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Written expression in boys with ADHD: The mediating roles of working memory and oral expression

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ABSTRACT

The written expression difficulties experienced by children with ADHD are widely recognized; however, scant empirical evidence exists concerning the cognitive mechanisms and processes underlying these deficiencies. The current study investigated the independent and potentially interactive contributions of two developmentally antecedent cognitive processes – viz., working memory (WM) and oral expression – hypothesized to influence written expression ability in boys. Thirty-three boys with ADHD-Combined Presentation and 27 neurotypical (NT) boys 8–12 years of age were administered standardized measures of oral and written expression, and multiple counterbalanced tasks to assess WM central executive (CE) processes, WM phonological short-term memory (PH STM), and WM visuospatial short-term memory (VS STM). Bias-corrected bootstrapped mediation analyses revealed a significant mediation effect, wherein the independent and interactive effects of PH STM and oral expression collectively explained 76% of the diagnostic status to written expression relation. The implications of the obtained results for clinical practice suggest that children with ADHD may benefit by incorporating a blended approach that simultaneously strengthens PH STM capacity and oral expression abilities as antecedents to engaging in writing-related activities.

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Attention-deficit/hyperactivity disorder (ADHD); working memory; written expression; oral expression; executive functions

The classification of attention-deficit/hyperactivity disorder (ADHD) as a neurodevelopmental disorder reflects its early onset and neurologically based cognitive deficits (American Psychiatric Association, 2013). The disorder affects an estimated 5%-7% of children worldwide (Polanczyk, Willcutt, Salum, Kieling, & Rohde, 2014) and is characterized by clinically impairing levels of inattentiveness, excessive gross motor activity, and impulsivity. A broad range of secondary features are evident in children with ADHD including difficulties with parent/peer relationships (Lifford, Harold, & Thapar, 2008, Normand et al., 2013), deficient organizational abilities (Kofler et al., 2018), and learning difficulties (DuPaul, Morgan, Farkas, Hillemeier, & Maczuga, 2016, Frazier, Youngstrom, Glutting, & Watkins, 2007), the latter of which contributes disproportionately to the overall cost of illness (Doshi et al., 2012, Pelham, Foster, & Robb, 2007).

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It is well established that children with ADHD experience considerable deficits in reading, math, and language-related problems (DuPaul, Gormley, & Laracy, 2013, Loe and Feldman, 2007, Martinussen, Hayden, Hogg-Johnson, & Tannock, 2005); however, their struggles with written expression are less well studied. This situation is particularly concerning given that a substantial proportion of children with ADHD (35–80%) experience writing difficulties ($ES = 0.67\text{--}0.80$), with reported prevalence rates comparable to or larger than that of children with ADHD and co-occurring reading or math difficulties (Frazier et al., 2007, Mayes and Calhoun, 2006, Mayes, Calhoun, & Crowell, 2000, Reid, 2012).

The ubiquity of written expression within modern society underscores its importance. Children are increasingly engaged in composing emails, texting, posting messages on social media, and interacting with friends and family. In school, writing competency is interwoven across curricula and underlies students' ability to communicate thoughts and knowledge during daily academic activities, take notes during academic instruction, and respond to written assignments. Writing is a particularly challenging skill to acquire and master because it requires the individual to recall and use proper lexical rules while simultaneously generating, maintaining, and organizing relevant ideas into a logical narrative (Kellogg, 2001, Vanderberg and Swanson, 2007).

Deficiencies in writing abilities are associated with near- and far-term negative outcomes. For example, poor writing abilities predict higher rates of grade retention, lower grade point averages, and lower scores on standardized academic achievement tests (Abbott, Berninger, & Fayol, 2010, Molitor, Langberg, Bouchtein, et al., 2016). Long-term consequences of underdeveloped writing abilities are also well documented and weigh heavily in many hiring/promotion decisions in the US, wherein businesses spend over \$3.1 billion annually on writing remediation training (National Commission on Writing, 2004).

Extant theoretical accounts of children's written expression abilities implicate two primary cognitive contributors – viz., working memory (WM) and oral expression abilities (Berninger, 1999, DeBono et al., 2012, Graham, Collins, & Rigby-Wills, 2017, Hayes, 1996, Kellogg, 2001). Although many contemporary models of WM exist (e.g., Baddeley, 2007, Engle, Tuholski, Laughlin, & Conway, 1999, Gray et al., 2017, Miyake et al., 2000), Baddeley's (2012) model is one of the most widely used due to its usefulness in exploring a wide range of WM-related mechanisms and processes and widespread adoption in child psychopathology research (Alderson, Rapport, Hudec, Sarver, & Kofler, 2010, Rapport et al., 2008). Baddeley views WM as a multicomponent system responsible for the temporary storage, rehearsal, maintenance, processing, updating, and manipulation of internally held information (Baddeley, 2007). The domain-general working component consists of a central executive (CE) supervisory system that controls attentional focus, reacts to multi-task demands, and oversees/coordinates two modality specific, anatomically distinct memory subsystems that upload information from long-term memory (Baddeley, 2012). These include phonological short-term memory [PH STM], localized in the left temporoparietal region and Broca's area, and visuospatial short-term memory [VS STM], localized in the posterior parietal and superior occipital cortices (Baddeley, 2003, Todd and Marois, 2005).

WM's involvement in children's written expression abilities is postulated to represent a balance between upper-level CE abilities and subsidiary PH STM capacity

(McCutchen, 1996, McCutchen, Covill, Hoyne, & Mildes, 1994). This hypothesized interplay reflects the expectation that as PH STM becomes increasingly automatized in its ability to upload lexical information from long-term memory (e.g., word forms, punctuation, grammatical rules), CE resources can be devoted to higher level processing of the information (e.g., text generation, emending word meanings, word sequencing, sentence restructuring) and contribute to proficient written expression (McCutchen, 2000, Swanson, 1996). The majority of empirical investigations examining the interplay among CE and PH STM processes in written expression have focused on neurotypical high school students whose PH STM capacity is usually fully developed and automatized by adolescence (Tillman, Eninger, Forssman, & Bohlin, 2011). As expected, CE abilities alone contribute significantly to youth's written expression abilities in these investigations (Hoskyn and Swanson, 2003, Vanderberg and Swanson, 2007).

