Hyperactivity in Boys with Attention-Deficit/Hyperactivity Disorder (ADHD): A Ubiquitous Core Symptom or Manifestation of Working Memory Deficits?

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Abstract Hyperactivity is currently considered a core and ubiquitous feature of attention-deficit/hyperactivity disorder (ADHD); however, an alternative model challenges this premise and hypothesizes a functional relationship between working memory (WM) and activity level. The current study investigated whether children's activity level is functionally related to WM demands associated with the domain-general central executive and subsidiary storage/rehearsal components using tasks based on Baddeley's (*Working memory, thought, and action*. New York: Oxford University Press 2007) WM model. Activity level was objectively measured 16 times per second using wrist- and ankle-worn actigraphs while 23 boys between 8 and 12 years of age completed control tasks and visuospatial/phonological WM tasks of increasing memory demands. All children exhibited significantly higher activity rates under all WM relative to control conditions, and children with ADHD ($n=12$) moved significantly more than typically developing children ($n=11$) under all conditions. Activity level in all children was associated with central executive but not storage/rehearsal functioning, and higher activity rates exhibited by children with ADHD under control conditions were fully attenuated by removing variance directly related to central executive processes.

The significance of excessive motor activity or hyperactivity in the conceptualization of attention-deficit/hyperactivity disorder (ADHD) has varied considerably during the past century. Hyperactivity was initially considered the disorder's dominant feature in early clinical (Still, 1902) and theoretical (Chess 1960; Laufer et al. 1957) descriptions, and continued its primacy throughout the 1970s as reflected by the diagnostic monikers *hyperkinetic impulse-disorder* (Laufer et al. 1957), *hyperactive child syndrome* (Chess 1960), and *hyperkinetic reaction to childhood* (American Psychiatric Association 1968). Empirical validation of motor excesses in ADHD was extensively documented during this time and afterward using a broad range of methodologies and technologies, ranging from rating scales (Werry 1968), analogue measures (Barkley 1991) and direct observations (Whalen et al. 1978) to pedometers (Plomin and Foch 1981), stabilimetric cushions (Conners and Kronsberg 1985) and actigraphs (Porrino et al. 1983).

A paradigm shift occurred during the mid to late 1970s following Douglas's (Douglas 1972; Campbell et al. 1971) seminal work documenting attentional difficulties in children with ADHD relative to children with specific learning disabilities and typically developing children. The relegation of hyperactivity to a secondary role was consummated in the third edition of the *Diagnostic and Statistical Manual of Mental Disorders* nomenclature (American Psychiatric Association 1980)—excessive movement was no longer considered a necessary criterion for diagnosing the disorder—and, motor activity and impulsivity descriptors were grouped together based on factor analytic findings (DuPaul et al. 1998).

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Cognitive and neurocognitive accounts of ADHD flourished during the ensuing two decades and continue to dominate contemporary theoretical models of the disorder. These models vary considerably in their expositions of hyperactivity. For example, the cognitive-energetic model (Sergeant 2005) focuses exclusively on information processing and consequently contains no testable or falsifiable predictions concerning the role of activity level in ADHD. A second model views hyperactivity as incidental motor behavior that accompanies attention shifts away from non-novel tasks or activities. These shifts occur because behavior-consequence relationships that are usually strengthened through operant conditioning extinguish too rapidly in children with ADHD unless immediate reinforcement is provided (Sagvolden et al. 2005). A third model hypothesizes that ADHD is due to a developmental delay in response inhibition (i.e., the ability to inhibit oneself in accordance with situational demands) that adversely influences executive functions such as working memory, self-regulation of affect/ emotion/arousal, and internalization of speech. Ubiquitous, non-goal directed motor movement (hyperactivity) reflects children's on-going struggle to inhibit task irrelevant behavior and regulate goal directed behavior (Barkley 1997). An alternative model envisions hyperactivity as a manifestation of subcortical impairment that remains relatively static throughout life and is unrelated to executive functions such as working memory (Halperin et al. 2008). Other models imply that increased activity level represents children's attempt to minimize the aversive nature of delayed consequences by engaging in avoidance or escape behavior (Sonuga-Barke et al. 1992), or combine elements of delay and behavioral inhibition models (Sonuga-Barke 2002). Collectively, most contemporary models of ADHD largely disregard the role of hyperactivity, view it as ubiquitous behavior secondary to pervasive cognitive deficits, or envision it as corollary behavior that accompanies frequent attentional shifts or efforts to escape situations involving delayed consequences. Only one study has empirically investigated the relationship between these model-implied deficits and children's activity level. The authors concluded that activity level is a manifestation of subcortical impairment and independent of executive functions such as working memory (Halperin et al. 2008).

The negligible role most contemporary ADHD models afford hyperactivity is at odds with the empirical literature. Activity level is the first enduring trait or personality characteristic to develop in humans (Eaton et al. 1996), is highly heritable (Wood et al. 2007; Saudino and Eaton 1991), and remains remarkably stable during preschool years despite differences in context and environment (Rapport et al. 2006). Above average motor activity predicts (beyond age four) a diagnosis of ADHD at age nine (Campbell and Ewing 1990; Palfrey et al. 1985) and

portends a wide range of pejorative outcomes. These include externalizing behavior problems (Keown and Woodward 2006), interpersonal and parent–child difficulties (Buss 1981; Fischer and Barkley 2006), scholastic underachievement (Fergusson et al. 1997) and deficient occupational functioning (Barkley et al. 2006; Mannuzza et al. 1993) among others. Excessive motor activity also appears to be the only empirically documented symptom that uniquely distinguishes children diagnosed with ADHD from those with other childhood disorders (Halperin et al. 1992). Finally, the recognition that hyperactive symptoms are conventionally used to diagnose the research participants upon which contemporary models are based is fraught with irony.

