

Reduced graphomotor procedural learning in children and adolescents with ADHD

Thomas A. Duda*, Joseph E. Casey, Amanda M. O'Brien, Natalie Frost, Amanda M. Phillips

University of Windsor, 401 Sunset Avenue, Windsor, Ontario N9B 3P4, Canada

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ABSTRACT

Purpose: The present study sought to determine if children and adolescents with ADHD demonstrate reduced procedural learning of a graphomotor program. **Method:** Thirty-two children and adolescents between age 9 and 15 with ($n = 16$) and without ADHD ($n = 16$) participated in the study. Each group of participants practiced a novel grapheme on a digitizing tablet 30 times. Participants with ADHD were off stimulant medication or were medication naïve. **Results:** Control participants demonstrated significant improvement in graphomotor fluency from the beginning to the end of practice, $T = 2$, $z = -2.534$, $p = .009$, whereas participants with ADHD did not, $T = 4$, $z = -1.810$, $p = .074$. **Conclusions:** Consistent with findings in adults with ADHD, results indicate that graphomotor procedural learning in children and adolescents with ADHD is attenuated. Findings have implications for future research that may inform remediation of handwriting difficulties, academic accommodations, and using digitizing technology for neuropsychological assessment.

1. Introduction

Attention-Deficit/Hyperactivity Disorder (ADHD) is a neurodevelopmental disorder defined diagnostically by behavioral symptoms associated with inattention and/or hyperactivity and impulsivity (American Psychiatric Association, 2013). Prevalence estimates vary between countries, but ADHD has been estimated to affect between 3.4% and 5% of children worldwide (Polanczyk, Salum, Sugaya, Caye, & Rohde, 2015). In addition, ADHD is often not diagnosed alone, and is highly comorbid with other neurodevelopmental disorders, including learning disabilities (LD) and developmental coordination disorder (DCD) (Kadesjo & Gillberg, 2001; Mayes & Calhoun, 2006; Piek, Pitcher, & Hay, 1999).

The etiology of ADHD and its symptomatic expression appear multifactorial in nature, involving an interplay between genetics, environmental factors, and neurological correlates (Barkley, 2006; Koziol, Budding, & Chidekel, 2013; McLoughlin, Palmer, Rijdsdijk, & Makeig, 2014; Merwood et al., 2014; J. Wu, Xiao, Sun, Zou, & Zhu, 2012; Wu, Gau, Lo, & Tseng, 2014). Across children and adults with ADHD, the most replicated, between-group differences found within the brain's gray matter indicate volumetric reductions of the dorsolateral prefrontal cortex, anterior cingulate, basal ganglia, and cerebellum (Amico, Stauber, Koutsouleris, & Frodl, 2011; Batty et al., 2010; Castellanos et al., 2002; Durston et al., 2004; McAlonan et al., 2007; Narr et al., 2009). Additionally, a recent meta-analysis reported differences in white matter structural integrity affecting the right anterior corona radiata, corpus callosum, bilateral internal capsules, and left cerebellum (van Ewijk, Heslenfeld, Zwiers, Buitelaar, & Oosterlaan, 2012). When viewed longitudinally,

* Corresponding author at: 401 Sunset Avenue, Windsor, Ontario N9B 3P4, Canada.
E-mail address: dudat@uwindsor.ca (T.A. Duda).

neuroimaging studies provide evidence for delays in the developmental trajectory of cortical thickness across the cerebrum, as well as developmental differences in subcortical regions and neural networks (Sato, Hoexter, Castellanos, & Rohde, 2012; Shaw et al., 2006, 2007, 2012, 2013, 2014; Tomasi & Volkow, 2014). Functional neuroimaging studies generally support structural findings and appear to implicate areas of the brain believed to be involved in attention (dorsolateral prefrontal cortex, parietal lobe, and cerebellum), inhibition (inferior frontal cortex, supplementary motor area, and anterior cingulate), and timing (left inferior prefrontal cortex, insula, cerebellum, and left inferior parietal lobe) (Hart, Radua, Mataix-Cols, & Rubia, 2012; Hart, Radua, Nakao, Mataix-Cols, & Rubia, 2013).

Although research using neuropsychological assessment has yet to identify a neurocognitive profile sufficiently sensitive and specific to ADHD as currently defined using behavioral criteria (Doyle, Biederman, Seidman, Weber, & Faraone, 2000; Nigg, Willcutt, Doyle, & Sonuga-Barke, 2005; Wasserman & Wasserman, 2012), non-diagnostic features commonly found in those with ADHD include motor skill deficits (Brossard-Racine, Majnemer, & Shevell, 2011; Cole, Mostofsky, Larson, Denckla, & Mahone, 2008; Dyck & Piek, 2014) and difficulties automatizing new skills (Ackerman, Anhalt, Holcomb, & Dykman, 1986; Adi-Japha, Fox, & Karni, 2011; Barnes, Howard, Howard, Kenealy, & Vaidya, 2010; Karatekin, White, & Bingham, 2009; Koziol et al., 2013). Germane to the current topic are the handwriting problems commonly found in children with ADHD.

