Hyperactivity in attention-deficit/hyperactivity disorder (ADHD): Impairing deficit or compensatory behavior?

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Abstract

Excess gross motor activity (hyperactivity) is considered a core diagnostic feature of childhood ADHD that impedes learning. This view has been challenged, however, by recent models that conceptualize excess motor activity as a compensatory mechanism that facilitates neurocognitive functioning in children with ADHD. The current study investigated competing model predictions regarding activity level's relation with working memory (WM) performance and attention in boys aged 8-12 years (*M*=9.64, *SD*=1.26) with ADHD (*n*=29) and typically developing children (TD; *n*=23). Children's phonological WM and attentive behavior were objectively assessed during four counterbalanced WM tasks administered across four separate sessions. These data were then sequenced hierarchically based on behavioral observations of each child's gross motor activity during each task. Analysis of the relations among intra-individual changes in observed activity level, attention, and performance revealed that higher rates of activity level predicted significantly better, but not normalized WM performance for children with ADHD. Conversely, higher rates of activity level predicted somewhat lower WM performance for TD children. Variations in movement did not predict changes in attention for either group. At the individual level, children with ADHD and TD children were more likely to be classified as reliably Improved and Deteriorated, respectively, when comparing their WM performance at their highest versus. lowest observed activity level. These findings appear most consistent with models ascribing a functional role to hyperactivity in ADHD, with implications for selecting behavioral treatment targets to avoid overcorrecting gross motor activity during academic tasks that rely on phonological WM.

Hyperactivity in attention-deficit/hyperactivity disorder (ADHD): Impairing Deficit or Compensatory Behavior?

Attention-deficit/hyperactivity disorder (ADHD) is a complex, chronic, and heterogeneous neurodevelopmental disorder whose cardinal behavioral features include developmentally inappropriate levels of inattention, impulsivity, and hyperactivity. Hyperactivity reflects a multifaceted construct that spans a broad range of verbal and physical child behaviors, with excess gross motor movement forming a key component as evidenced by its explicit inclusion in 4 of the 6 DSM-5 hyperactivity symptoms (APA, 2013). Subjective measures (e.g., symptom ratings, clinical interviews) are the most frequent indices of the hyperactivity construct; however, correlations with direct measures of gross motor activity are typically modest (.32 to .58; Rapport et al., 2006). This modest agreement between hyperactivity indices may reflect informant reporting biases (Harris & Lahey, 1982) and/or the substantial psychometric overlap of ratings with conceptually distinct behavioral dimensions such as impulsivity and inattention (DuPaul, 1991), and highlights the need for objective methods to clarify the role of excess gross motor activity in ADHD.

Developmental investigations of children's objectively-recorded gross motor movement indicate that activity level follows a curvilinear pattern (inverted U-shaped) across the lifespan (Eaton, McKeen, & Campbell, 2001). Activity level is the first enduring trait to develop in humans, with individual differences in motor activity at 28 weeks gestation reliably predicting children's motor activity in early infancy (Walters, 1965), which in turn predicts objectively measured motor activity in early childhood (Eaton, McKeen, & Saudino, 1996). The emergence of hyperactivity, or excess gross motor activity, displays an inverse relation with positive behavior outcomes across development. Heightened activity level following the neonatal period is associated with desirable behavioral attributes such as positive social interactions, motor and mental maturity, inquisitiveness

(Rapport et al., 2006), and better developed behavioral and inhibitory control (Campbell et al., 2002). This positive association rapidly reverses itself during the preschool and early elementary school years, at which time children are required to regulate their gross motor activity while interacting with others and in accordance with classroom and cognitive demands (Eaton et al., 2001).