In contrast to other contemporary models, the nascent working memory (WM) model makes specific, testable predictions concerning the functional role of hyperactivity in children with ADHD (Rapport et al. 2001, 2008b). Specifically, the model postulates that challenges to underlying working memory components engender increased movement in all children as a process that augments arousal necessary for task performance. The relationships among CNS arousal, increased activity level, and task performance are well established (for reviews, see Andreassi 1995; Barry et al. 2005; Zentall and Zentall 1983). Higher rates of movement are predicted to occur under WM conditions in children with ADHD relative to typically developing children to help compensate for the chronic cortical under-arousal associated with the disorder. Evidence for prefrontal cortical hypo-activation[1] as a core underlying physiological process in ADHD has been consistently verified by studies reporting increased slow wave (theta) and decreased fast wave (beta) activity in children with ADHD while performing academic (Mann et al. 1992) and cognitive tasks (Clark et al. 1998; Dickstein et al. 2006; El-Sayed et al. 2002) relative to typically developing children. Similar evidence has emerged from fMRI studies (Castellanos et al. 1996; Rubia et al. 1999).

Two studies recently examined predictions stemming from the WM model (Martinussen and Tannock 2006; Willcutt et al. 2005a); however, neither found a significant relationship between hyperactivity and WM performance despite finding deficient WM in children with ADHD relative to typically developing children. Two methodological confounds may have precluded the detection of a WM-activity level relationship in the studies: (a) the discrepant time parameters for measuring hyperactivity relative to WM performance, and (b) the reliance on subjective parent/ teacher rating scale scores to estimate children's activity level. The ratings scales used by Willcutt et al. (2005a) and

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[1] Prefrontal cortical hypo-activation refers to deficient task-related changes in arousal.

Martinussen and Tannock (2006) reflect adult retrospective perceptions of children's activity level throughout the day across multiple settings for the preceding week and month, respectively, in contrast to the brief time (typically 5 to 15 min) required to complete the WM tasks in both studies (i.e., digit and location span tasks). Controlling for setting and time parameter effects, however, does not remedy the low agreement ($r=0.32$ to 0.58) conventionally found between subjective (e.g., rating scale scores) and objective measures of children's activity. These values indicate that 66% to 91% of the variability in activity rating scale scores is not linearly related to variability in actigraph scores in the same children measured at the same time in the same setting (Rapport et al. 2006). This discrepancy is potentially problematic given (a) the ability of actigraphs but not hyperactivity ratings to differentiate hyperactive from impulsive subtypes of ADHD (Marks et al. 1999), and (b) the improved predictive validity of actigraphs for differentiating groups of ADHD children from both typically developing and other clinical groups compared to hyperactivity ratings (Halperin et al. 1992).

The present study is the first to investigate the relationship between children's WM and objectively measured activity level using experimental paradigms based on Baddeley's (2007) model.[2] Baddeley's model views WM as a multi-component system consisting of two independent subsystems—phonological (PH) and visuospatial (VS)—that are each equipped with unique input processors, a buffer for the temporary store of modality specific information (PH, VS), and a rehearsal mechanism. The domain-general central executive (CE) provides oversight and coordination of the two subsystems, reacts to changing attentional/multi-task demands, and provides a link between WM and long-term memory. The distinct functioning of the two subsystems, their storage/rehearsal components, and the domain-general CE are supported by extensive neuropsychological (Baddeley 2003), neuroanatomical (Smith et al. 1996), neuroimaging (Fassbender and Schweitzer 2006), and factor analytic (Alloway et al. 2006) investigations.

Children with ADHD and typically developing children were both expected to exhibit increased motor activity while performing WM tasks relative to control conditions as predicted by the WM model (Rapport et al. 2001, 2008b). No predictions were offered concerning whether motor activity would increase to some minimal threshold level to reflect general WM task demands (i.e., reflect primarily CE processing capabilities such as focused attention), or rise incrementally in response to the greater number of stimuli to be recalled (i.e., reflect storage/rehearsal processes). The issue was addressed statistically, however, by isolating and subsequently comparing activity level associated with the domain-general CE and subsystem (PH, VS) processes. Children with ADHD also were predicted to exhibit significantly higher rates of motor activity relative to typically developing children across both WM modalities. This prediction was based on recent experimental (Rapport et al. 2008a) and meta-analytic (Martinussen et al. 2005) findings demonstrating deficient CE, phonological, and visuospatial WM processes in children with ADHD relative to typically developing children. Finally, the two groups were compared under minimal WM control conditions before and after removing variance associated with WM performance to address the conventionally held belief that hyperactivity in children with ADHD is ubiquitous and unrelated to setting/task variables (Porrino et al. 1983).

Method

Participants

The sample was comprised of 23 boys aged 8 to 12 years ($M=9.04$, $SD=1.36$), recruited by or referred to the Children's Learning Clinic-IV (CLC-IV) through community resources (e.g., pediatricians, community mental health clinics, school system personnel, self-referral). The CLC-IV is a research-practitioner training clinic known to the surrounding community for conducting developmental and clinical child research and providing *pro bono* comprehensive diagnostic and psychoeducational services. Its client base consists of children with suspected learning, behavioral or emotional problems, as well as typically developing children (those without a suspected psychological disorder) whose parents agree to have them participate in developmental/clinical research studies. A psychoeducational evaluation was provided to the parents of all participants.

Two groups of children participated in the study: children with ADHD, and typically developing children without a psychological disorder. All parents and children gave their informed consent/assent to participate in the study, and the university's Institutional Review Board approved the study prior to the onset of data collection.

Group Assignment

All children and their parents participated in a detailed, semi-structured clinical interview using the Kiddie Schedule for Affective Disorders and Schizophrenia for School-Aged Children (K-SADS; Kaufman et al. 1997). The K-SADS

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[2] Children with ADHD were previously shown to exhibit significant WM deficits relative to typically developing children in CE and both working memory subsystems using these paradigms (Rapport et al. 2008a).

assesses onset, course, duration, severity, and impairment of current and past episodes of psychopathology in children and adolescents based on DSM-IV criteria. Its psychometric properties are well established, including interrater agreement of 0.93 to 1.00, test–retest reliability of 0.63 to 1.00, and concurrent (criterion) validity between the K-SADS and psychometrically established parent rating scales.