Extant studies of written expression abilities in children with ADHD have conventionally compared their score profiles to neurotypical children on various qualitative (e.g., lexicon, grammar, structure) and quantitative (e.g., text length, accuracy) expressive writing indices (cf. Graham, Fishman, Reid, & Hebert, 2016, for a review). These investigations report significant written expression deficits in ADHD that are not attributable to basic knowledge about writing (Re and Cornoldi, 2010) or core clinical symptoms such as inattentiveness and hyperactivity (DeBono et al., 2012), although Langmaid et al., (2014) reported a significant relation between inattention symptom severity and poorer hand writing in children with ADHD. Only one study to date, however, has attempted to examine the relative influence of CE and PH STM processes on writing expression ability in children with ADHD relative to neurotypical (NT) children (Rodriguez, Torrance, Betts, Cerezo, & Garcia, 2017). Children's performance on a battery of simple (e.g., digit span forward) and complex (e.g., reading span) memory span tasks were analyzed using a series of hierarchical linear regression models, and revealed significant contributions of both CE and PH STM on written expression competence. The unique contribution of these processes and whether they act in tandem, however, could not be addressed in the study because complex span measures involve both WM storage (PH STM) and processing (CE) components.

An additional factor hypothesized to influence children's written expression is their oral expression abilities, which develop shortly after the PH STM subsystem is able to upload information from long-term memory (Diamond, Gerde, & Powell, 2008, Gathercole and Baddeley, 1993, Hoskyn, 2010) and maintain the information in conscious awareness. These collective abilities precede the development of written expression by approximately two years (Vygotsky, 1978). As expected, young children are superior in oral expression relative to written expression, and written expression is highly dependent upon oral language, particularly among nascent writers (cf. Shanahan, MacArthur, Graham, & Fitzgerald, 2006, for a review). Extant evidence suggests that children diagnosed with ADHD experience particular difficulties using language to express thoughts orally and emit coherent oral narratives (Purvis and Tannock, 1997, Tannock, Purvis, & Schachar, 1993, Tannock and Schachar, 1996). They often neglect to apply correct mechanical structure grammar, syntax, and semantics, and fail to acknowledge the pragmatic requirements of oral expression (Bruce, Thernlund, & Nettelbladt, 2006, Green, Johnson, & Bretherton, 2014).

The interplay between two of the three factors theorized to underlie written communication abilities discussed thus far – viz., PH STM and oral expression abilities – has also been considered in recent years. For example, oral expression among children with poor PH STM abilities is often characterized by the use of short utterances (Adams & Gathercole, 1995, Blake, Austin, Cannon, Lisus, & Vaughan, 1994), immature syntax, and limited vocabulary (Adams & Gathercole, 1995). As a result, CE and PH STM abilities are implicated in both the buffering of oral expression prior to speech and the uploading of learned lexical and grammatical rules from long-term memory. The extent to which they work independently or act as coordinated cognitive processes to influence children's written expression, however, is unknown currently.

The current study is the first to fractionate the anatomically distinct CE from the PH STM subsystem while concomitantly examining the potential contribution of oral expression to ascertain their unique and potentially interactive relations to ADHD-related written expression difficulties. Understanding the relative contributions of these processes and whether the CE and/or PH STM work in tandem with oral expression abilities is critical to designing and targeting effective interventions. For example, designing interventions for written expression deficits may require strengthening CE, PH STM, and oral expression singularly or in tandem depending on their unique and/or interactive contributions.

Upper level CE, lower level modality-specific PH STM subsystem, and Oral Expression were hypothesized to serve as significant mediators of ADHD-related written expression difficulties in the planned mediation analyses, whereas the VS STM subsystem was expected to play a nonsignificant role based on extant literature (Adams & Gathercole, 1995, McCutchen, 2000, Vanderberg and Swanson, 2007). The unique contributions of CE and PH STM were scrutinized afterwards using a parallel multiple mediation model based on the expectation that both variables would prove to be significant mediators of ADHD-related written expression deficits, but that one may represent an epiphenomenon of the other when modeled conjointly (Orban, Rapport, Friedman, Eckrich, & Kofler, 2018). A defining feature of the parallel multiple mediation model is the constraint that each potential mediator is modeled controlling for (holding constant) all other mediators in the model to allow competing hypotheses of mechanisms or processes to be examined independently of one another (Hayes, 2018).

A final serial multiple mediator model involving WM components and Oral Expression was planned based on the expectation that one or both WM components and Oral Expression would prove to be significant mediators in the preceding analyses to determine the extent to which these variables influence the Diagnostic Status to Written Expression relation independently and/or collectively. WM components found to be significant mediators in the parallel model were entered initially, followed by Oral Expression based on extensive research confirming the developmental sequencing of these processes and abilities in children (Diamond et al., 2008, Hoskyn, 2010). We hypothesized that one or both WM variables and Oral Expression would continue to be significant, independent mediators of the ADHD-Written Expression relation, but would also work in tandem based on theoretical accounts that oral expression is developmentally contiguous to PH STM. If supported, this finding would indicate that ADHD-related written expression difficulties reflect both an independent contribution by and interplay between WM and oral expression processes.

Method

Participants

The sample comprised boys ($n = 60$) aged 8–12 years ($M = 9.35$, $SD = 1.28$) referred to or recruited by an urban university-based child study clinic. All participants lived within 60 miles of the greater Orlando, FL area and were recruited through community resources (e.g., referrals from primary care physicians, community mental health clinic, public, and homeschool systems, self-referral) whose parents agreed to have them participate in developmental/clinical research studies. A psychoeducational evaluation was provided *pro bono* to the parents of all participants. Two groups of children participated in the study: boys with ADHD-Combined Presentation and neurotypical (NT) boys without a psychological disorder. Sample ethnicity was mixed consistent with the surrounding population demographics and included 40 Caucasian non-Hispanic (67%), 13 Hispanic fluent English-speaking (22%), 2 African-American (3%), and 5 (8%) boys of mixed racial/ethnic background. None of the participating children spoke multiple languages at home (i.e., English only). Children with a history of (a) gross neurological, sensory, motor impairment, or seizure disorder by parent report, (b) psychosis, or (c) Full-Scale IQ score ≤ 80 were excluded. All parents and children provided their informed written consent/assent to participate in the study, and the university's Institutional Review Board approved the study prior to the onset of data collection. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Group assignment