1.1. Graphomotor functioning in childhood ADHD

Research on graphomotor functioning in children with ADHD has often found that their handwriting can be described as frequently illegible, error-prone, and less organized than the handwriting of children without ADHD, which in turn affects academic achievement (Barkley, 2006; Brossard-Racine, Majnemer, Shevell, Snider, & Belanger, 2011; Brossard-Racine, Majnemer, Shevell, & Snider, 2008). Investigations using digitizing technology (e.g., computer tablets) and kinematic analysis of handwriting movements (i.e., the analysis of movements using variables of time, acceleration, speed, and their derivatives) in those with ADHD has provided an additional method of inquiry using objective and quantifiable markers (Viviani & Terzuolo, 1982). This type of analysis is particularly relevant to graphomotor research in those with ADHD when considering that poor handwriting in this population does not appear to be related to purely visual-perceptual or linguistic difficulties, but instead is suspected to be associated with basic parameter setting (e.g., regulation of force, speed, and size of movements), motor control, and timing of movements (Adi-Japha et al., 2007; Brossard-Racine et al., 2008; Marcotte & Stern, 1997; Rommelse et al., 2008; Schoemaker, Ketelaars, van Zonneveld, Minderaa, & Mulder, 2005).

Kinematic analyses of handwriting in those with ADHD has provided further insight into the motor control differences within this population that cannot be studied using other methods. For example, children with ADHD without a history of developmental coordination disorder have demonstrated significantly greater variability in stroke size compared to typically-developing peers (Chang & Yu, 2009). Other kinematic variables (e.g., ballisticity, which is the degree to which movements can be characterized as sudden) have been shown to correlate with ADHD diagnostic symptomatology such as impulsivity (Frings et al., 2010; Langmaid, Papadopoulos, Johnson, Phillips, & Rinehart, 2014). Whereas treatment with stimulant medication appears to result in improved handwriting performance based on qualitative analysis (Brossard-Racine et al., 2015; Lerer, Artner, & Lerer, 1979; Whalen, Henker, & Finck, 1981), studies using kinematic analyses have also found that children with ADHD (but not adults) produce more dysfluent and less automatized handwriting when being treated with stimulant medication relative to both controls and children with ADHD who have discontinued medication treatment, with an effect that appears unrelated to medication side-effects (Flapper, Houwen, & Schoemaker, 2006; Tucha & Lange, 2001, 2004, 2005). One kinematic study of adults with ADHD who were counterbalanced on and off stimulant medication identified greater between-trial intraindividual variability in graphomotor program execution regardless of medication status, but this effect was only evident when learning a novel grapheme (Duda, Casey, & McNeven, 2014). This was interpreted as indicating potential differences in procedural learning of a graphomotor program in adults with ADHD who had discontinued stimulant medication treatment.

1.2. Procedural learning in ADHD

Consistent with this interpretation, some researchers suggest that children and adults with ADHD may show delays in procedural learning and the automatization of skills – such as handwriting – that require effortful attentional control and practice over time (Lange et al., 2007). Evidence exists to support this position. In one study, researchers used a serial motor sequence learning task to study implicit learning in children with ADHD (Barnes et al., 2010). Results indicated that children with ADHD demonstrated a variable rate of learning relative to controls when completing motor sequences (i.e., they showed a reduced priming effect) that was not attributable to poor perceptual-motor abilities.

In another study, researchers used two motor learning tasks to study procedural learning in children with ADHD and comorbid reading disability and developmental coordination disorder (Magallon, Crespo-Eguilaz, & Narbona, 2015), conditions that have high rates of co-occurrence with ADHD (Brossard-Racine et al., 2011; Westendorp, Hartman, Houwen, Smith, & Visscher, 2011). Results demonstrated improvements in procedural learning after three practice trials on the motor learning tasks for this group, yet all participants were on methylphenidate medication at the time of the study. This stimulant medication is known to improve motor skills in children with ADHD and developmental coordination disorder (Bart, Daniel, Dan, & Bar-Haim, 2013; Brossard-Racine, Shevell, Snider, Belanger, & Majnemer, 2012). As such, the improvements in procedural motor learning may have been mediated by effects of stimulant medication in these children.

Similar procedural learning differences have also been identified in adults with ADHD. Researchers investigating procedural

learning in young women with ADHD found that control participants demonstrated significant improvement in speed and accuracy, whereas participants with ADHD only showed an improvement in speed and demonstrated significantly less accurate performance at 24-hour and 2-week post-training follow-up assessments (Adi-Japha et al., 2011). Evidence of attenuated procedural learning specific to graphomotor functioning was also recently demonstrated in adults with ADHD (Duda, Casey, & McNevin, 2015). In this experiment, the graphomotor fluency of control participants and participants with ADHD was compared at the beginning and end of practicing a novel grapheme, fluency being defined as the smoothness and automaticity with which a graphomotor program was executed. Despite equivalent practice, control participants demonstrated a statistically significant improvement in graphomotor fluency, whereas participants with ADHD who had discontinued stimulant medication did not. These findings in turn provided additional evidence that graphomotor procedural learning may develop differently in those with ADHD relative to typically developing peers. Indeed, these findings also suggest that individuals with ADHD who are off medication do not demonstrate similar improvements in graphomotor fluency as do controls.