Twelve children met the following criteria and were included in the ADHD-Combined Type group: (1) an independent diagnosis by the CLC-IV's directing clinical psychologist using DSM-IV criteria for ADHD-Combined Type based on K-SADS interview with parent and child which assesses symptom presence and severity across home and school settings; (2) parent ratings of at least 2 *SD*s above the mean on the Attention Problems clinical syndrome scale of the Child Behavior Checklist (CBCL; Achenbach and Rescorla 2001), or exceeding the criterion score for the parent version of the ADHD-Combined subtype subscale of the Child Symptom Inventory (CSI; Gadow et al. 2004); and (3) teacher ratings of at least 2 *SD*s above the mean on the Attention Problems clinical syndrome scale of the Teacher Report Form (TRF; Achenbach and Rescorla 2001), or exceeding the criterion score for the teacher version of the ADHD-Combined subtype subscale of the CSI (Gadow et al. 2004). The CSI requires parents and teachers to rate children's behavioral and emotional problems based on DSM-IV criteria using a 4-point Likert scale. The CBCL, TRF, and CSI are among the most widely used behavior rating scales for assessing psychopathology in children. Their psychometric properties are well established (Rapport et al. 2008b). All children in the ADHD group met criteria for ADHD-Combined Type, and six were comorbid for Oppositional Defiant Disorder (ODD).

Eleven children met the following criteria and were included in the typically developing group: (1) no evidence of any clinical disorder based on parent and child K-SADS interview; (2) normal developmental history by maternal report; (3) ratings below 1.5 *SD*s on the clinical syndrome scales of the CBCL and TRF; and (4) parent and teacher ratings within the non-clinical range on all CSI subscales. Typically developing children were actively recruited through contact with neighborhood and community schools, family friends of referred children, and other community resources.

Children that presented with (a) gross neurological, sensory, or motor impairment, (b) history of a seizure disorder, (c) psychosis, or (d) Full Scale IQ score less than 85 were excluded from the study. None of the children were receiving medication during the study—seven of the children with ADHD had previously received trials of psychostimulant medication. Demographic and rating scale data for the two groups are provided in Table 1.

Measures and Statistical/Methodological Overview

Measures

Actigraph An actigraph is an acceleration-sensitive device that measures motor activity. The estimated reliability for actigraphs placed at the same site on the same person ranges from 0.90 to 0.99 (Tryon 1985). Actigraphs are moderately correlated with parent and teacher ratings of activity level ($r=0.32$ to 0.58), and have superior predictive validity relative to parent and teacher ratings of hyperactivity for differentiating among children with ADHD, typically developing children, and children with other psychopathological disorders (Halperin et al. 1992; Rapport et al. 2006). Actigraphs generate a current (voltage) each

Table 1 Sample and Demographic Variables

Variable	ADHD		Typically developing		
	$\overline{X}$	SD	$\overline{X}$	SD	*F*
Age	8.75	1.29	9.36	1.43	1.17
FSIQ	100.92	15.22	110.18	13.11	2.43
SES	43.46	12.25	52.50	7.57	6.13*
CBCL					
Attention problems	78.50	10.53	55.64	7.06	36.68***
TRF					
Attention problems	66.25	8.83	48.73	16.92	9.94**
CSI-parent					
ADHD, combined	12.67	3.85	3.00	4.98	27.42***
CSI-teacher					
ADHD, combined	9.83	5.32	2.73	3.93	13.06**

ADHD attention-deficit/hyperactivity disorder, *CBCL* Child Behavior Checklist, *CSI* Child Symptom Inventory, *FSIQ* full scale intelligence quotient, *SES* socioeconomic status, *TRF* Teacher Report Form

*$p\leq0.05$; **$p\leq0.01$; ***$p\leq0.001$

time the instrument is moved. The current is passed through an amplifier and filtered, resulting in an analog waveform—a histogram of measured voltage over time—from which data regarding movement frequency, intensity, or duration may be extracted and analyzed (for a detailed review, see Rapport et al. 2006). MicroMini Motionlogger® (Ambulatory Monitoring Inc. 2004) actigraphs were used to measure children's activity level. The acceleration-sensitive devices resemble wristwatches and were set to Proportional Integrating Measure (low-PIM) mode, which measures the intensity of movement (i.e., quantifies gross activity level). Movement was sampled 16 times per second (16 Hz) and collapsed into 1-minute epochs. Data were downloaded via a hardware interface and analyzed using the Action-W2 software program (Ambulatory Monitoring Inc. 2004) to calculate mean activity rates for each child during the control and WM tasks described below.

Children were told that the actigraphs were "special watches" that let them play the computer learning games. The Observer (Noldus Information Technology 2003) live observation software was used to code start and stop times for each task, which were matched to the time stamps from the actigraphs. Actigraphs were placed immediately above children's left and right ankles using velcro watch bands. Ankle placement was used *in lieu* of trunk placement due to the improved sensitivity of the former for detecting movement (Eaton et al. 1996). A third actigraph was placed on children's non-dominant wrist only, because the visuospatial and both control tasks required movement using the dominant hand.

Phonological (PH) Working Memory Task The phonological WM task is similar to the Letter–Number Sequencing subtest on the WISC-IV (Wechsler 2003), and assesses phonological WM 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 was counterbalanced across trials to appear 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 correctly recalled as 2 4 6 H). Two trained research assistants, shielded from the participant's view, independently recorded oral responses (interrater reliability=95.6% agreement).

Visuospatial (VS) Working Memory Task Children were shown nine 3.2 cm squares arranged in three vertical columns on a computer monitor. The columns were offset

from a standard 3×3 grid to minimize the likelihood of phonological coding of the stimuli (e.g., by equating the squares to numbers on a telephone pad). 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 but one dot presented within the squares was black—the exception being a red dot that was counterbalanced across trials to appear an equal number of times in each of the nine squares, but never presented as the first or last stimulus in the sequence to minimize potential primacy and recency effects. Each dot was displayed for 800 ms followed by a 200 ms interstimulus interval. A green light appeared at the conclusion of each 3, 4, 5, and 6 stimulus sequence. Children were instructed to indicate the serial position of black dots in the order presented by pressing the corresponding squares on a computer keyboard, and to indicate the position of the red dot last. The last response was followed by an intertrial interval of 1,000 ms and an auditory chime that signaled the onset of a new trial.