All children and their parents participated in a detailed, semi-structured clinical interview using all modules of the Kiddie Schedule for Affective Disorders and Schizophrenia for School-Aged Children (K-SADS). The K-SADS assesses onset, course, duration, severity, and impairment of current and past episodes of psychopathology in children and adolescents based on DSM-5 criteria. Its psychometric properties are well established, including interrater agreement of 0.93 to 1.00, test-retest reliability of 0.63–1.00, and concurrent (criterion) validity between the K-SADS and psychometrically established parent rating scales (Kaufman et al., 1997). Thirty-three boys meeting all of the following criteria were included in the ADHD-Combined Presentation group: (1) an independent diagnosis by the directing clinical psychologist using DSM-5 criteria for ADHD-Combined Presentation based on K-SADS interview with parent and child; (2) parent ratings exceeding the 90th percentile on the Attention-Deficit/Hyperactivity Problems DSM Oriented scale of the Child Behavior Checklist (CBCL; Achenbach and Rescorla, 2001) or exceeding the criterion score on the parent version of the ADHD-Combined subtype subscale of the Child Symptom Inventory-4: Parent Checklist (CSI-P; Gadow, Sprafkin, Salisbury, Schneider, & Loney, 2004); and (3) teacher ratings exceeding the 90th percentile on the Attention-Deficit/Hyperactivity Problems DSM Oriented scale of the Teacher Report Form (TRF; Achenbach and Rescorla, 2001) or exceeding the criterion score on the teacher version of the ADHD-

Combined subtype subscale of the Child Symptom Inventory-4: Teacher Checklist (CSI-T; Gadow et al., 2004). Eleven (33%) of the ADHD boys were on a psychostimulant regimen for treatment of their ADHD symptoms (24-h washout period prior to each testing session). Twenty-seven boys met all of the following criteria and were included in the NT group: (1) no evidence of any clinical disorder based on parent and child K-SADS interview; (2) normal developmental history by parental report; (3) ratings within 1.5 *SDs* of the mean on all CBCL and TRF scales; and (4) parent and teacher ratings within the nonclinical range on all CSI subscales.

Measured intelligence and socioeconomic status

Children were administered the WISC-IV or -V to obtain an overall estimate of intellectual functioning based on each child's estimated Full-Scale IQ (FSIQ; Wechsler, 2003; 2014). The changeover to the fifth edition was due to its release during the course of the study and to provide parents with the most up-to-date intellectual evaluation possible. Hollingshead Four Factor Index of Social Status (Hollingshead, 1975) was used to calculate socioeconomic status (SES) based on parental education, occupation, age, and marital status. Standardized mean SES scores for families of children with ADHD and NT children were 49.63 (*SD* = 9.97) and 53.17 (*SD* = 10.17), respectively.

Procedures

The WM tasks (described below) were programmed using SuperLab Pro 2.0 (Cedrus Corporation, 2002) and administered as part of a larger battery that required the child's presence for approximately 3 h per session across four consecutive Saturday assessment sessions. Participants completed all tasks while seated alone, approximately 0.66 m from a computer monitor, in an assessment room. Performance was monitored at all times by the examiner, who was stationed just outside the child's view to provide a structured setting while minimizing performance improvements associated with examiner demand characteristics (Power, 1992). All participants received brief (2–3 min) breaks following each task, and preset longer (10–15 min) breaks after every two to three tasks to minimize fatigue. The Kaufman Test of Educational Achievement 2nd edition (KTEA-II; Kauffman and Kauffman, 2004) was administered during two separate week-day testing sessions to minimize fatigue.

Measures

Written expression

Age-corrected, standardized scores from the Written Expression subtest of the KTEA-II (Kauffman and Kauffman, 2004) served as the dependent variable to measure the extent to which boys were able to communicate effectively in writing. The examiners (trained doctoral level graduate students) presented questions orally using a visual aid (stimulus book with written instruction visible to the child) per standardized instructions while the child followed along with an age/grade-appropriate storybook. Children follow a set of characters throughout the story and perform a variety of tasks during the telling of

the story including: writing sentences from dictation, inserting punctuation and capitalization, filling in missing words with correct subject-verb agreement, completing sentences, combining sentences, writing compound and complex sentences, and writing an essay based on the story. The participant's written responses were scored following standardized KTEA-II manual directions for the five categories (task completion, sentence structure, word form, capitalization, and punctuation) and converted to a standardized score based on age normative values. Neither spelling nor writing fluency contributes to the overall score. The psychometric properties and expected patterns of relationships between the KTEA-II Written Expression subtest and other measures of educational achievement are well established (Kauffman and Kauffman, 2004).

Oral expression

Age-corrected, standardized Oral Expression subtest scores from KTEA-II (Kauffman and Kauffman, 2004) were used to assess how well boys communicate ideas in speech. Participants were shown an illustration of a real-life scenario and verbally instructed to perform increasingly complex speaking tasks while utilizing appropriate pragmatics, syntax, semantics, and grammar. Their answers were recorded manually by the examiner and scored following standardized KTEA-II manual directions. Raw scores were converted to standardized scores based on age normative values. The psychometric properties and expected patterns of relations between the KETA-II Oral Expression subtest and other measures of educational achievement are well established (Kauffman and Kauffman, 2004).

Working memory tasks

The working memory tasks used in the current study are identical to those described by Rapport et al. (2008)¹ Each child was administered four phonological conditions (i.e., set sizes 3, 4, 5, and 6) and four visuospatial conditions (i.e., set sizes 3, 4, 5, and 6) across the four testing sessions. The four working memory set size conditions each contained 24 unique trials of the same stimulus set size, and were counterbalanced across the four testing sessions to control for order effects and potential proactive interference effects across set size conditions. Stimuli correct per trial served as the dependent variable for the WM tasks. Previous studies of ADHD and NT children reveal large magnitude between-group differences on these tasks (Rapport et al., 2008). Past investigations report strong reliability and validity of the PH WM task evidenced by high internal consistency ($r = 0.82\text{--}0.97$), significantly large correlations ($r = 0.50\text{--}0.71$) with established measures of working memory such as the WISC-IV Working Memory Index (Alderson et al., 2015, Raiker, Rapport, Kofler, & Sarver, 2012), and the expected pattern of relations between the isolated CE construct and ecologically valid outcomes such as objectively measured activity level (Rapport et al., 2009) attentive behavior (Kofler, Rapport, Bolden, Sarver, & Raiker, 2010), and impulsivity (Raiker et al., 2012).

¹PH WM and VS WM performance data for a small subset of the current sample were used in previous studies (references masked for review) to examine conceptually unrelated hypotheses. The associations among the Written Expression, Oral Expression, and WM tasks used in the current study have not been examined previously.

Phonological working memory (PH WM)

The PH WM tasks are similar to the Letter-Number Sequencing subtest on the WISC-V (Wechsler, 2014), and assess phonological working memory based on Baddeley's (2007) model. Children were presented a series of jumbled numbers and a capital letter on a computer monitor. Each number and letter (4 cm height) appeared on the screen for 800 ms, followed by a 200 ms interstimulus interval. The letter never appeared in the first or last position of the sequence to minimize potential primacy and recency effects, and trials were counterbalanced to ensure that letters appeared an equal number of times in the other serial positions (i.e., position 2, 3, 4, or 5). Children were instructed to recall the numbers in order from smallest to largest, and to say the letter last (e.g., 4 H 6 2 is recalled correctly as 2 4 6 H). All children completed five practice trials prior to each administration and achieved the minimum of 80% accuracy on training trials. Two trained research assistants, shielded from the participant's view, recorded oral responses independently. Interrater reliability was calculated for all task conditions for all children, and ranged from 0.98 to 0.99.