Parental reports of difficulty regulating gross motor activity beyond age four predicts an ADHD clinical diagnosis at age nine (Campbell & Ewing, 1990), and heightened gross motor activity after age five is associated with undesirable characteristics and outcomes. For example, objective observations of gross motor behavior predict observed classroom off-task behavior (Abikoff et al., 2002), parental ratings of ADHD symptoms (Ebenegger et al., 2012), and more variable cognitive test performance (Teicher, Ito, Glod, & Barber, 1996). In addition, parental report of hyperactive behaviors is associated with aggression and oppositional behavior (Waschbusch, 2002), peer difficulties (Diamantopoulou et al., 2007) and parent relationship problems (Wymbs et al., 2008) for children with ADHD. These difficulties continue into middle childhood for most children with ADHD, and set the stage for a lifetime of functional impairments despite the diminution in subjective reports – but not objective measures (Halperin et al., 2008) – of excess gross motor activity during adolescence and young adulthood for many individuals with ADHD (Biederman et al., 2000).

The excess gross motor activity exhibited by children with ADHD has been subjected to considerable empirical scrutiny for nearly a half a century using a broad range of methodologies and expanding number of innovative technologies. Early approaches relied on rating scales (Werry, 1968), direct observations (Abikoff & Gittelman, 1984; Whalen et al., 1978), and floor grid-crossing counts (Milich et al., 1982), and have been followed by technologically more sophisticated measures such as pedometers (Plomin & Foch, 1981), stabilimetric cushions (Conners & Kronsberg, 1985), infrared motion analysis (Teicher et al., 1996), and actigraphs (Halperin et al., 1992). Collectively, these and more recent studies uniformly report significantly more frequent and intense gross motor activity in children with ADHD relative to typically developing children at home (Porrino et al., 1983), in school (Imeraj et al., 2011), while asleep (Cortese et al., 2009), and while completing a diverse range of laboratory and clinical tasks (Dane et al., 2000; Rapport et al., 2009) regardless of the technology employed.

Theoretical accounts of the excess gross motor activity in ADHD and its association with task performance and attention have varied considerably over the years. For example, the *behavioral inhibition* model describes hyperactivity as ubiquitous, non-goal oriented motor movement that reflects the outcome of ADHD children's difficulty inhibiting task-irrelevant behavior and regulating goal-directed behavior (Barkley, 1997). Evidence for a relation between inhibition and hyperactivity appears mixed, however. For example, most (Nigg, 1999) but not all (Brocki et al., 2010) studies fail to find significant correlations between inhibition tasks performance and hyperactivity/impulsivity ratings. Experimental evidence also indicates that increasing inhibition demands fails to increase actigraph-measured activity level (Alderson et al., 2012). In contrast, the more recent *subcortical deficit model* describes hyperactivity as a manifestation of early occurring and static subcortical impairment rather than an outcome or correlate of underdeveloped executive functions such as inhibition or working memory (Halperin & Schulz, 2006; Halperin et al., 2008). It specifically proposes a developmental, but not cross-sectional relation wherein maturation of executive functions such as WM facilitates longitudinal recovery from ADHD symptoms. Support for this model includes evidence that developmental changes in neuropsychological test performance, but not baseline scores, predict longitudinal changes in ADHD symptom severity (Rajendran et al., 2013).

Collectively, both ADHD models predict relatively stable and high activity level for children with ADHD that is either unrelated cross-sectionally (subcortical impairment) or negatively related (inhibition) to ADHD-related deficits on tests of executive functions such as working memory. In contrast, only one contemporary model hypothesizes that the higher rates of gross motor behavior observed in children with ADHD are functional (Rapport et al., 2009). In the *functional working memory* model, hyperactivity in ADHD is hypothesized to serve one of two primary purposes: (a) augmenting ADHD children's well-documented prefrontal cortical hypoactivation while engaged in academic (Mann, Lubar, Zimmerman, Miller, & Muenchen, 1992) and cognitive (Dickstein et al., 2002) activities that place demands on working memory; and, in a more limited number of situations, (b) terminating the perceived aversiveness of cognitively demanding activities via escape or avoidance behavior (Rapport et al., 2009). This model therefore predicts a positive relation between gross motor activity and performance within the context of task engagement (attentive behavior).