1.3. Rationale and hypothesis

Although studies have investigated the development of graphomotor fluency, automaticity, and procedural learning with the use of kinematic analysis in typically developing individuals, in children with other neurodevelopmental disorders (e.g., developmental coordination disorder), and in adults with ADHD (Chang & Yu, 2009; Duda et al., 2015; Portier & Van Galen, 1992), still lacking in the literature are studies investigating this development within the child and adolescent ADHD population, particularly when off medication. Further, investigation into graphomotor procedural learning in ADHD is especially relevant in these age groups given the negative effects handwriting problems can have on academic achievement, as well as the potential justification for academic accommodations should differences be present. As such, the current study sought to investigate the development of graphomotor fluency in children and adolescents with and without a history of ADHD as they practiced a novel grapheme using kinematic analysis. Based on previous research, a significant group by practice interaction was predicted in which children and adolescents with ADHD would demonstrate reduced ability to automatize a newly learned grapheme relative to their peers without ADHD despite equivalent practice.

2. Method

2.1. Participants

Power analysis was conducted using G*Power software (Buchner, Erdfelder, Faul, & Lang, 2009), which indicated that 32 total participants were needed for adequate power ($1 - \beta = 0.80$) to detect a statistically significant interaction effect ($\alpha = 0.05$) of approximately medium effect size ($r = 0.26$). Following clearance by the University's Research Ethics Board, 46 total child and adolescent participants between the age of 9 and 15 took part in the study. Participants included only those with normal or corrected to normal vision, and those without neurological or cognitive deficits that would significantly affect their ability to complete tasks or reasonably influence outcomes (e.g., cerebral palsy affecting the upper extremities or intellectual disability). Data from one potential control participant were removed due to observably poor cooperation, and data from three potential control participants were removed due to suboptimal performance based on calculation of a Reliable Digit Span score of ≤ 6 (Kirkwood, Hargrave, & Kirk, 2011). Data from an additional 10 potential control participants were removed to create equal group sizes and maximize the robustness of statistical analyses and interpretation. Rather than randomly removing participants, participant removal was conducted such that control and ADHD participants were matched as closely as possible on age, estimated general intellectual functioning, and socioeconomic status (SES). Altogether, data were analyzed from 32 child and adolescent participants (ADHD $n = 16$, Control $n = 16$). See Table 1 below for participant descriptive statistics for sex, handedness, race/ethnicity, age, estimated intelligence, and SES. Additional data were gathered regarding potential diagnostic comorbidities, medication use, psychosocial functioning, and academic and fine motor skill skills, which can be found in Tables 2 and 3. See the Materials and Apparatus and Procedures sections below for further details regarding measures selected and how they were used.

2.1.1. Group assignment and potential comorbid diagnoses

Participants assigned to the ADHD group included those who 1) were previously diagnosed with ADHD by a health care professional and continued to meet DSM-5 criteria, 2) did not have a pre-existing diagnosis of ADHD but met DSM-5 criteria, or 3) had a Conners 3 ADHD Index Probability Score ≥ 64 , which indicates a high probability that a classification of ADHD is warranted. Classification of ADHD presentation as primarily inattentive, primarily hyperactive-impulsive, or combined was based on DSM-5 symptom criteria for those who met full criteria. Participants assigned to the ADHD group based on the Conners 3 ADHD Index Classification were classified as Specified ADHD (see below for further details pertaining to this group). All participants not meeting any of these criteria were assigned to the control group.

Of the 16 participants with ADHD, only three did not meet strict DSM-5 diagnostic criteria for primarily inattentive, primarily hyperactive-impulsive, or combined type, but met inclusion criteria based on the Conners 3 ADHD Index. This was the result of one participant having impairments reported in only one setting and two participants demonstrating five rather than the required six symptoms in the domains of inattention and hyperactivity-impulsivity. Otherwise, all other DSM-5 criteria were met by these participants, and difficulties in psychosocial functioning and ADHD-related symptomatology were highly similar to participants who met full DSM-5 criteria. That is, each of these three participants had Conners 3 Inattention and/or Hyperactivity/Impulsivity ratings in the

Table 1
Descriptive Statistics of Participant Sample.

		Control		ADHD			
		<i>n</i>	<i>M</i>	<i>SD</i>	<i>n</i>	<i>M</i>	<i>SD</i>
Sex	Female	8			5		
	Male	8			11		
Handedness	Right	13			14		
	Left	3			2		
Race/Ethnicity	Asian	0			0		
	Black/African/Caribbean	1			2		
	Caucasian/European/White	9			12		
	Hispanic/Latina/Latino	0			0		
	Multiracial	1			0		
	Native/Aboriginal	4			0		
	Other	1			1		
Age			11.89	2.03		11.24	1.86
Estimated FSQ			104.69	9.71		104.31	15.78
	Block Design Subtest		11.06	1.98		10.81	3.73
	Vocabulary Subtest		10.56	2.48		10.69	2.47
SES			40.41	14.02		40.31	11.15

Note. Estimated FSQ = estimate of general intellectual functioning derived from Block Design and Vocabulary Subtests of the Wechsler Intelligence Scale for Children, 4th Edition (WISC-IV).