Visuospatial working memory (VS WM)

Children were shown nine squares arranged in three offset vertical columns on a computer monitor. A series of 2.5 cm diameter dots (3, 4, 5, or 6) were presented sequentially in one of the nine squares during each trial such that no two dots appeared in the same square on a given trial. All dots presented within the squares were black; the exception being a red dot that never appeared as the first or last stimulus in the sequence. Children were instructed to indicate the serial position of black dots in the order presented by pressing the corresponding squares arranged in three offset vertical columns on a computer keyboard, and to indicate the serial position of the red dot last.

Working memory factors

Estimates of the central executive (CE), phonological short-term memory (PH STM), and visuospatial short-term memory (VS STM) were computed at each set size. Briefly, the PH and VS STM systems are functionally and anatomically independent, with the exception of a shared (domain-general) CE controller (Baddeley, 2007). Consequently, statistical regression techniques were used to provide reliable estimates of the CE and its subsidiary PH and VS STM subsystems as described in the following section. Precedence for using shared variance to statistically derive CE and/or PH/VSTM variables is found for working memory components in Alderson et al. (2015), Engle et al. (1999), Kofler, Sarver, and Wells (2015), Raiker et al. (2012), Rosen and Engle (1997), and Swanson and Kim (2007).

PH/VSTM factors

PH STM composite scores were computed by averaging each child's score across set sizes using the following procedure. Scores on the VS WM task were regressed out of scores on the PH WM task to remove common variance associated with the domain-general central executive. The four PH STM scores were then fixed to one factor via principal components factor analysis (factor loadings = 0.62–0.81) using scores at each of the four set sizes to provide an overall estimate of the contribution of PH STM independent of shared CE

influences. A complementary procedure was performed whereby scores on the PH WM task were regressed out of scores on the VS WM and fixed to one factor (factor loadings = 0.58–0.75) to obtain an overall estimate of VS STM independent of CE influences.

CE factor

Two unstandardized predicted scores were computed by regressing VS WM scores onto PH WM scores at each set size, and vice versa. The two scores at each set size were averaged to provide an estimate of CE functioning at each set size. These four CE scores were then fixed to one factor via principal components factor analysis (factor loadings = 0.76–0.86) to provide an overall estimate of CE independent of the two STM subsystems.

Results

Preliminary analysis

All independent, dependent, and mediating variables were screened for multivariate outliers using Mahalanobis distance tests ($p < 0.001$) and univariate outliers as reflected by scores exceeding 3.0 *SDs* from the mean in either direction. No significant outliers were identified. As expected, scores on the parent and teacher behavior rating scales were significantly higher for the ADHD group relative to the NT group (see Table 1). Boys with ADHD and NT boys did not differ on age ($p = 0.31$), SES ($p = 0.18$), or FSIQ ($p = 0.11$). Four ADHD and one NT child met diagnostic criteria for co-occurring learning disorder² in written expression. We considered including FSIQ as a covariate despite the lack of between-group differences due to the investigation's focus on children's WM processes; however, doing so would result in removing substantial variance associated with WM from WM (Dennis et al., 2009, Miller and Chapman, 2001). Consistent with past studies (e.g., Alderson et al., 2010, Friedman et al., 2018; Kofler et al., 2014, Rapport et al., 2008), we adopted a more conventional approach that involved removing reliable variance associated with the WM CE factor (described above) from FSIQ and examining between-group differences in FSIQ without the influence of the CE.³

We examined whether ADHD-related WM deficits are differentially related to the varying Written Expression subdomains (i.e., Punctuation, Word Form, Capitalization, Structure, and Task) by conducting a mixed-model ANOVA (2 Diagnostic Status x 5 Written Expression Subdomain) examining potential between-group differences in KTEA Written Expression components. The results revealed a significant main effect of Diagnostic Status such that children with ADHD made more errors than NT children on the Written Expression subdomains overall, $F(1, 42) = 5.61, p = .023$. There was also a significant main effect for Written Expression subdomains, $F(4, 168) = 48.54, p < .001$. Post hoc tests revealed that there were significantly more

²Criteria for a learning disorder (LD) in written expression was defined by a Standard Score (SS) of more than 1 SD (15) below the normative mean (i.e., $SS < 85$) on the KTEA-II Written Expression subtest. The association between diagnostic status and written expression was not moderated by the presence of a LD in written expression ($p = .92$). Further, although 15 ADHD children received educational accommodations, the presence of such interventions did not moderate ($p = .62$) the diagnostic status/written expression relation.

³Alternative approaches were considered but not adopted because they also share substantial variance with WM (e.g., the WISC-IV and -V General Ability Index [GAI] is comprised of the Verbal Comprehension and Perceptual Reasoning Indices, and shares 25% to 40% of variance with WM, respectively).

Table 1. Sample and demographic variables.

	ADHD		Neurotypical		Total	
	Mean	SD	Mean	SD	<i>t</i>	Cohen's <i>d</i>
Age	9.18	1.21	9.52	1.34	1.02	0.27
FSIQ	106.36	10.82	110.63	9.52	0.11	0.42
FSIQ _{res}	-0.64	10.59	0.78	9.46	0.54	0.14
SES	49.63	9.97	53.17	10.19	1.38	0.35
CBCL AD/HD Problems	72.00	7.26	54.52	8.67	8.50**	2.19
TRF AD/HD Problems	67.63	6.91	53.00	5.40	3.82**	2.35
CSI-P: ADHD, Combined	77.06	9.27	49.00	12.17	10.13**	2.59
CSI-T: ADHD, Combined	67.82	14.55	47.96	9.98	6.28**	1.48
Oral Expression	99.15	13.63	106.41	13.63	2.05*	0.53
Written Expression	100.21	12.91	107.48	12.83	2.18*	0.56
Phonological STM Factor Score	-0.35	1.10	0.43	0.65	3.24**	0.86
Visuospatial STM Factor Score	-0.28	0.92	0.34	0.99	2.51*	0.65
Central Executive Factor Score	-0.49	0.87	0.60	0.82	4.94**	1.29

Note: ADHD: attention-deficit/hyperactivity disorder; FSIQ: Full-Scale Intelligence Quotient; FSIQ_{res}: Full-Scale Intelligence Quotient with central executive factor score removed; SES: socioeconomic status; CBCL: Child Behavior Checklist; TRF: Teacher Report Form; CSI-P: Child Symptom Inventory: Parent severity *T*-scores; CSI-T: Child Symptom Inventory: Teacher severity *T*-scores; STM: short-term memory; * $p \leq 0.05$, ** $p \leq 0.01$.