The diverse predictions stemming from these three ADHD models were investigated in a recent experimental study that used actigraphs to objectively record the intensity of children's gross motor activity while they completed tasks with minimal or high working memory demands (Rapport et al., 2009). Children with ADHD were significantly more active than typically developing children, and both groups exhibited significantly higher intensity movement during high working memory relative to minimal working memory conditions; however, increasing set size demands (i.e., short-term memory) did not significantly impact activity level for either group. Despite the experimental methodology and use of high precision actigraphs to measure the intensity of children's gross motor activity, the study was unable to test directly the extent to which gross motor activity may facilitate, impair, or fail to influence working memory performance as predicted by the working memory, inhibition, and subcortical impairment models, respectively.

Understanding the complex interplay among these constructs is particularly critical for clinical/school psychologists and other mental health professionals charged with designing, implementing, and monitoring psychosocial treatments for children with ADHD. For example, empirically supported psychosocial interventions for ADHD include a wide range of contingencies to address the disruptive behavior and functional impairments exhibited by these children in the classroom, including specific consequences for reducing gross motor activity that interferes with classroom functioning (e.g., getting out of seat; Barkley, 2002; Wells et al., 2000). Other behavioral interventions have used a more direct approach, such as having children wear actigraphs that emit visual and vibratory feedback whenever gross motor activity exceeds a pre-determined threshold, with positive contingencies administered for reduced movement (Tryon et al., 2006).

The indirect or direct targeting of children's activity level with behavioral interventions reflects the assumption that excess gross motor activity interferes with children's ability to actively engage in, and complete, learning-related activities such as classroom assignments and homework. This perspective is supported by the strong covariation between actigraph-measured activity level and academic achievement (Reichenbach, Halperin, Sharma, & Newcorn, 1992) and variable cognitive test performance (Teicher et al., 1996), combined with the replicated association between hyperactivity and inattention based on both subjective ratings (Willcutt et al., 2012) and objective classroom observations (Abikoff et al., 2002). Such recommendations to target hyperactivity may be contraindicated, however, if increasing motor activity serves a positive function for children with ADHD—that is, increasing cortical arousal and facilitating alertness (Zentall & Zentall, 1983) during engagement with learning-related activities that routinely challenge executive functions such as working memory (Rapport et al., 2009). As such, it is critical to consider both inattention and activity level when investigating the relation between gross motor movement and task performance to ensure that any impact of activity level is not better accounted for by inattentive behavior.

The present study is the first to address this pivotal issue by providing an initial examination of the extent to which naturally occurring variations in children's gross motor movement are associated with changes in working memory performance and attentive behavior for children with ADHD relative to typically developing children. To accomplish this goal, children's gross motor movement and attentive behavior were quantified using objective, reliable, and continuous observations while children completed a counterbalanced series of working memory tasks across four testing days in a controlled setting shown previously to evoke attentive behavior rates similar to rates observed in regular classroom settings (Kofler et al., 2010). Children with ADHD were hypothesized to exhibit higher rates of gross motor activity and lower rates of attentive behavior relative to typically developing (TD) children across all conditions consistent with previous studies (Abikoff et al., 2002; Dane et al., 2000). Finding that variations in activity level are positively associated with working memory performance for children with ADHD would be consistent with working memory model predictions regarding the functional role of hyperactivity in facilitating neurocognitive functioning for children with ADHD. In contrast, a negative association would be consistent with models describing hyperactivity as a core deficit or ubiquitous feature of the disorder (Barkley, 1997), while no association would be consistent with models predicting that no cross-sectional relation between ADHD symptoms and underdeveloped executive functions such as working memory (Halperin & Schultz, 2006). As a final step, we sought to characterize individual patterns in children's cognitive performance correlates of gross motor activity. This examination was rooted in the well-documented heterogeneity among children with ADHD with regard to behavioral symptoms and neurocognitive functioning, as well as the potential for differential responses to arousal in individual children (Fair, 2012; Pelham et al., 2011). The extent to which activity level was associated with working memory performance for individual subgroups of ADHD and TD children was accomplished by applying the Jacobson & Truax (1991) model of reliable change to individual differences in this relation.

Method

Participants

The sample included 52 boys aged 8 to 12 years recruited by or referred to a children's learning clinic through community resources (e.g., pediatricians, community mental health clinics, school system personnel, and self-referral). The clinic is a research-practitioner training center 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. Sample ethnicity was mixed and included 34 White non-Hispanic (65%), 12 Hispanic English-speaking (23%), 2 African American (4%), and 4 children of mixed racial/ethnic background (8%). 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 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 inter-rater agreement of .93 to 1.00, test-retest reliability of .63 to 1.00, and concurrent (criterion) validity between the K-SADS and psychometrically established parent rating scales (Kaufman et al., 1997).