Control (C) Conditions Children's activity level was assessed while they used the Microsoft® Paint program for five consecutive minutes both prior to (C1) and after (C2) completing the phonological and visuospatial WM tasks during four consecutive Saturday assessment sessions. The Paint program served as pre and post conditions to assess and control for potential within-day fluctuations in activity level (e.g., fatigue effects). Children sat in the same chair and interacted with the same computer used for the WM tasks while interacting with a program that placed relatively modest demands on WM (i.e., the Paint program allows children to draw/paint anything they like on the monitor using a variety of interactive tools[3]). The four pre and four post activity level control conditions were separately averaged to create pre and post composite scores secondary to preliminary analyses that found no significant differences in children's pre or post condition activity level across days (all $p>0.10$).

Measured Intelligence All children were administered either the Wechsler Intelligence Scale for Children third or fourth edition to obtain an overall estimate of intellectual functioning. The changeover to the fourth edition was due to its release during the conduct of the study and to provide parents with the most up-to-date intellectual evaluation possible. Full Scale IQ (FSIQ) was not analyzed as a covariate because it shares significant variance with WM

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[3] Successful interaction with the Paint program requires central executive processes such as focused attention and interaction with long-term memory, as well as limited phonological and visuospatial storage/rehearsal processes.

and would result in removing substantial variance associated with WM from WM (Ackerman et al. 2005). Instead, a residual FSIQ score was derived using a latent variable approach. Briefly, the derived central executive, phonological storage/rehearsal, and visuospatial storage/rehearsal performance variables described below were covaried out of FSIQ ($R^2=0.31$). Residual FSIQ scores represent IQ that is unrelated to estimated WM functioning, and were examined as a potential covariate in the analyses described below.

Procedures

The phonological and visuospatial tasks were programmed using Superlab Pro 2.0 (2002). All children participated in four consecutive Saturday assessment sessions at the CLC. The phonological, visuospatial, and control conditions were administered as part of a larger battery of laboratory-based tasks that required the child's presence for approximately 2.5 h per session. Children completed all tasks while seated alone in an assessment room. All children received brief (2–3 min) breaks following every task, and preset longer (10–15 min) breaks after every two to three tasks to minimize fatigue. Each child was administered eight control (pre and post on each of the 4 days), four phonological, and four visuospatial conditions (i.e., PH and VS set sizes 3, 4, 5, and 6) across the four testing sessions. Each phonological and visuospatial set size consisted of 24 trials. Details concerning the administration of practice blocks for the visuospatial and phonological paradigms are described in Rapport et al. (2008a). The eight WM conditions were counterbalanced to control for order effects. The control conditions always occurred as the first and last tasks each day. Children were seated in a caster-wheel swivel chair approximately 0.66 m from the computer monitor for all tasks.

Dependent Variables

Total extremity scores (TES) were calculated by summing activity level across the three actigraph sites (2 ankle, 1 non-dominant hand) to compute an estimate of overall movement for each of the ten conditions (C1, PH and VS set sizes 3, 4, 5, and 6, and C2).[4] An aggregate measure of activity level was employed *in lieu* of reporting separate extremity activity rates or using data reduction techniques such as averaging due to expected inter-individual differences

in movement across children's extremities while completing cognitive tasks (Eaton et al. 1996). This approach has the additional advantage of conserving power while providing the broader sampling of children's activity level needed to test hypotheses regarding the relationship between overall activity level and WM.

Performance data (PH and VS stimuli correct per trial) were computed and used to statistically isolate the relationship between activity level and specific components of WM.

Statistical Analysis

A 4-tier analytic approach was used to examine (a) potential overall group differences in activity level between WM modalities (PH,VS); (b) group differences and changes in activity level associated with overall phonological and visuospatial WM demands; (c) the extent to which activity level is directly related to individual WM component processes, and whether this relationship differs between children with ADHD and typically developing children; and (d) whether hyperactivity is a ubiquitous feature of ADHD or functionally related to WM. Measurement of activity level while children performed WM tasks allowed direct examination of the relationship between WM and hyperactivity, providing incremental benefit beyond the correlational studies described earlier. Hedges' g effect sizes were computed to estimate the magnitude of all between-group differences while correcting for sample size (Lipsey and Wilson 2001).

Results

Data Screening

Power Analysis An average effect size (ES) of 0.72 was calculated from two studies providing actigraph means and *SD*s for children with ADHD and typically developing (TD) children during laboratory tasks (Dane et al. 2000; Halperin et al. 1992). GPower software version 3.0.5 (Faul et al. 2007) was used to determine needed sample size using this ES, with power set to 0.80 as recommended by Cohen (1992). For an ES of 0.72, $\alpha=0.05$, power $(1-\beta)=0.80$, 2 groups, and six repetitions (C1, set sizes 3–6, C2 as described below), 18 total subjects are needed for a repeated measures ANOVA to detect differences and reliably reject H_0.

Outliers Each of the ten tasks (C1, PH set sizes 3–6, VS set sizes 3–6, C2) was screened for univariate outliers (i.e., ≥ 3.5 SD above or below group mean). No univariate outliers were identified.

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[4] Site placement contrasts for each task revealed that non-dominant hand movement was greater than left and right foot movement across most conditions (i.e., NH > LF = RF). The pattern of results across conditions for the three actigraph recording sites, however, did not differ significantly from those reported for TES in the Results.

Preliminary Analyses

Demographic data are shown in Table 1. Sample ethnicity was mixed with 16 Caucasian (69%), 5 Hispanic (22%), and 2 African American (9%) children. All parent and teacher behavior rating scale scores were significantly higher for the ADHD group relative to the TD group as expected (see Table 1). Children with ADHD and TD children did not differ on age ($p=0.14$), or intelligence (WISC-III or WISC-IV FSIQ; $p=0.13$). A univariate ANOVA revealed significant between-group differences in SES ($p=0.02$). On average, children with ADHD had lower Hollingshead (1975) SES scores than TD children. Age and SES were not significant covariates of any of the Tier I, II, III, or IV analyses (all $p\geq 0.24$). Residual FSIQ did not differ between groups, $F(1,21)= 0.48$, $p=0.83$. The residual FSIQ score was a significant covariate of all Tier I, II, and IV analyses (all $p\leq0.05$), but did not change the pattern of any results. Residual FSIQ was not a significant covariate of the Tier III analysis ($p=0.97$). We therefore report simple model results with no covariates. Means, SDs, and F-values are presented in Tables 2, 3, and 4.