Punctuation errors than any other errors. There were also significantly more Capitalization and Structure errors than Word Form and Task errors. As expected from the literature (Graham et al., 2016), there was no significant interaction between Diagnostic Status and Written Expression subdomain, $F(4, 168) = .748, p = .561$. We elected not to include each of these subdomains measures in the mediation models due to a lack of power and high risk of family-wise error.

Tier I: Intercorrelations and simple mediation models

Zero-order intercorrelations between all factor scores were computed and are shown in Table 2.

Separate simple mediation models were tested to examine the extent to which WM and Oral Expression variables attenuated the relation between Diagnostic Status and Written Expression. Mediation analyses were completed using bias-corrected bootstrapping to minimize Type II error as recommended by Shrout and Bolger (2002). Bootstrapping was used to establish the statistical significance of all total, direct, and indirect effects. All continuous variables were standardized *z*-scores based on the full sample to facilitate between-model and within-model comparisons and allow unstandardized regression coefficients (*B* weights) to be interpreted as Cohen's *d* effect sizes when predicting from a dichotomous grouping variable (Hayes, 2009). The PROCESS script for SPSS (Hayes, 2014) was used for all analyses, and 10,000 samples were derived from the original sample ($n = 60$) by a process of resampling with replacement (Shrout and Bolger, 2002). Effect ratios (indirect effect divided by total effect) were calculated to estimate the proportion of each significant total effect that was attributable to the mediating pathway (indirect effect). Cohen's *d* effect sizes, standard errors, 90% confidence intervals, and effect ratios are shown in Figure 1. Ninety percent confidence intervals were selected over 95%, due to concerns that the latter is less conservative in determining full vs. partial mediation. Briefly, the narrower 90% confidence interval is less likely to include 0.0, and therefore is likely to result in a more conservative conclusion

Table 2. First-order correlations.

	1	2	3	4	5	6	7	8
1. Diagnostic status(NT = 0, ADHD = 1)								
2. Age	-.13							
3. SES	-.18	-.14						
4. FSIQ	-.21	-.15	.26*					
5. Central Executive	-.54*	.41*	.14	.26*				
6. PH STM	-.39*	.19	.18	.23	.68*			
7. VS STM	-.31*	.35*	.01	.10	.63*	-.14		
8. Oral Expression	-.26*	-.07	.15	.42*	.22	.28*	-.01	
9. Written Expression	-.28*	.13	.25	.46*	.34*	.45*	-.03	.40*

ADHD: attention-deficit/hyperactivity disorder, PH STM: phonological short-term memory, SES: Socioeconomic Status, FSIQ = Full-Scale IQ, NT: Neurotypical, VS STM: visuospatial short-term memory. Correlations reflect Pearson's correlation coefficients. *Correlation is significant based on $p < 0.05$.

regarding the magnitude of the relation between Diagnostic Status and Written Expression after accounting for the mediator. In contrast, the wider 95% confidence interval increases the likelihood that the confidence interval for direct effect will include 0.0, indicating that Diagnostic Status and Written Expression are no longer related significantly after accounting

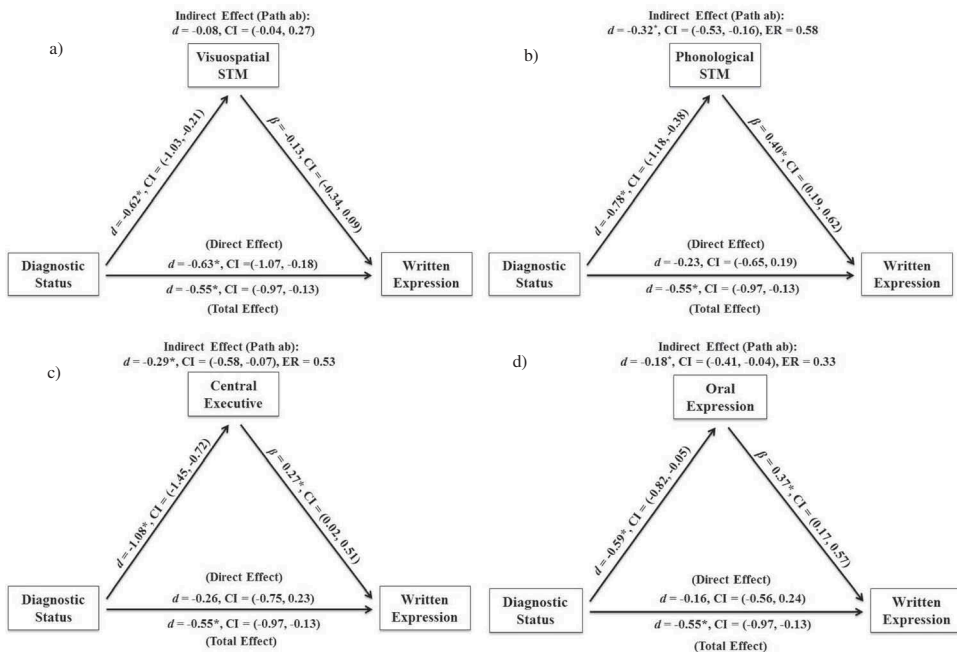


Figure 1. CI: confidence interval (90%), ER: effect ratio, STM: short-term memory. Schematics depicting the effect sizes, standard errors and B coefficients of the total, direct, and indirect pathways for the mediating effect of (a) Visuospatial Short-Term Memory (b) Phonological Short-Term Memory, (c) Central Executive, and (d) Oral Expression on Written Expression. Cohen's d for the direct and total effect pathways reflects the impact of ADHD Diagnostic Status on Written Expression before (total effect) and after (direct effect) taking into account the mediating variable. *Effect size (or B weight) is significant based on 90% confidence intervals that do not include 0.0 (Shrout and Bolger, 2002); values from the mediator to Written Expression reflect B weights due to the use of two continuous variables in the calculation of the direct effect.

for the mediator. For discussion and specific examples of this phenomenon, see Shrout and Bolger (2002).