Table 2
Participant Diagnostic Information and Medication Use.

		Control <i>n</i>	ADHD <i>n</i>
ADHD Subtypes:			
	ADHD Combined	0	6
	ADHD Inattentive	0	1
	ADHD Hyperactive-Impulsive	0	6
	Specified ADHD	0	3
At-Risk Additional Diagnosis/Comorbidities [†] :			
	Anxiety Disorder	0	4
	Conduct Disorder	0	2
	Developmental Coordination Disorder	1	9
	Oppositional Defiant Disorder	0	6
	Mood Disorder	0	5
	Specific Learning Disability	3	12
	Speech/Language Disorder	1	1
	Tic Disorder	0	1
Prescribed Stimulant medication (Total):			
	Vyvanse	0	1
	Adderall	0	1
	Stimulant Medication Naïve	16	14

Note. [†]At-Risk Additional Diagnosis/Comorbidities were based on positive findings from screening measures or parent-report of an existing diagnosis. These are not, however, diagnostic by themselves, and represent increased risk for these comorbidities.

elevated range or greater (i.e., T scores ≥ 65), and at-risk or clinically significant difficulties in most psychosocial domains measured. Data collected by interview and rating scales that indicated potential diagnostic comorbidities were recorded as indicating the participant was “at-risk” for that potential comorbid diagnosis. See Table 4 regarding determination of “at-risk” designation for potential comorbid diagnoses.

2.2. Materials and Apparatus

2.2.1. Demographic, psychiatric, and neuropsychological assessment

Participant demographic, psychiatric, and medical information was collected via structured interview with parents and participants. Socioeconomic status was calculated using the four-factor method of social status (Hollingshead, 1975), with higher calculated scores based on caregiver education, occupation, and marital status indicating higher SES. Neuropsychological testing included assessment of the following domains: psychosocial functioning by parent-report using the Behavioral Assessment System for Children,

Table 3
Participant Psychosocial Functioning and Academic Performance.

		Controls		ADHD	
		<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
BASC-2	Externalizing	43.88*	6.13	59.38	8.55
	Internalizing	41.81*	7.33	60.38 [†]	13.55
	Behavioral Symptom Index	42.19*	6.10	64.06 [†]	8.24
	Adaptive Skills	58.63*	6.94	45.06	9.03
	Hyperactivity	43.06*	6.02	64.88 [†]	10.32
	Aggression	43.88*	5.54	55.75	11.29
	Conduct Problems	46.50*	6.83	54.69	8.57
	Anxiety	44.06*	10.55	59.31	14.16
	Depression	44.06*	7.22	62.25 [†]	11.73
	Somatization	42.19*	5.93	56.69	12.32
	Atypicality	42.56*	4.45	60.56 [†]	12.40
	Withdrawal	45.19*	13.11	60.06 [†]	19.70
	Attention Problems	45.00*	7.62	62.00 [†]	6.57
	Adaptability	57.00*	7.70	44.94	8.81
	Social Skills	56.00	7.66	51.56	11.47
Leadership	60.00*	7.99	48.81	11.85	
Activities of Daily Living	56.31*	7.32	40.94	7.77	
Communication	57.44*	6.28	42.75	11.52	
Conners 3	Inattention	48.31*	8.34	73.31 [‡]	10.21
	Hyperactivity-Impulsivity	47.44*	6.49	76.81 [‡]	14.25
	Learning Problems	47.13*	7.98	67.69 [†]	14.40
	Executive Function	47.56*	8.27	66.06 [†]	11.33
	Defiance/Aggression	47.25*	5.79	60.94	16.29
	Peer Relations	48.81*	12.64	66.31 [†]	19.07
	DSM-5 ADHD Inattentive	48.44*	7.60	72.00 [‡]	9.77
	DSM-5 ADHD Hyperactive-Impulsive	47.94*	7.34	75.25 [‡]	15.55
	DSM-5 Conduct Disorder	46.69*	4.25	53.69	11.11
DSM-5 Oppositional Defiant Disorder	48.25*	6.31	66.19 [†]	15.49	
DCDQ'07	Control During Movement	27.88*	4.43	22.56	6.95
	Fine Motor Skills	19.50*	3.72	12.50	5.09
	General Coordination	21.75*	4.01	16.69	5.28
	Total	67.75*	9.90	51.75	15.24
WRAT 4	Word Reading	105.19	12.48	102.69	15.92
	Spelling	107.63	16.97	100.50	17.10
	Math Calculation	99.63*	16.07	89.75	11.42
Grooved Pegs	Dominant	45.37	9.18	42.18	8.68
	Non-Dominant	46.45	9.71	41.26	9.81

Note. Statistically significant difference between controls and participants with ADHD, * $p < .05$. [†] = At-Risk or Elevated problems. [‡] = Clinically Significant or Very Elevated problems.