Tier I: Composite Scores

The initial analysis examined overall differences in activity level between WM modalities (PH, VS) and groups (ADHD, TD). Results are depicted in Table 2. Phonological and visuospatial composite scores were computed separately by averaging activity level across set sizes. A Mixed-model ANOVA indicated significant main effects for WM modality ($p=0.004$) and group ($p<0.001$). Across groups, children were significantly more active during the phonological relative to the visuospatial task; children with ADHD were significantly

Table 2 Phonological and visuospatial Total Activity Level Composite Scores

	Phonological $\overline{X}$ (SD)	Visuospatial $\overline{X}$ (SD)	Composite $\overline{X}$ (SE)	F
ADHD	43785.61 (10504.79)	39626.21 (7071.20)	41705.91 (2136.72)	
TD	25477.21 (7557.72)	20574.21 (6737.30)	23025.71 (2231.74)	
Composite	35029.42 (12983.89)	30514.39 (11845.82)	–	10.14**
F				36.55***

Actigraph Proportional Integrating Measure (PIM) values can range from 0 (no movement) to 65,535

ADHD attention-deficit/hyperactivity disorder; SE standard error; TD typically developing children

[a] Modality × group interaction, $p=0.80$, ns

**$p<0.01$; ***$p<0.001$

more active than TD children across all tasks. The modality by group interaction was not significant ($p=0.80$).

Tier II: Set Sizes

The second set of analyses examined the effects of increasing phonological and visuospatial memory load on children's activity level (see Tables 3 and 4). Using Wilks' criterion, a significant one-way MANOVA on all ten conditions (C1, set sizes 3–6 for both modalities, C2) by group (ADHD, TD) confirmed the overall relationship between activity level and WM, Wilks' $\lambda=0.19$, $F(10,11)=4.64$, $p=0.009$. Phonological and visuospatial Mixed-model ANOVAs with LSD post hocs were conducted separately to examine group (ADHD, TD) by condition (C1, set sizes 3–6, C2) differences.

Phonological ANOVA For activity level during the phonological and control conditions (C1, PH set sizes 3–6, C2), the Mixed-model ANOVA was significant for group ($p<0.0005$) and set size ($p<0.0005$). The post hoc test for set size is reported in Table 3. The group by set size interaction was also significant ($p=0.001$). LSD post hoc tests for the interaction revealed that children with ADHD demonstrated greater activity level across all control and phonological set size conditions compared to TD children (all $p\leq0.04$). The pattern of activity level between control and phonological conditions for both groups was also similar. Both groups exhibited higher rates of activity under all phonological conditions (set sizes 3–6) relative to both control conditions (C1, C2; all $p\leq0.05$). Children with ADHD exhibited significantly higher rates of movement than TD children across all conditions, but their rates were not statistically different across the four phonological set size conditions (i.e., ADHD PH 3=4=5=6; all $p\geq 0.31$). Typically developing children also evidenced a stable pattern of activity level across the phonological condition with one exception—their activity level was moderately lower under set size 3 relative to set sizes 4 ($p=0.05$) and 6 ($p= 0.03$). Computation of Hedges' g indicated that the average magnitude difference between children with ADHD and TD children was 1.49 standard deviation units (range: 0.93 to 2.10). Results are depicted in Table 3 and Fig. 1 (top).

Visuospatial ANOVA For activity level during the visuospatial and control conditions (C1, VS set sizes 3–6, C2), the Mixed-model ANOVA was significant for group ($p< 0.0005$) and set size ($p<0.0005$). The post hoc test for set size is reported in Table 4. The group by set size interaction was also significant ($p=0.02$). LSD post hoc tests for the interaction revealed that children with ADHD exhibited significantly higher rates of activity across all control and visuospatial conditions relative to TD children (all $p\leq 0.003$). The pattern of activity level between control and visuospatial conditions for both groups was similar. Both

Table 3 Phonological Activity Level Set Size Analyses

	Phonological set size[a]								
	C1	PH 3	PH 4	PH 5	PH 6	C2	Group composite		
	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$		
	(SD)	(SD)	(SD)	(SD)	(SD)	(SD)	(SE)	F	Set size contrasts
ADHD	16120.23	45963.33	41953.19	41776.05	46613.42	21346.11	35628.72	28.93***	C1<C2< 3=4=5=6
	(7210.24)	(13999.73)	(17143.45)	(11428.42)	(10308.03)	(9799.32)	(2409.48)		
TD	8582.92	19285.65	28141.98	23858.93	30622.28	12055.10	20424.48	14.77***	C1<C2<3< 4=5=6; 3=5
	(2703.27)	(9933.66)	(10161.86)	(10360.81)	(12623.68)	(4557.34)	(2409.48)		
Set size composite	12351.57	32624.49	35047.59	32817.49	38617.85	16700.59	–	42.16***	C1<C2< 3=4=5=6
	(6566.19)	(18075.29)	(15462.29)	(14049.54)	(13908.84)	(8844.52)			
Group F	9.96**	28.10***	4.85*	17.17***	10.09**	8.13**	45.57***		
Group contrasts	A>TD	A>TD	A>TD	A>TD	A>TD	A>TD	A>TD		

Actigraph Proportional Integrating Measure (PIM) values can range from 0 (no movement) to 65,535; ADHD $n=11$ for all analyses involving C2 condition due to missing data for one participant; A = ADHD; C1 = control condition (pre); C2 = control condition (post); PH = phonological (3, 4, 5, & 6 set size); SE = standard error; TD = typically developing children

[a] Phonological group x set size interaction, $F(5,100)=4.28$, $p=0.001$; *$p<0.05$; **$p\leq0.01$; ***$p\leq0.001$

groups exhibited higher levels of activity under all four visuospatial conditions (set sizes 3–6) relative to both control conditions (C1, C2; all $p\leq0.006$). In addition, activity level for both groups of children (ADHD, TD) remained relatively stable across the four visuospatial set size conditions (all $p\geq0.34$). The significant interaction effect was due to the disproportionate decrease in activity level from WM to control conditions for children with ADHD relative to TD children (i.e., a significant ES decrease of 0.66 in the magnitude of group differences from visuospatial WM tasks to C2, one-sample $t(7)=2.67$, $p=0.03$). When the control conditions were removed from the analysis, neither the main effect for set size nor the group by set size interaction remained significant (both $p\geq0.61$). Hedges' g effect size indicated that the average magnitude difference in activity level between children with ADHD and TD children during visuospatial WM tasks was 1.83 standard deviation units (range=1.47 to 2.67). Children in both groups were some-