A large magnitude effect size was predicted based on established relations between ADHD and WM ($d_s = 1.89, 2.31$; Rapport et al., 2008), ADHD and Written Expression ($d = 1.09$; Kauffman and Kauffman, 2004), WM and Written Expression ($r = 0.53$; Vanderberg and Swanson, 2007), and Written Expression and Oral Expression ($r = 0.65$; Kauffman and Kauffman, 2004). Mediation analysis using bias-corrected bootstrapping requires 34 total participants to achieve 0.80 power (Fritz and Mackinnon, 2007) and 60 individuals participated in the current study.

Examination of the total effect of Diagnostic Status (ADHD, NT) on Written Expression for all models (Figure 1) revealed a significant relation ($d = -0.55$, CI = $-0.97, -0.13$), such that a diagnosis of ADHD was associated with medium effect size Written Expression differences prior to accounting for potential mediators. As expected, VS STM (Figure 1a) was not a significant mediator of the Diagnostic Status to Written Expression relation (90% CI included 0.0). In contrast, PH STM was a significant, full mediator ($d = -0.32$; 90% CI = $-0.53, -0.16$) and accounted for 58% of the variance in the Diagnostic Status to Written Expression relation (Figure 1b). CE ($d = -0.29$; 90% CI = $-0.58, -0.07$) and Oral Expression ($d = -0.18$; 90% CI = $-0.41, -0.04$) were also significant, full mediators and accounted for 53% and 33% of the variance in ADHD-related Written Expression deficits (Figure. 1c and 1d), respectively.

Tier II: Parallel and serial multiple mediation models of the diagnostic status to written expression relation

Power analysis for parallel and serial multiple mediation models

Based on available evidence, a sample of 60 participants is powered adequately to conduct bias-corrected, bootstrapped tests of two mediators, as in the serial and parallel models reported below. Specifically, Briggs (2006) reported a series of simulations to estimate power for models with two mediators using a smaller sample ($n = 50$) and reported power of 0.90–0.92 to detect the total indirect effect and 0.74–0.75 for each independent mediator. Assuming full mediation, which is justifiable based on our Tier I finding that the relation between Diagnostic Status and Written Expression was significantly attenuated after including PH STM or Oral Expression within the model, power for a sample size of 50 is 0.89–0.92 for the total indirect effect, and 0.73–0.77 for each individual indirect effect. Our sample of 60 is powered appropriately for testing models with two mediators given evidence of sufficient power for individual effects that approach .80 with a sample size of 50.

Parallel multiple mediation model

Tier II involved a two-step process to determine the most parsimonious model for characterizing WM's association with ADHD-related deficits in Written Expression. A parallel multiple mediation model (Hayes, 2013) that included both CE and PH STM (Figure 2) was used initially to examine whether one or both processes were responsible for the Tier I findings.⁴ This model was predicated on empirical evidence that PH STM

⁴A defining feature of the parallel multiple mediation model is the constraint that each potential mediator is modeled controlling for (holding constant) all other mediators in the model to allow competing hypotheses of mechanisms or processes to be examined independently of one another (Hayes, 2018).

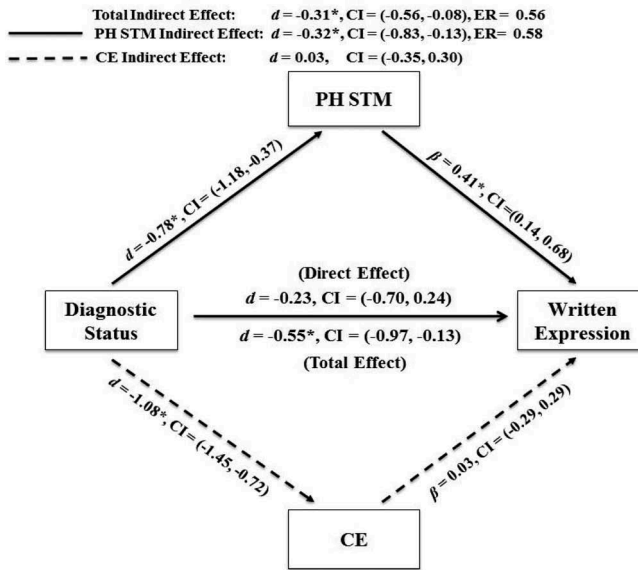


Figure 2. CI: confidence interval (90%), ER: effect ratio. Effect sizes, standard errors, and β coefficients of the total, direct, and indirect pathways for parallel mediation of PH STM and CE on the relation between Diagnostic Status and Written Expression are depicted in the figure. PH STM Indirect Effect = mediating effect of PH STM independent of Central Executive on Written Expression. CE Indirect Effect = mediating effect of CE independent of PH STM on Written Expression. Total Indirect Effect = collective influence of both mediation pathways. PH STM: Phonological Short-Term Memory; CE: Central Executive. *Indicates significant path (90% CI does not include 0.0).

and CE processes stem from neuroanatomically distinct cortical structures (D'Esposito et al., 1995) but are expected to be intercorrelated because individuals with well-developed CE typically have well-developed PH STM and vice versa ($r = 0.68$). Inspection of Figure 2 indicates that PH STM ($d = -0.32$, ER = 0.58) but not CE (90% CI includes 0.0) explained unique variance in the ADHD/Written Expression relation. As a result, only PH STM was included in further model testing.

Serial multiple mediation model

A serial multiple mediation model was used subsequently to examine the extent to which significant Tier I (Oral Expression) and the contributing parallel mediator model variable (PH STM) alone and/or interactively account for between-group differences in Written Expression (Figure 3) using the PROCESS script for SPSS (Hayes, 2014). PH STM was entered into the model initially followed by Oral Expression based on extensive empirical evidence (a) confirming the temporal development of these processes and abilities (Diamond et al., 2008, Gathercole and Baddeley, 1993, Hoskyn, 2010), and (b) that PH STM must be activated to upload and maintain information from long-term memory (e.g., words, syntax, grammatical rules) as a requisite to expressing oneself orally. Despite the established temporal development of PH STM and Oral Expression based on extant research, causal inferences cannot be confirmed in the present study due to the use of a cross-sectional design.

