving forward it is important to investigate what affects the amount of language children with ADHD do produce (e.g., characteristics of children with ADHD or the narrative tasks applied to measure production). Understanding what affects the productive speech of children with ADHD will hopefully improve our understanding of the language difficulties in children with ADHD – and, by extension, how to assess and treat them.

In the fluency category there was no significant effect (Hedges' $g = 0.27$), meaning that there is nothing to suggest that children with ADHD have less fluent narratives than typically developing children (although this is likely affected by a scarcity of studies [$K = 4$]). There are however different definitions of the concept "fluency". In the present study, fluency was defined as false starts, repeating words etc., similar to symptoms of Childhood Onset Fluency Disorder (i.e. stuttering; APA, 2013). However, in other studies fluency has been applied somewhat synonymous with coherence of speech, that is, fluency is also often defined as language that is easy to follow, with unambiguous references and without disruptive errors (Guo et al., 2008). Though the type of fluency defined in the current study might not be impaired in children with ADHD, there is a lack of studies that address fluency (Tannock, 2018) and this lack of studies makes the association between these language disturbances and ADHD in need of further study.

Only two studies investigated internal state language, and were therefore not feasible to combine in a meta-analysis. Both studies found children with ADHD to include fewer references to internal states than typically developing children (Hedges' $g = 1.01$ and 0.25 respectively). This area of narrative production is considered a measure of social and emotional understanding (Siller et al., 2014). As ADHD is associated with emotional and social difficulties (Graziano & Garcia, 2016; Ros & Graziano, 2018), it appears relevant to investigate this area of narrative production in future studies.

The categorization scheme included in the present study made it possible to synthesize results from different and methodologically diverse studies. The traditional heterogeneity analyses supported the use of this scheme, as the true variation was close to zero or small in all categories but fluency and production [category]. Consequently, the outcomes combined in each category appear to all measure the same aspect of narrative ability, providing preliminary evidence of the validity of the categorization scheme. The scheme could be of use in future studies to further the investigation of narrative language in children with ADHD, perhaps providing a collective starting point for a more coherent research field.

This study had several strengths, including being the first systematic review and meta-analysis of narrative abilities in children with ADHD, organizing narrative abilities into categories, and including relevant heterogeneity analyses (e.g., prediction intervals). There are also some limitations mainly pertaining to the studies included in this review. First, there was an overall scarcity of studies addressing narrative abilities in ADHD. Small samples impeded the statistical examination of publication bias in the majority of analyses and may have affected the precision of the meta-analysis as well as the heterogeneity analysis. Second, the NOS quality ratings revealed a lack of control for relevant aspects (e.g., age and socio-economic status) in the majority of studies, which makes it uncertain if the differences found between children with and without ADHD were in fact due to ADHD or some other variable. Future studies would benefit from investigating the predictive effect of variables that previous studies have found to be associated with language, such as age, IQ (Rohrer-Baumgartner & Zeiner, 2014), executive function (Martinussen & Tannock, 2006; Sjöwall & Thorell, 2014), and social economic status (Hart

& Risley, 1995). Additionally, because a majority of studies did not include general language ability in the matching process or as a covariate in the analyses, we cannot determine whether it is the child's language abilities in general rather than their ADHD that determines their ability to produce a coherent narrative. Moving forward, the investigation of the circumstances during which it is difficult (or easy) for children with ADHD to produce coherent language will likely provide relevant knowledge for the assessment and treatment of language difficulties in children with ADHD. Third, the general lack of studies meant it was not feasible (or possible) to conduct subgroup analyses or moderation analyses to investigate possible contributors to heterogeneity. This includes investigating the effect of medication status or the use of different DSM versions to diagnose ADHD. The lack of studies also meant there were too few studies to perform feasible subgroup analysis of recall vs online tasks – but as the variance (the Q, T, and I^2 statistics) in all categories but fluency and production [category] was negligible, it suggests that there was no variation to explore. Future studies should investigate whether narrative abilities of children with ADHD vary according to type of narrative task or medication status as it is still unclear whether these variables affect how much or how coherent a story children with ADHD produce (Boudreau, 2008; Francis et al., 2001).

The results from the present meta-analysis suggest that it is important to include language assessment in the standard clinical assessment of ADHD. Often, language issues in children with ADHD go unnoticed and even when language problems are identified children rarely receive targeted treatment (Sciberras et al., 2014; Tannock, 2018). As language and communication are important to social cognition (Astington & Baird, 2015), social abilities (Green et al., 2014; Staikova et al., 2013), and academic abilities (Curenton, 2011; Klecan-Aker & Swank, 1987), children with ADHD and language impairment may also be more functionally impaired in other aspects of life than children with ADHD without such impairment. Additionally, language impairments may potentially compromise the effectiveness of psychosocial treatments for ADHD (e.g., cognitive behavioral therapy or social skills training), as these often include verbal components. Consequently, including a thorough assessment of language abilities in children with ADHD would enable not only targeted treatment towards possible language impairment, but addressing these impairments in interventions directed towards ADHD.

In conclusion, the results from the current meta-analysis suggests that children with ADHD have impairments in their narrative language. In particular, children with ADHD produce narratives that are less coherent, less cohesive, less syntactically complex, and include more disruptive errors than typically developing children do. The results add to the growing evidence for an association between ADHD and language difficulties. Additionally, difficulties pertaining to coherent language could support an association between ADHD and social pragmatic communication disorder (Green et al., 2014; Tannock, 2018). Further research is needed to establish how robust the current findings are as well as what variables affect narrative production, such as executive functions.

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