Table 4 Visuospatial Activity Level Set Size Analyses

	Visuospatial set size[a]								
	C1	VS 3	VS 4	VS 5	VS 6	C2	Group composite		
	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$	$\overline{X}$		
	(SD)	(SD)	(SD)	(SD)	(SD)	(SD)	(SE)	F	Set size contrasts
ADHD	16120.23	42156.64	39179.69	40050.91	39029.78	21346.11	32980.56	13.52***	C1<C2< 3=4=5=6
	(7210.24)	(13138.28)	(14222.47)	(7751.39)	(14447.63)	(9799.32)	(1847.52)		
TD	8582.92	22673.08	21456.97	18403.58	19763.22	12055.10	17155.81	12.43***	C1<C2< 3=4=5=6
	(2703.27)	(9843.00)	(7846.21)	(7862.40)	(7806.69)	(4557.34)	(1847.52)		
Set size composite	12351.57	32414.86	30318.33	29227.24	29396.50	16700.59	–	23.63***	C1<C2< 3=4=5=6
	(6566.19)	(15091.54)	(14418.80)	(13445.39)	(15021.23)	(8844.52)			
Group F	9.96**	16.68***	11.45**	43.71***	16.47***	8.13**	22.63***		
Group contrasts	A>TD	A>TD	A>TD	A>TD	A>TD	A>TD	A>TD		

Actigraph Proportional Integrating Measure (PIM) values can range from 0 (no movement) to 65,535; ADHD $n=11$ for all analyses involving C2 condition due to missing data for one participant; A = ADHD; C1 = control condition (pre); C2 = control condition (post); SE = standard error; TD = typically developing children; VS = visuospatial (3, 4, 5, & 6 indicate set size)

[a] Visuospatial group x set size interaction, $F(5,100)=2.94$, $p=0.016$; **$p\leq0.01$; ***$p\leq0.001$
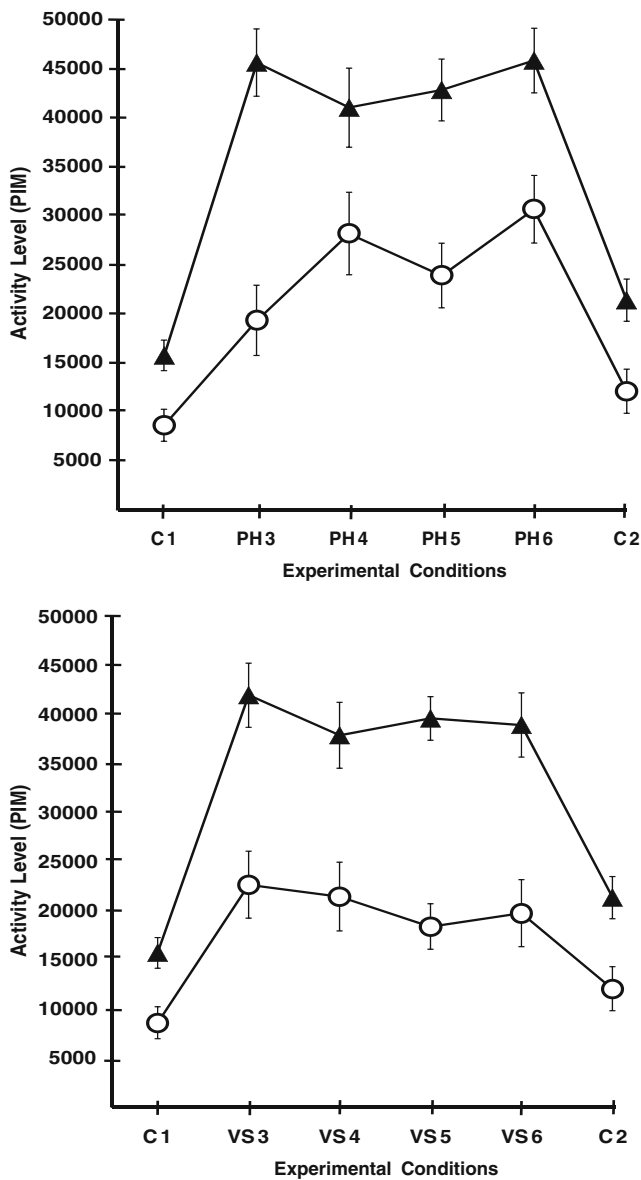
Fig. 1 Total extremity activity level (right foot, left foot, and non-dominant hand) expressed in PIM (Proportional Integrated Measure) units for children with ADHD (*triangles*) and typically developing children (*circles*) under control (C1, C2) and four phonological (top graph: PH 3, 4, 5, 6) and four visuospatial (bottom graph: VS 3, 4, 5, 6) working memory set size conditions. *Vertical bars* represent standard error

what more active during the second relative to the first control condition (both $p \leq 0.01$). Results are depicted in Fig. 1 (bottom).

Tier III: Components of Working Memory

Latent variable analyses were undertaken to determine the extent to which group differences in activity level reported above were associated with the domain-general central executive relative to the two subsidiary systems (PH or VS

storage/rehearsal). Latent variable analysis is currently the best practice for estimating the independent contribution of WM component processes (cf. Swanson and Kim 2007).

Phonological Storage/Rehearsal Latent variable analyses were used to estimate shared variance between the derived phonological storage/rehearsal performance variables (described above) and phonological activity level at each set size (i.e., activity level directly related to PH storage/rehearsal functioning). Results indicated that phonological storage/rehearsal functioning was not a significant contributor to objectively measured activity level (average $R^2 = 0.10$; values ranged from 0.06 to 0.21 and were all nonsignificant with one exception[5]). The planned t-test for group differences was not conducted because children's activity level and phonological storage/rehearsal functioning were not significantly related.