Twenty-nine children met the following criteria and were included in the ADHD-Combined Type group[1]: (1) an independent diagnosis by the directing clinical psychologist using DSM-IV criteria for ADHD-Combined Type based on K-SADS interview with parent and child which assesses symptom presence, onset, severity, and impairment 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 & Rescorla, 2001) or exceeding the criterion score for the parent version of the ADHD-Combined subtype subscale of the Child Symptom Inventory (CSI; Gadow, Sprafkin, Salisbury, Schneider, & Loney, 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 & 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, and their psychometric properties are well established (Rapport, Kofler, Alderson, & Raiker, 2008). All children in the ADHD group met criteria for ADHD-Combined Type, and 8 (28%) met criteria for Oppositional Defiant Disorder (ODD).

Twenty-three 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

[1] A review of individual children's records indicated that all children in the ADHD Combined-Type group would meet current DSM-5 diagnostic criteria for ADHD-Combined Presentation.

CSI subscales. Typically developing children were recruited through contact with neighborhood and community schools, family friends of referred children, and other community resources.

Children presenting 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. Ten children with ADHD had been prescribed psychostimulants previously; eight of these children were prescribed psychostimulants at the time of assessment, which were withheld for a minimum of 24 hours prior to participating in all assessment sessions.

Procedures

All children participated in four consecutive Saturday assessment sessions at one-week intervals following baseline diagnostic evaluation, psychoeducational assessment, and group assignment. The phonological working memory tasks were administered as part of a larger battery of neurocognitive tasks that required the child's presence for approximately 2.5 hours per session. Tasks were counterbalanced within and across the four testing sessions, one task per week, to minimize order, practice, and fatigue effects. Performance was monitored at all times by the examiner, who was stationed just out of the child's view to provide a structured setting while minimizing performance improvements associated with examiner demand characteristics (Gomez & Sanson, 1994). All children received brief (2-3 m) breaks following each task, and preset longer (10-15 m) breaks after every 2-3 tasks to minimize fatigue.

Measures

Phonological working memory. The phonological working memory task was programmed in Superlab Pro 2.0 (2002) and is similar to the Letter-Number Sequencing subtest on the WISC-IV (Wechsler, 2003). The task assesses phonological working memory based on Baddeley's (2007) model by requiring both central executive processing/manipulation and phonological storage-rehearsal. 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 inter-trial stimulus 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. 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). These tasks were administered as part of an ongoing series of studies investigating neurocognitive processes in ADHD[2].

Each child was administered the phonological task at four different cognitive loads (i.e., phonological set sizes consisting of 3, 4, 5, and 6 stimuli) across the four testing sessions. The four working memory set size conditions each contain 24 unique trials of the same stimulus set size, and were counterbalanced across the four testing sessions to control for order effects and potential proactive interference effects across set size conditions (Conway et al., 2005). Five practice trials were administered before each task and all children were required to achieve 80% accuracy before advancing to the full task (for additional details see Rapport et al., 2008). Evidence for reliability and validity of the four working memory tasks includes high internal consistency ($\alpha = .82$), and demonstration of the expected magnitude of relations (Swanson & Kim, 2007) with an established measure of short-term memory (WISC-III or -IV Digit Span raw scores: $r = .36$ to $.58$). Two trained research assistants, blind to diagnostic status and seated out of the child's view, independently recorded children's verbal responses. Inter-rater reliability was 96.3%; discrepancies were handled

[2] Performance and attention data for a subset of the current sample were used in separate studies to evaluate conceptually unrelated hypotheses (REFS removed for blind review). We have not previously reported behavioral observations of gross motor activity for any children in the current sample. Additionally, we elected to concentrate on phonological rather than visuospatial working memory given that ADHD-related effect size differences between these two systems are nearly identical (Kasper et al., 2012), and due to previous findings of greater activity level during phonological relative to visuospatial tasks (Rapport et al., 2008).

via audio-video review.