Table 4
Criteria for Indicating “At-Risk” Potential Comorbid Disorders.

Diagnosis	Criteria: Parent-report of diagnosis and/or...
Anxiety Disorder	BASC-2 Anxiety T Score ≥ 70
Conduct Disorder	Conners 3 Symptom Count criteria met
Developmental Coordination Disorder	Positive Screen based on DCDQ'07 age norms
Learning Disability	Conners 3 Learning Problems T score ≥ 65
Mood Disorder	BASC-2 Depression T Score ≥ 70
Oppositional Defiant Disorder	Conners 3 Symptom Count criteria met

2nd Edition (BASC-2) (Reynolds & Kamphaus, 2004); ADHD and disruptive behavior symptomatology by parent-report based on the Conners 3 parent rating scale with DSM-5 update (Conners, 2008); developmental coordination disorder (DCD) symptomatology by parent-report using the Developmental Coordination Disorder Questionnaire 2007 (DCDQ'07); general intellectual functioning (i.e., FSIQ) derived from Block Design (BD) and Vocabulary (VC) subtest performance on the Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV) (Wechsler, 2003) as suggested by Sattler (2008); a performance validity measure based on Reliable Digit Span performance calculated using the Digit Span subtest of the WISC-IV; academic skills indicated by performance on the Wide

Range Achievement Test-Fourth Edition (WRAT 4) (Wilkinson & Robertson, 2006); and fine motor skills indicated by performance on the Grooved Pegboard test.

2.2.2. Handwriting software

A WACOM Cintiq 21UX digitizing tablet and MovAlyzeR software (NeuroScript, 2009, LLC; Tempe, AZ, USA) were used to record handwriting performance and analyze kinematic data, respectively. The digitizing tablet provided real-time visual feedback using a special non-inking pen as participants performed tasks. A maximum sampling rate of 200 Hz was used, x-y coordinates were low-pass filtered at 12 Hz, and movements were broken down into strokes using interpolated vertical velocity zero crossings. The digitizing tablet was calibrated and accuracy maximized according to MovAlyzeR software protocol.

2.3. Procedures

After study eligibility was determined, participants taking stimulant medication were asked to discontinue its use 24–48 h prior to testing (time-frame based on drug information indicating very low drug plasma concentrations after this period; see U.S. Food & Administration, 2007). Guardian consent and child assent were obtained, and all procedures were completed in one session at a university research center. Parents and participants were both present during the structured interview, which was followed by parents completing rating forms and participants completing neuropsychological assessment and experimental tasks with a separate examiner. Researchers confirmed all forms were complete, followed-up on critical items and took action as needed, and answered parent questions to ensure understanding.

The order of neuropsychological assessment and the experimental handwriting task was counterbalanced. Prior to engaging in the experimental handwriting task, participants were provided the opportunity to become familiar with the tablet and pen, and adjust their chair and the tablet to a comfortable position. Practice consisted of writing their name on the digitizing tablet five times and following an on-screen cursor in various patterns six times. Additional instruction and corrections were provided to ensure participants held the pen and wrote as naturally as possible.

2.3.1. Novel grapheme copy task

The experimental grapheme copy task, labelled as the Learning task, consisted of copying a novel grapheme from the participant's left to right 30 times within a box displayed on the digitizing tablet. A card with the novel grapheme was present throughout the experiment and the participant was allowed to move it at the beginning to a location that was best seen. Participants were instructed to recreate the design as accurately as they could and make it about the same size. They were also told that the design did not have to be “perfect” and to “just do their best” if performance concerns were expressed or observed. See Fig. 1 below for a sample tracing of the novel grapheme that was drawn, as well as sections indicating each element of the grapheme (see below).

The dependent variable, normalized jerk, (NJ) was calculated for each individual trial (described in further detail below). To analyze change with practice, comparison was made between the beginning of practice and the ending of practice. Beginning of practice was operationalized as the mean NJ of the first three valid trials, whereas the ending of practice was defined as the mean NJ of the last three valid trials.

Trials were deemed as valid by observation if they were accurately representative of the novel grapheme, which consisted of the six elements pictured: first wavy stroke including pointed stroke, lower loop, large upper loop, small loop, lower section of large ‘S’, upper section of ‘S’ stroke. Trials deemed invalid by observations were subsequently removed from analysis. After observationally invalid trials were removed, extreme values of NJ for any given trial for each specific participant were removed. Values were deemed