Visuospatial Storage/Rehearsal An identical latent variable approach was used to estimate activity level directly related to visuospatial storage/rehearsal functioning. Results indicated that visuospatial storage/rehearsal functioning was not a significant contributor to objectively measured activity level (average $R^2 = 0.07$; values ranged from less than 0.001 to 0.14 and were all nonsignificant). The planned t-test for group differences was not conducted because children's activity level and visuospatial storage/rehearsal functioning were not significantly related.

Central Executive Latent variable analyses were again used to derive predicted scores that reflect shared variance between the derived CE performance variables (described above) and children's activity level during the phonological and visuospatial tasks at each set size (i.e., activity level directly related to CE functioning). Results indicated that CE functioning was a significant contributor of objectively measured activity level (average $R^2 = 0.32$; values ranged from 0.17 to 0.61; all $p \leq 0.04$). A composite score was computed by averaging the four predicted scores for each task to provide an overall estimate of children's activity level directly associated with CE functioning. An independent samples t-test on the derived CE-activity level variable indicated a significant between-group difference, $t(21) = 7.54$, $p < 0.0005$, with children with ADHD evincing higher rates of activity directly associated with CE functioning relative to TD children. Hedges' g effect size indicated that the average magnitude difference between children with ADHD and TD children was 3.03 standard deviation units ($SE = 0.60$).

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[5] The relationship between phonological storage/rehearsal functioning and activity level at set size 5 ($R^2 = 0.21$) was significant at $p = 0.03$.

Tier IV: Control Conditions

Latent variable analyses were used in the final tier to assess the extent to which observed group differences in activity level during the two control conditions (C1, C2) represent ubiquitous hyperactivity in children with ADHD (Porrino et al. 1983) or the influence of minimal WM demands associated with the Paint program (Rapport et al. 2008a). Residual scores were computed for both control tasks by regressing the CE composite performance variable onto C1 ($R^2=0.26$) and C2 ($R^2=0.25$) activity level to remove variance associated with CE functioning.[6] A 2 (group) by 2 (condition: C1, C2) Mixed-model ANOVA was nonsignificant for group, condition, and the group by condition interaction (all $p \geq 0.52$), indicating that children with ADHD were not ubiquitously more motorically active than typically developing children during the control conditions after accounting for task-related WM demands. Hedges' g effect size indicated that the average magnitude difference between children with ADHD and TD children was 0.20 standard deviation units ($SE=0.29$), with a confidence interval that included 0.0.

Discussion

This is the first experiment to demonstrate a functional relationship between working memory and children's activity level. Children with ADHD and typically developing children both exhibited significantly higher rates of movement while performing phonological and visuospatial WM tasks relative to minimal WM control conditions. This finding contradicts the subcortical impairment model (Halperin et al. 2008), but is consistent with WM model predictions and provides initial evidence that movement is functionally related to the imposition of WM demands (Rapport et al. 2001, 2008b). Children's activity level was also moderately higher under the four phonological relative to visuospatial set size conditions, which may reflect differences in children's developing phonological and visuospatial abilities despite using identical cognitive loads across the two modalities (Alloway et al. 2006). The finding may also reflect subtle differences in processing demands and strategic resource use between the two tasks. Maintaining a limited set of over-learned items such as digits and letters within the phonological storage/rehearsal subsystem typically relies to some extent on long-term memory knowledge to clean up the memory trace during

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[6] Phonological and visuospatial storage/rehearsal composite scores were also used in the analysis initially but did not share significant variance with C1 and C2 activity level.

repeated rehearsal and/or at final recall (Baddeley 2007). The visuospatial subsystem cannot adopt this strategy to help recall unfamiliar material such as a novel matrix pattern. Coupled with our related finding that activity level is associated with CE and not storage/rehearsal processes, the higher activity rates under the phonological relative to the visuospatial WM conditions may reflect the increased demands on CE resources that facilitate the interplay between WM and long-term memory. An alternative explanation for the finding is that the moderately higher activity rates during the phonological relative to visuospatial conditions reflect subtle differences in response demands between the two tasks. Children attend to a computer monitor to view WM stimuli during both tasks; however, the phonological task requires a verbal response (which can be emitted while moving), whereas the visuospatial task requires a keyboard response that necessarily restricts movement to some extent.

Examination of between-group activity level differences revealed a relatively consistent pattern of results. Children with ADHD were more active than typically developing children across both modalities, and their activity level remained stable despite increases in cognitive demand. Typically developing children also evidenced a stable pattern of activity level across the phonological and visuospatial conditions with one exception—their activity level was moderately lower under the smallest phonological set size condition. The similar pattern of results across groups suggests that increases in children's activity level between control and phonological/visuospatial WM conditions primarily reflect general task demands associated with central executive processing rather than increases in cognitive load imposed on the storage/rehearsal loop subsidiary systems. This interpretation was confirmed statistically by isolating the unique contributions of the domain-general central executive, phonological storage/rehearsal, and visuospatial storage/rehearsal to children's activity level. Latent variable analysis revealed that WM performance attributable to central executive functioning—but not phonological or visuospatial storage/rehearsal functioning—was significantly related to children's activity level.

The significant relationship between CE functioning and activity level appears at odds with previous correlational studies that failed to find a significant relationship between WM and activity level (Martinussen and Tannock 2006; Willcutt et al. 2005a). This apparent discrepancy may reflect previously discussed methodological differences among the studies (e.g., nonconcurrent measurement of WM and activity level, subjective measures of activity level). A more likely explanation is that the WM tasks used in earlier studies (e.g., digits forward and backward tasks) primarily reflect visuospatial and phonological storage/ rehearsal processes (Swanson and Kim 2007), whereas

only CE processes appear to be functionally related to children's activity based on the current results. In this case, our finding that storage/rehearsal processes are not significantly related to children's activity level is consistent with previous correlational findings.

Collectively, children with ADHD showed disproportionately higher motor activity relative to typically developing children under both control and all WM conditions. This finding is consistent with those reported in previous investigations of actigraph-measured activity in laboratory and classroom settings. Extant studies uniformly reported higher activity level in children with ADHD relative to typically developing controls during laboratory-based experimental tasks (Dane et al. 2000; Halperin et al. 1992, 1993; Inoue et al. 1998). The extent to which performance on these tasks (e.g., CPT, Stop-signal Task) is mediated by WM processes, however, is currently unknown. Higher activity rates are also consistently observed in children with ADHD relative to typically developing children while completing in-seat academic assignments (Porrino et al. 1983; Tsujii et al. 2007[7]), which are known to place moderate to heavy demands on WM resources (Gathercole et al. 2004).