Performance data was calculated using partial-credit unit scoring (proportion of stimuli correct per trial) due to its preferred psychometric properties relative to all-or-nothing scoring (Conway et al., 2005) and control for differences in the number of stimuli available for recall across the set sizes.[3]

Measured Intelligence. All children were administered the Wechsler Intelligence Scale for Children third or fourth edition to obtain an overall estimate of intellectual functioning based on each child's estimated Full Scale IQ (FSIQ; Wechsler, 1991; 2003).

Behavioral Codes

A ceiling-mounted digital video camera was used to record children's gross motor activity and attentive behavior while they completed each of the tasks described above. The camera was situated such that children's full bodies were visible without obstruction at all times. Two trained observers, blind to children's diagnostic status, independently coded all behaviors using Observer XT 10.5 (Noldus Information Technology, 2011) behavioral observation software. Interrater reliability was assessed for all children across all tasks and observed behavior categories. Observer pairs were required to obtain ≥ 80% agreement. For observations with < 80% agreement, observer pairs met to review individual discrepancies and then independently re-coded the video recordings until ≥ 80% agreement was achieved. Overall interrater percent agreement was 97.6%, and agreement across all experimental conditions for all movement and attentive behavior codes was high (all codes ≥ 97% agreement). Disagreements were resolved using the identical method described above, when needed. A continuous observation method with partial interval behavioral definitions was used to most closely match the approach used in previous ADHD classroom observation studies (Kofler et al.,

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[3] Partial-credit methods count each stimulus on a trial as correct if it is emitted in the correct serial location. It differs from all-or-nothing scoring approaches that count trials correct only if *all* stimuli in a trial are emitted in the correct sequence. Partial-credit scoring is associated with significantly higher internal consistency and concurrent validity relative to all-or-nothing scoring (Conway et al., 2005).

2008). Children were observed continuously, and behavioral states were changed (e.g., from visually oriented to not oriented) whenever the new behavioral state was present for $\geq$ 2 consecutive seconds. Behavioral observations were selected to maximize ecological validity by providing an index of children's gross motor and inattentive behavior frequency that more closely matches the frequency-based metric on commonly-used parent/teacher questionnaires (e.g., the CSI-IV queries each hyperactivity symptom frequency as occurring between *never* to *almost always*; Gadow et al., 2004).

Gross Motor Activity

Chair movement. Chair movement was coded as a continuous variable to quantify gross motor movement. Children were seated in a caster-wheel swivel chair approximately 0.66 meters from the computer monitor for all tasks. Three mutually exclusive states of chair movement were coded based on in-chair movement occurring relative to imagined stationary axes extending from the child to the computer monitor and 90 degrees left and right of the child: (a) *Swinging* included all chair movements that crossed fewer than three fixed axes in a continuous motion (i.e., $\leq$ 180 degrees); (b) *Spinning* included movements that crossed three or more fixed axes in a continuous motion (i.e., > 180 degrees); and (c) *Auxiliary chair movement* included all other forms of chair movements while seated exclusive of those described above (e.g., rolling the chair forward/backwards while seated).

Out of seat movement. Out of seat movement was coded any time that a child's movement resulted in his buttocks and/or both knees losing physical contact with the chair seat, mutually exclusive of the chair movement code described above.[4]

Foot movement. Foot movement was coded as a continuous variable independent of attention and the other movement codes to further quantify gross motor activity; the camera and computer

[4] The overall mean duration of out of seat behavior was computed as a validity check to ensure that the out-of-seat code did not misidentify stationary standing behavior (i.e., standing still) as being motorically active. This analysis revealed that out-of-seat behavior was relatively brief for children with ADHD (*M* = 2.7 to 8.6 s) and TD children (*M* = 0.1 to 7.4 s), indicating that misidentification was extremely unlikely.

table were arranged such that children's feet and legs were continuously visible. Foot movement included all observable episodes of foot and ankle movement (e.g., foot tapping, fidgeting, foot/feet swinging) that occurred for ≥ 2 consecutive seconds. During active chair movement, foot/ankle movements were coded only when the child's foot/ankle movement was in excess of the kinetic movement associated with the moving chair (e.g., wriggling feet while spinning in the chair). This procedure was followed to preserve the distinction between chair and foot movement codes.