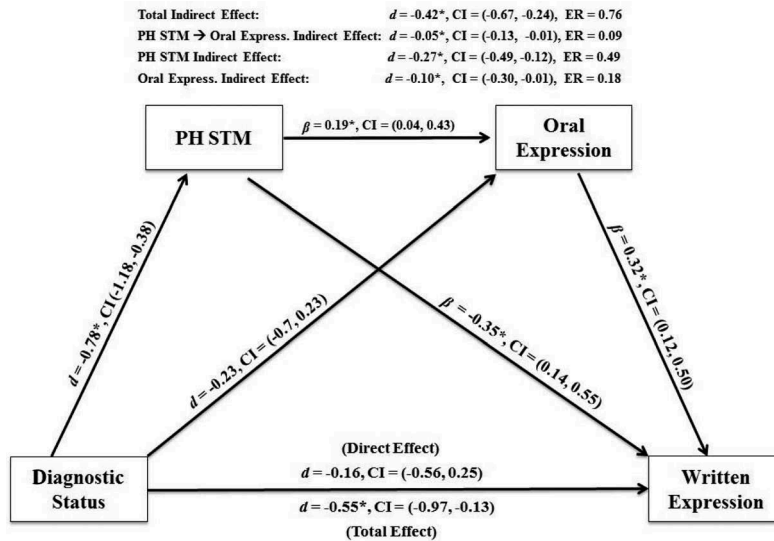


Figure 3. CI: confidence interval, ER: effect ratio. Diagram depicting effect sizes, standard errors, and β coefficients of the total, direct, and indirect pathways for serial multiple mediation of PH STM and Oral Expression on the relationship between Diagnostic Status and Written Expression. Cohen's d for the Direct and Total pathways reflects the impact of ADHD Diagnostic Status on Written Expression before (Total Effect) and after (Direct Effect) taking into account the mediating variables. *Effect size (or β -weight) is significant based on 90% confidence intervals that do not include 0.0 (Shrout and Bolger, 2002); values from the mediating variables to Written Expression are described as β -weights due to the use of two continuous variables in the calculation of the direct effect. PH STM Indirect Effect represents the mediating effect of PH STM independent of Oral Expression on Written Expression. The Oral Expression Indirect Effect represents the mediating effect of Oral Expression independent of PH STM on Written Expression. The PH STM \rightarrow Oral Expression indirect effect represents the mediating effect of the shared influence of PH STM and Oral Expression on Written Expression. Total Indirect Effect represents the collective influence of all three mediation pathways.

The total effect of Diagnostic Status on Written Expression, $d = -0.55$, was significantly attenuated when PH STM and Oral Expression were included as mediators, $d = -0.42$, such that the combined effect of all three mediating pathways accounted for 76% of the Diagnostic Status/Written Expression relation (Figure 3, Total Indirect Effect), and the direct effect of Diagnostic Status on Written Expression was no longer detectable (90% CI included 0.0, indicating no effect). This combined effect was carried primarily by the mediating role of PH STM ($d = -0.27$) independent of Oral Expression, and accounted for 49% of the variance between Diagnostic Status and Written Expression (Figure 3, PH STM Indirect Effect). The second largest effect was carried by the mediating role of Oral Expression ($d = -0.10$) independent of the influence of PH STM, and accounted for 18% of the variance between Diagnostic Status and Written Expression (Figure 3, Oral Expression Indirect Effect). Lastly, a small albeit significant effect, $d = -0.05$, resulted from the mediating role of PH STM working in tandem with Oral Expression and accounted for 9% of the variance in the Diagnostic Status to Written Expression relation.

Collectively, these findings indicate that the moderate magnitude influence of PH STM and Oral Expression on Written Expression observed in Tier I is largely driven by

PH STM's impact on the boys' ability to perform written expression activities. The high effect ratio (76% of variance explained), coupled with the nonsignificant residual association between Diagnostic Status and Written Expression, indicates that the interactive effects of PH STM deficits and oral expression difficulties play an important role in understanding the written expression difficulties commonly observed among boys with ADHD.

Discussion

The current study is the first to fractionate the anatomically distinct PH STM and VS STM subsystems from domain-general CE working memory processes while concomitantly examining oral expression abilities to quantify their potentially unique and shared contributions to ADHD-related written expression difficulties. Planned mediation analyses were conducted – beginning with simple models and proceeding to multiple mediation models based on established developmental sequencing of hypothesized processes and abilities – to elucidate the extent to which they independently and/or jointly contributed to the established ADHD-related Written Expression difficulties relation.

Results gleaned from the simple mediation models were consistent with past investigations involving NT children, wherein CE and PH STM but not VS STM accounted for significant differences in children's written expression abilities (McCutchen, 1996, Rodriguez et al., 2017, Vanderberg and Swanson, 2007). The finding is also consistent with the premise that the association between PH STM and writing occurs in young children because they depend on similar language processes (Gathercole and Baddeley, 1990). The independent contribution of WM CE processes to the Diagnostic Status/Written Expression relation did not remain when examined in the context of a multiple mediator parallel model, which estimates the unique influence of each variable (CE, PH STM) entered into the model while holding the other constant. Similar findings are often reported in cases in which two variables are correlated and significant mediators when modeled separately, but only one is a true mediator of the relation and the other a correlated process that arises from but does not causally influence the process (cf. Hayes, 2018, for an expanded discussion of epiphenomena influences).

The nonsignificant contribution of CE processes to the Diagnostic Status/Written Expression relation may appear unexpected *prima facie*, but likely reflects methodological and age differences between the current study and past studies investigating the phenomenon. For example, Rodriguez et al. (2017) reported significant contributions of both CE and PH STM to written expression differences in 8- to 12-year-old children with ADHD relative to NT children; however, CE was assessed using a complex memory span task that also required PH STM processes such that their independent contributions to written expression could not be determined. Significant CE but not PH STM to written expression relations have also been reported by other investigators (Hoskyn and Swanson, 2003, Vanderberg and Swanson, 2007); however, the participants in both studies were high school aged NT youths whose PH STM capacity was likely fully developed and automatized in its ability to upload lexical information (e.g., grammatical rules, spelling) from long-term memory based on longitudinal studies of WM development (Tillman et al., 2011). Under these circumstances, CE resources can

be devoted to higher level processing of the information such as text generation, word sequencing, and sentence restructuring that contribute to written expression abilities (McCutchen, 2000). In contrast, children within the pediatric age range rely primarily on PH STM when engaged in writing and previous investigations indicate that both PH STM subsidiary system components (viz., storage and maintenance via covert rehearsal) are significantly underdeveloped in children with ADHD (ES = 1.15–1.98; Bolden, Rapport, Raiker, Sarver, & Kofler, 2012).

The potential contribution of a third factor – oral expression abilities – was also hypothesized to mediate ADHD-related written expression difficulties based on recent findings that children with ADHD often demonstrate difficulties applying the mechanical and pragmatic requirements of oral expression (Green et al., 2014), coupled with the established nature of oral expression as a developmental antecedent to children's ability to communicate their thoughts in writing (Shanahan et al., 2006, Vygotsky, 1978). As expected, Oral Expression served as a significant stand-alone mediator, and also interacted with its developmental precursor (viz., PH STM) to mediate the Diagnostic Status to Written Expression relation when evaluated in the context of a multiple serial mediation model.