Although previous actigraph studies are consistent in documenting higher activity rates in children with ADHD, little is known about the underlying processes responsible for these differences. Prevailing hypotheses suggest that higher activity in children with ADHD relative to typically developing children is ubiquitous and largely independent of task and situational demands (Porrino et al. 1983, p. 685). Our finding that children with ADHD were more motorically active relative to their peers under minimal WM conditions appeared consistent with this view, but at odds with predictions stemming from the WM model of ADHD (Rapport et al. 2001, 2008b). Their higher motor activity during control conditions, however, was fully attenuated by removing the influence of WM demands associated with these tasks. This finding, coupled with the previously discussed results, suggests that activity may serve a purposeful function in all children to the extent that arousal is necessary for CE processing, and becomes excessive in some children to compensate for chronic cortical under-arousal (Dickstein et al. 2006; El-Sayed et al. 2002; Mann et al. 1992). Experimental studies concurrently examining WM performance, motor activity, physiological arousal, and cortical activity are needed to further explicate the complex interplay among these processes. The outcome of these studies will help clarify whether hyperactivity might be better characterized as an effect educed by CE processing deficits rather than as a core causal variable.

The current study's unique contribution was the objective measurement of activity level during concurrent manipulation of domain-general CE processing and subsidiary storage/rehearsal demands while controlling for IQ, age, and SES. Several caveats require consideration when interpreting the present findings despite these and other methodological refinements (i.e., controlling IQ-WM co-variation, pre/post activity level measurement, and WM component partitioning). The generalization of results from highly controlled, laboratory-based experimental investigations with stringent inclusion criteria to the larger population of children with ADHD is always limited to some extent. Independent experimental replication with larger samples that include females, older children, and other ADHD subtypes is recommended to address these potential limitations. Our cell sizes were nevertheless sufficient based on the *a priori* power analysis. The large magnitude between-group differences in motor activity associated with the imposition of WM demands observed in the study may be related to our stringent inclusion criteria, and would likely be attenuated to the extent that children exhibit fewer or less disabling ADHD-related symptoms. This supposition is consistent with the strong genetic contribution associated with activity level (Wood et al. 2007; Saudino and Eaton 1991) and evidence that ADHD behavioral symptoms represent continuous rather than categorical dimensions (Gjone et al. 1996). Several of the children with ADHD also met diagnostic criteria for ODD; however, the degree of comorbidity may be viewed as typical of the ADHD population based on recent epidemiological findings (i.e., 59%; Wilens et al. 2002), and previous investigations indicate that the excess motor activity observed in ADHD is independent of ODD (Halperin et al. 1992). The specificity of disproportionately high activity rates found in our ADHD sample is currently unknown and merits investigation. Actigraph studies comparing children with ADHD and children with other clinical disorders, such as anxiety and conduct disorder, are thus far inconclusive due to insufficient statistical power (Halperin et al. 1993). Children with other clinical disorders are likely to exhibit lower activity rates than children with ADHD but higher rates than typically developing children to the extent that CE processes are disrupted. A final caveat worth noting is that actigraph-measured activity reported herein may differ from rates reported in other studies due to measurement differences. Proportional integrating measure, rather than the zero-crossing mode used in previous studies, was selected because it quantifies movement intensity over time (i.e., how much movement occurs) rather than counting the frequency that a child's movement crosses a preset intensity threshold (which may underestimate activity). The use of multiple actigraphs provided a broader sampling of children's activity level, and wrist/

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[7] Tsujii et al. 2007 found these differences only during the afternoon hours.

ankle actigraph placements provided enhanced precision over truncal placement for assessing both gross and distal movements (cf. Rapport et al. 2006, and Eaton et al. 1996, for reviews).

The current findings indirectly address anecdotal parent and teacher reports that children with ADHD remain engaged in particular tasks and activities with no apparent excessive motor activity (e.g., computer activities, playing LEGO® or video games, watching TV), yet move excessively during most in-seat academic/learning activities (e.g., homework, classroom academic assignments). Volitional control deficits are often invoked to explain this apparent incongruity (McInerney and Kerns 2003). The current findings, however, suggest that activity rates in children with ADHD may vary among these activities as a function of differences in CE demands. Experimental paradigms that systematically vary a wider range of CE processing demands than used in the current study, while simultaneously measuring activity level, are needed to address this issue. The results also potentially shed light on the reduced motor movement observed in children prescribed psychostimulants—an effect described as paradoxical in years past. Psychostimulants are known to enhance cognitive performance in children (Douglas et al. 1995; Rapport and Kelly 1991) and WM in particular (Bedard et al. 2007). The accompanying reduced motor activity (Bedard and Tannock 2008) may reflect increased cortical arousal and improved CE processing (Lawrence et al. 2005).

Considering hyperactivity as a secondary symptom—whose presence reflects ongoing CE processing demands in the environment, rather than a core causal feature of the disorder—has several implications for intervention planning and treatment. Behavioral programs designed to reduce excessive gross motor activity in children through conventional behavior management techniques may be counterproductive and unintentionally decrease CE functioning. Programs specifically targeting CE functions such as focused attention, in contrast, are likely to prove beneficial as evidenced in past outcome studies (DuPaul et al. 1992). Efforts to develop interventions that promote the early development of WM abilities in children at risk for ADHD also appear warranted based on accumulating evidence from recent experimental investigations (Rapport et al. 2008a) and meta-analytic reviews (Martinussen et al. 2005; Willcutt et al. 2005b). To date, however, there is scant empirical support to indicate that direct training of WM capacity in children is beneficial (for an exception, cf. Klingberg et al. 2005). An alternative intervention approach—with growing empirical support—is to adopt curricula systems and methods that avoid and/or minimize WM failure in children. These include a wide variety of techniques that involve restructuring complex tasks, simplifying mental processing, and encouraging the use of memory aids such as memory cards,

information key rings, and audio devices (cf. Gathercole and Alloway 2008).

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