Movement frequency, defined as the proportion (%) of the total task duration during which each child displayed one or more of the gross motor movements defined above, served as the primary index of activity level for each task condition. The mean activity frequency score across the four conditions exhibited the expected magnitude relation with parent/teacher CSI ratings of hyperactivity/impulsivity symptoms ($r = .30$, $p < .05$), suggesting that movement exhibited during the lab sessions was similar to that observed in the classroom/at home over the past month.

Attentive Behavior

Visual attention to task. Visual attention to task was coded as a continuous variable to quantify children's attentive behavior during each of the experimental task conditions. Observers coded behavior into one of two mutually exclusive states. Children were coded as *oriented* to task (i.e., attentive) when their head was directed within 45° vertically/horizontally of the center of the computer monitor on which the task is displayed. Children were coded as *not oriented* when their head direction exceeded a 45° vertical/horizontal tilt away from the center of the monitor for greater than two consecutive seconds during the tasks. The oriented and not oriented codes used in the present study are analogous to on- and off-task definitions used in most laboratory and classroom observation studies (Kofler et al., 2008). Attentive behavior was coded independently of gross motor movement, such that a child could be attentive or inattentive while moving or not moving.

Percent oriented, defined as the proportion (%) of the total task duration during which each child was visually attentive, served as the primary index of attention for each task condition. The mean attention frequency across the four conditions was related significantly to parent/teacher CSI inattention symptom ratings ($r = .55$, $p < .001$), and the mean attentive behavior frequency (~75%) for children with ADHD was highly similar to the observed attentive behavior frequency in classroom settings estimated via meta-analysis (Kofler et al., 2008), suggesting that our laboratory setting and tasks evoked the expected levels of attentive behavior observed in classroom settings and similar levels of attentive behavior reported by parents/teachers.

Data Analytic Approach

Three unique data points were collected concurrently for each child during each of the four phonological working memory task conditions: (a) *Activity Level* (percent of task engaged in at least one of the gross motor behaviors described above) to index the frequency of children's gross motor activity; (b) *Working Memory Performance* (percent of stimuli correct per trial across each working memory condition) to index phonological working memory functioning; and (c) *Attention* (percent of time visually oriented to task) to index children's attention or on-task behavior.

To investigate the role of activity level on working memory performance and attention, the four working memory task conditions were ordered sequentially for each child individually based on their objectively observed, naturally occurring activity level (from least active to most active; Activity conditions 1-4). Thus, the Activity 1-4 conditions shown in Figures 1 and 2 reflect the ascending order of independent conditions during which each child was least active (Activity 1) to most active (Activity 4) across the four working memory tasks.

Statistical approach. The study's two central aims were to (a) investigate the role of activity level (gross motor movement) on working memory performance and attention (Tier I analyses), and (b) examine the intra-individual heterogeneity in children's cognitive correlates of activity level (Tier

II analyses).

For Tier I, repeated measures ANOVAs were conducted for the performance and attention variables separately using SAS 9.4 PROC MIXED. A mixed model approach was used to account for the correlated data structure that included the nesting of multiple measurements within children. For these analyses, subjects were included as random effects, and the effects of activity level and diagnosis were treated as fixed effects. A lag-1 autoregressive covariance assumption was modeled to allow for within-subject correlation. Because there appeared to be some departure of homogeneity of variance over time, this assumption was relaxed and time-dependent variances were assumed. A restricted maximum likelihood (REML) algorithm was employed to obtain estimates of the effect of diagnostic group (ADHD, TD), activity level, and the group by activity level interaction. Following any significant interaction effects, the simple effects were estimated using contrast statements that used the power of the mixed model but considered differences at each activity level condition. Bonferroni corrections were applied to post-hoc contrasts to adjust for multiple comparisons.