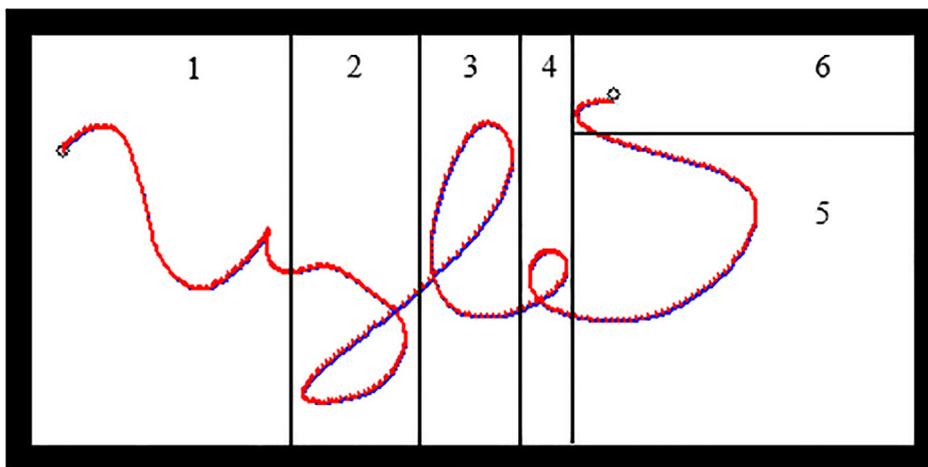


Fig. 1. Example image capture of novel grapheme with box copied during the Learning task. Numbers represent the six elements of the grapheme that were required to be drawn by each participant.

extreme when performance reflected an intraindividual Z score (calculated with removal) of ≥ 5 . These criteria resulted in the removal of approximately 23% of all trials (224 of 960). Of these, 211 trials were deemed invalid observationally and 13 trials were deemed extreme. Due to significant negative correlations found between age and general intellectual functioning and the dependent variable of graphomotor fluency performance (i.e., NJ), invalid trials were deemed to be not missing at random and imputation of these missing values was therefore not used.

An ANOVA was performed to determine if there was a significant difference in the number of invalid trials produced between the ADHD and control groups. There was, however, no statistically significant difference between the number of invalid trials produced by control ($M = 5.50$, $SD = 4.98$) versus ADHD ($M = 8.50$, $SD = 6.78$) participants, $F(1, 31) = 2.03$, $p = .164$, $\omega^2 = 0.03$.

2.3.2. Novel grapheme analysis

The dependent variable of interest derived using MovAlyzeR software was Normalized Jerk (NJ), which represented the operational definition of the degree of graphomotor fluency, automaticity, and procedural learning. NJ is a continuous measure of writing smoothness and fluency that is similar to the measure “number of inversions of acceleration/velocity” frequently found in the ADHD literature (Flapper et al., 2006; Schoemaker, Ketelaars, van Zonneveld, Minderaa, & Mulder, 2005; Tucha & Lange, 2001, 2004; Tucha, Paul, & Lange, 2003), but provides the advantage of comparing patterns of varying duration and size because data are normalized (Teulings, Contreras-Vidal, Stelmach, & Adler, 1997). Higher values of NJ indicate more dysfluent, less automatized, and less well-learned movement (Teulings et al., 1997; Yan, Rountree, Massman, Doody, & Li, 2008). With practice, graphomotor fluency and automatization improve as indicated by lower values of NJ (Portier & Van Galen, 1992; van Mier, Hulstijn, & Petersen, 1993) resulting from fewer “stops and starts” during movement production and improved sensorimotor coherence.

Due to the experimental nature of this measure and variability resulting from different data-capturing tools (e.g., monitor-style digitizing tablets versus hand-held digitizing tablets, visual feedback with trace using non-inking pens versus use of inking pens or pens with no visual feedback) and individual computer processing differences, there currently exists no systematically determined reliability or validity data for this specific tool. However, dozens of studies with various clinical populations (e.g., ADHD, Parkinson's Disease, DCD, and other disorders) have yielded replicable results using kinematic analysis (for examples, see Chang & Yu, 2009; Gangadhar et al., 2009; Schoemaker et al., 2005; Smits-Engelsman, Wilson, Westenberg, & Duysens, 2003; Tucha & Lange, 2001, 2004; Tucha et al., 2006), and normative datasets of kinematic data are beginning to be constructed (Accardo, Genna, & Borean, 2013).

3. Results

All statistical analyses were performed using IBM SPSS Statistics, Version 22. Higher values of mean NJ represented greater dysfluency (i.e., less automaticity) and lower values represented less dysfluency (i.e., greater automaticity). Due to significant non-normality and heterogeneity of variance found within the data combined with small sample size, parametric statistics were deemed inappropriate and nonparametric statistics were thus used for analyses. Between-group comparisons were completed using the Mann-Whitney U test whereas within-task analysis was completed using the Wilcoxon Signed-Rank test. Effect sizes were estimated using r , such that effect sizes of 0.1, 0.3, and 0.5 were interpreted as indicating a small, medium, and large effect size, respectively. A Bonferroni adjusted alpha level of 0.017 was used due to multiple comparisons. All assumptions of the Mann-Whitney U and Wilcoxon Signed-Rank tests were assessed and met (i.e., at least ordinal level dependent variables, a dichotomous independent variable, independence of observations, similarly shaped distributions between groups, and, as unique to the Wilcoxon Signed-Rank test, difference scores that were symmetrical in distribution in each group).