Collectively, the contributions of PH STM and Oral Expression in mediating ADHD-related Written Expression difficulties accounted for 76% of the variance and reflect several cognitive processes. For example, the independent contribution of PH STM likely reflects the extent to which children can briefly store and maintain words and grammatical rules (e.g., lexical, syntactic, punctuation, capitalization) uploaded from long-term memory, whereas Oral Expression's independent contribution represents pragmatic applications such as idea generation, translation of ideas into words, and forming sentences. These latter processes consume a greater proportion of available resources of the limited capacity PH STM subsystem and interact by decreasing the amount of information that can be stored and processed when writing.

Despite incorporating multiple methodological (e.g., multi-method/multi-informant diagnostic procedures, comprehensive measurements of WM, age-normed, nationally standardized assessments of oral and written expression) and statistical (e.g., bootstrapped mediation) refinements in the study, several caveats warrant mention. For example, the Oral Expression to Written Expression relation may be overestimated moderately due to drawing both subtests from the same achievement battery. The KTEA-II intra-battery correlation for the two subtests ($r = 0.50$; Kauffman and Kauffman, 2004), however, is highly similar to the inter-battery correlation between KTEA-II Oral Expression and WIAT-II Written Expression ($r = 0.46$), and between KTEA-II Oral Expression and WJ-III Written Expression ($r = 0.44$). The inter-battery similarities suggest that the two skills are highly correlated because oral expression abilities include knowledge of lexical content necessary for expressing oneself in writing. Other related competencies such as spelling (Capodieci, Serafini, Dessuki, & Cornoldi, 2018, Kroese, Hynd, Knight, Hiemenz, & Hall, 2000) and/or handwriting (Capodieci, Lachina, & Cornoldi, 2018, Langmaid et al., 2014) may also contribute to ADHD-related written expression difficulties. These abilities, however, were neither assessed in the current study nor contributed to the standardized scoring of the KTEA Written Expression subtest. We also acknowledge that children's performance on

cognitive tasks administered in highly controlled settings may not generalize to dissimilar settings.

The generalizability of the results of the current study is limited to boys with ADHD-Combined Presentation. The decision to study this population was based upon the comparatively lower rate of writing difficulties among girls (Berninger, Nielsen, Abbott, Wijsman, & Raskind, 2008, Farrington et al., 2014) and recent studies demonstrating morphological (Dirlikov et al., 2015; Mahone et al., 2011) and neurocognitive (O'Brien, Dowell, Mostofsky, Denckla, & Mahone, 2010) differences that may differentially impact written and oral expression abilities. Similarly, youths diagnosed with ADHD-I exhibit a significantly different neurocognitive profile than those diagnosed with ADHD-C (Thaler, Bello, & Etcoff, 2013, cf. Willcutt, Doyle, Nigg, Faraone, & Pennington, 2005, for a meta-analytic review). Despite recent studies indicating that rates of writing disorders are similar among children with ADHD-C and ADHD-I (Mayes and Calhoun, 2006, Mayes, Frye, Breaux, & Calhoun, 2018), the cognitive processes underlying written and/or oral expression difficulties may be particular to the presentation type. For example, children with ADHD-I are often characterized by exhibiting slow processing speed suggesting poor *updating* of information from long-term memory – a separate mechanism from deficient memory storage (STM) and/or manipulation (WM) of information used to complete both oral and written expression tasks. Additionally, further investigation is warranted among children with ADHD and comorbid Specific Learning Disability in written expression, and children with disorders where WM performance deficits are suspected – e.g., depression (Harvey et al., 2004), anxiety (Tannock, Ickowicz, & Schachar, 1995), and a wide range of developmental disabilities. Examining an older age range of youths may also prove valuable given their expected ability to invoke WM CE resources as PH STM becomes more automatized. Within this context, separable CE components (e.g., interference control, updating, dual processing, and sequencing) should be scrutinized to determine the extent to which they may be underdeveloped in youth with ADHD and other disorders that involve WM deficits and contribute to written expression difficulties.

We also acknowledge that mediation analysis assumes a temporal ordering of the mediating and dependent constructs and that causality cannot be substantiated. The temporal sequencing adopted in the current study, however, was based on extant research and theoretical accounts that provide compelling evidence regarding the developmental sequence of PH STM, oral expression, and written expression in humans. Despite this potential limitation, an important contribution of including multiple variables in mediational models is to elucidate the possible mechanisms and/or processes that may explain the established relation between diagnostic status and particular outcomes prior to initiating longitudinal research to confirm causal relations. Nevertheless, longitudinal studies are necessary in the future to confirm the findings reported in the present investigation.

Because written expression abilities as operationalized in the present paper represent an amalgamation of various skills necessary to produce coherent written work (e.g., capitalization, punctuation, syntax, grammar), follow-up studies are necessary to determine whether the mediational influences of ADHD-related deficits are consistent across domain-specific written expression components. Although previous studies investigated the multi-component nature of writing abilities in older children with ADHD (Casas,

Ferrer, & Fortea, 2013, Molitor, Langberg, & Evans, 2016), such analysis was not possible in the current investigation due to insufficient power and concerns of increased family-wise error as a result of multiple tests. A recent meta-analytic review (Graham et al., 2016), however, found that children with ADHD perform equally poorly across all elements of writing (e.g., handwriting, syntax, grammar, punctuation, & task), and suggests that the neurocognitive abilities underlying the various writing skills may not differ.

Finally, complimentary functional neuroimaging studies are warranted to examine the extent to which overlapping patterns of activation (e.g., dorsolateral prefrontal cortex, angular gyrus, frontal premotor cortex) during WM and written expression tasks identified in children with Specific Learning Disorder in written expression and in neurotypical samples (Richards, Nagy, Abbott, & Berninger, 2016) are consistent with those identified in children with ADHD. Identifying neural networks specific to PH STM and written expression deficits may be used to inform the design of personalized interventions consistent with the NIMH Research Domain Criteria (RDoC) initiative (Insel, 2014).

The discovery of PH STM and oral expression as significant contributors to written expression difficulties among boys diagnosed with ADHD has potentially promising clinical implications. For example, extant interventions tend to focus on teaching writing processes that place a large demand on CE abilities (Lienemann and Reid, 2008), whereas the results reported herein suggest that writing interventions for boys with ADHD may benefit by incorporating training exercises that strengthen PH STM capacity and improve students' ability to retrieve, hold, and express writing rules related to syntax, grammar, and word forms. Doing so would likely increase the automaticity by which these processes are deployed, allowing children to utilize upper-level cognitive resources required for advanced level writing assignments.

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The authors declare that they have no conflicts of interest.

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