A statistically significant main effect was observed for practice in which the graphomotor fluency and automatization of participants, when grouped together, improved from the beginning ($Mdn = 525.66$) to the ending ($Mdn = 313.11$) of practice, $T = 6$, $z = -3.011$, $p = .002$. The effect size was large, $r = -0.532$. However, as an individual group, participants with ADHD did not demonstrate a statistically significant improvement in graphomotor fluency from the beginning ($Mdn = 565.10$) to the ending ($Mdn = 362.79$) of practice, $T = 4$, $z = -1.810$, $p = .074$, $r = -0.32$. In contrast, control participants did show significant improvement in graphomotor fluency and automatization (beginning NJ: $Mdn = 428.49$; ending NJ: $Mdn = 248.76$), $T = 2$, $z = -2.534$, $p = .009$, and the effect size was medium to large $r = -0.448$. In sum, 2 more control participants evidenced a significant improvement in graphomotor fluency with practice than participants with ADHD.

The statistically significant main effect for practice combined with data indicating only control participants significantly improving from the beginning to the ending of practice suggested an interaction effect, but this interpretation was qualified with further nonparametric analyses. Results from follow-up analyses using the Mann-Whitney U test indicated no statistically significant between-group differences when comparing controls with ADHD participants at the beginning, $U = 104.00$, $z = -0.905$, $p = .381$, $r = -0.160$, or the ending of practice, $U = 100.00$, $z = -1.055$, $p = .305$, $r = -0.187$. When reviewing mean rankings that were largely equivalent between groups and within conditions, as well as negative and positive ranks with similar directionality, results did not appear to support an appreciable group by practice interaction effect. See Table 5 for additional summary information regarding nonparametric analyses.

4. Discussion

Using kinematic analysis, the present study sought to determine if child and adolescent participants with ADHD would demonstrate similar ability to learn a novel graphomotor program given the same amount of practice as those without the disorder.

Table 5
Summary of Nonparametric Test Results.

	<i>n</i>	<i>Mean</i> NJ	<i>SD</i> NJ	Mann-Whitney U Test		Wilcoxon Signed-Rank Test		
				Mean Rank	Sum of Ranks	Negative Ranks	Positive Ranks	Ties
Beginning								
Controls	16	535.16	322.93	15.00	240.00			
ADHD	16	834.83	840.58	18.00	288.00			
Ending								
Controls	16	406.47	400.20	14.75	236.00			
ADHD	16	590.79	593.84	18.25	292.00			
Beginning – Ending								
Main Effect						26	6	0
Controls						14	2	0
ADHD						12	4	0

Note. NJ = normalized jerk, with lower values indicating more fluent and automatized graphomotor program execution.

Control participants were expected to demonstrate a statistically significant improvement in graphomotor fluency and automaticity (i.e., lower NJ at the ending relative to the beginning of practice), whereas ADHD participants who had discontinued stimulant medication or were medication naïve were hypothesized to show relatively reduced improvement. Results of the current study were consistent with previous research (Duda et al., 2015) and hypothesized outcomes: control participants demonstrated a statistically significant improvement in graphomotor fluency and automaticity whereas participants with ADHD did not. These data were interpreted to suggest that children and adolescents with ADHD not taking stimulant medication at time of study may exhibit reduced procedural learning while practicing a new grapheme despite similar practice as peers without the disorder.

These results are in contrast to previous findings with children who have comorbid ADHD and DCD and are prescribed methylphenidate medication (Magallon et al., 2015). Researchers demonstrated that children with ADHD, RD, and DCD perform significantly better on motor learning tasks after three practice trials albeit to a lesser degree than controls. This study differs from the current project in one major way: the current participants were mostly medication naïve or had discontinued medication for 24–48 h before participating ($n = 2$). When children were off medication, it appears those with ADHD exhibit reduced procedural learning compared to control participants despite similar practice of 30 trials. These differences echo existing research suggesting that methylphenidate medications improve motor functioning in children with ADHD and DCD (Bart et al., 2013). Indeed, similar discrepancies in procedural learning of the same novel grapheme have been shown in adults with ADHD who were on and off stimulant medication (Duda et al., 2015). Researchers found that control participants and participants with ADHD on medication showed improvements in graphomotor fluency after 30 practice trials whereas participants with ADHD off medication did not despite equivalent practice. Findings suggest that graphomotor procedural learning in adults with ADHD may occur more slowly than typically developing peers (Duda et al., 2015). Together with findings from the present study, it appears that attenuated graphomotor procedural learning occurs in children, adolescents, and adults with ADHD.

Kinematic analysis of graphomotor fluency conducted with digitizing technology, as was utilized in the current study, possesses the advantage of analyzing motor skill acquisition and procedural learning with the use of an objective and quantifiable measure. A caveat here is that this tool lacks systematically determined reliability and validity data at this time, limiting its direct applicability in neuropsychological assessment at this time. In addition, data collected from these analyses were non-normal and create some interpretive difficulty. For example, as seen in Table 5, children with ADHD evidenced a greater mean change in NJ with practice, which could suggest greater improvement within this group if only mean values were considered. It is important to again point out that failure to meet the assumptions of parametric analyses led to the necessity of non-parametric analyses using medians rather than means, which was appropriate given the data and improved interpretability. Beyond this point, however, it may not be entirely unexpected for children and adolescents with ADHD to demonstrate higher NJ values at baseline (and thus greater dysfluency) given the above described history of handwriting difficulties in ADHD. Indeed, ADHD is a neurodevelopmental disorder affecting motor skill development and acquisition, and there is evidence that 1) poor writers show greater dysfluency compared to typically developing peers at the beginning of tasks and 2) children with better graphomotor functioning have less improvement to make with practice or intervention and can thus show less improvement on average (Hurschler, Wicki, & Falmann, 2018; Overvelde & Hulstijn, 2010). Nevertheless, methods used in the current study do possess ecological validity when considering that graphomotor fluency – and in turn handwriting – is an important aspect of academic performance and children are actively involved in automatizing their handwriting. In addition, automated handwriting is often a required basal skill for future academic endeavors (e.g., being able to take notes while simultaneously paying attention to teacher instructions).

Small sample sizes inherently raise concern for sampling bias. This is a consideration in the current study given that only two more control participants showed improvement in graphomotor fluency than did participants with ADHD. Despite a small sample size, adequate statistical power and interpretability of data were achieved through nonparametric analyses. In future studies, larger sample sizes may enable the use of more powerful statistical analyses. The small sample also consisted of a relatively broad age range of participants (i.e., pre-pubertal children through adolescents), which also clouds interpretation from a developmental perspective. This became evident when considering the complexity of the grapheme used to examine procedural graphomotor learning. As

mentioned previously, child and adolescent participants from both groups combined to produce a large proportion of invalid trials. This suggests that the novel grapheme was too complex for many participants, particularly younger participants. Participant age within the context of sex must also be taken into account given findings of motor learning differences between males and females that appear to increase between childhood and adolescence (Dorfberger, Adi-Japha, & Karni, 2009). Together, future research would benefit from analyses stratified by narrow age-bands, sex, and the use of an array of designs with differing levels of complexity to better control for and understand maturational factors involved in procedural graphomotor learning.

Although common in the graphomotor fluency literature, the generalizability of these findings is also limited by the small number of participants with each presentation of ADHD (i.e., primarily inattentive, hyperactive-impulsive, combined presentation, and specified ADHD) and combining them for analyses. Indeed, there is a great need within this literature to improve upon our understanding regarding the effects of ADHD-subtype on graphomotor fluency and development. In addition, 3 ADHD group participants in the current study – although very similar to the other 13 participants in this group – met ADHD group inclusion criteria based on a combination of subthreshold DSM-5 symptomatology and the Conners ADHD index. There was also concern for greater diagnostic comorbidity than might be expected in a typical community sample. It must again be highlighted, however, that comorbidities as described in this study for both control and clinical groups are conceptualized as “At-Risk” rather than true diagnostic comorbidity. Indeed, these screening metrics were used to gain information about *risk* for comorbidity but are insufficient by themselves to make a formal diagnosis because they are sensitive to symptoms/problems but not specific. Importantly regarding potential diagnostic comorbidity are limitations associated with the degree to which reduced automatization can be attributed to ADHD itself versus the presence of a potentially comorbid condition. This is especially the case within the context of potential comorbid DCD, of which 63% of participants screened positive. However, it is also of note that of the four participants with ADHD who did not demonstrate an improvement in graphomotor fluency, two screened positive for DCD whereas two did not. These results are also in contrast to research showing improved procedural learning in children with DCD + ADHD + RD who are medicated with methylphenidate (Magallon et al., 2015). Noting these observations and combined with other research demonstrating difficulties with procedural motor learning and increased neuromotor noise in children with a diagnosis of DCD (Chang & Yu, 2009; Huau, Velay, & Jover, 2015), future research must investigate which effects are related to ADHD alone or in combination with formally-diagnosed DCD or other disorders.

5. Conclusions

In sum, results of the current study provide support that differences in graphomotor procedural learning exist between those diagnosed with ADHD and those without the disorder. That is, whereas control participants demonstrated significantly improved graphomotor fluency and automaticity across learning trials, those with ADHD did not demonstrate equivalent improvement. The results were interpreted to suggest that children and adolescents with ADHD not taking stimulant medication may exhibit reduced procedural learning while practicing a new grapheme despite similar practice as peers without the disorder. This finding expands upon the existing literature by indicating that reduced procedural learning is evident not only in adults (Duda et al., 2015), but also children and adolescents with ADHD. Given additional supporting research that addresses the limitations described above, findings may have clinical implications for 1) academic accommodations provided to children and adolescents whose performance appraisals depend upon handwriting (e.g., extended time to practice), 2) remedial interventions (e.g., additional time spent learning handwriting to improve automatization or how interventions can be tailored to address automatization; see Tucha & Lange, 2005), and 3) the use of kinematic analysis as an additional neuropsychological tool to assess motor learning difficulties in children and adolescents with ADHD or other neurodevelopmental disorders.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.humov.2018.06.018>.

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