For Tier II, we examined intra-individual heterogeneity using the Jacobson and Truax (1991) Reliable Change Index (RCI). Specifically, each child was classified as Improved, No Change, or Deteriorated based on whether their working memory performance during their most active condition was reliably different than their working memory performance during their least active condition (i.e., difference exceeded chance). Each child's RCI was computed as the ratio of the difference in performance between these two test scores divided by standard error (computed using the measure's test-retest reliability and the *SD* of the TD control group; Rule B; Jacobson & Truax, 1991). Test-retest reliability for these tasks was reported as .76 to .90 (Kofler et al., 2010); we conservatively selected the lower bound of this range (.76) to compute RCI.

Results

Preliminary Analyses

All variables were screened for univariate/multivariate outliers and tested against $p < .001$. Activity level for one child with ADHD at the highest activity condition was replaced with a value equal to one percentage point greater than the next most extreme score for the ADHD group as recommended (Tabachnick & Fidell, 2007). No other univariate or multivariate outliers were identified. Partial observational data was available for four children with ADHD due to video malfunction (unavailable data included 1 child's set size 3 condition, 1 child's set size 4 condition, and 2 children's set size 6 condition). Missing data represented 0.08% of available data points and were replaced with the ADHD group mean for the specific set size in which the video failed to record based on recommendations (Tabachnick & Fidell, 2007). The results and interpretation of analyses presented below were unchanged when including or excluding these four cases. Multicollinearity diagnostics were within recommended limits (all VIF values < 10).

All parent and teacher behavior rating scale scores were significantly higher for the ADHD group relative to the TD group as expected (Table 1). Children with ADHD and TD children did not differ significantly on Hollingshead (1975) SES scores ($p = .19$) or FSIQ ($p = .24$); however, children with ADHD were younger ($p < .01$) and required somewhat longer intervals to complete each WM task ($p < .05$) than TD children. Age and task duration were not significant covariates for any of the analyses presented below (all p values $\geq .13$). Therefore, the simple model results are reported with no covariates.

Data integrity checks. Two series of tests were conducted to assess the integrity of ordering each child's task conditions according to ascending activity level. First, we conducted a 2 (Group) x 4 (Activity Conditions 1-4) mixed-model ANOVA on the activity level data to confirm that children's activity level increased across the four ordered conditions. Results are shown in Figure 1 and revealed a significant main effect of activity condition ($p < .001$), with post-hocs indicating significant differences in activity level between all conditions (all contrasts $p < .0005$). Children with ADHD were more active than TD children across conditions ($p = .01$); however, group differences were similar across the four ordered activity level conditions (group x condition interaction $p = .81$).

Second, we examined set size and administration order as potential confounds. Results indicated that the ADHD and TD groups did not differ systematically in the working memory set size conditions associated with each activity level condition (all χ^2 $p \geq .18$), and no set size was overrepresented in any activity level condition (all χ^2 $p \geq .23$). These analyses indicate that manipulating short-term memory load (i.e., storage/rehearsal demands) within the context of a phonological working memory task was not significantly related to activity level as expected (Rapport et al., 2009). Coupled with the counterbalancing procedures described above, these findings suggest that any changes in attention or performance across the activity conditions are associated with variation in activity level rather than set size, practice, fatigue, or order effects.

Tier I: The Role of Activity Level in Working Memory Performance and Task Attention

Phonological working memory performance. The mixed-model ANOVA examining the association between children's activity level and phonological working memory performance revealed significant main effects for group ($p < .0001$), activity level ($p = .018$), and a significant group by activity level interaction ($p < .001$). Post hoc contrasts for the interaction revealed large magnitude between-group differences in phonological WM performance under the two lowest activity level conditions (Cohen's $d = 1.28$ to 1.44, both $p < .0001$) that were no longer detectable under the two highest activity level conditions ($d = 0.52$ to 0.42, $p = .06$ to .21) due to the divergent pattern of performance changes between the two groups (Figure 2).

For TD children, variation in naturally occurring activity level significantly predicted working memory performance (p = .04). Post hocs revealed that performance was not significantly different among most activity level set sizes (all p = .07 to .59), with one exception (Activity3 performance < Activity1 performance; p = .008). Trend analysis revealed a significant, linear pattern of declining performance associated with greater levels of activity for the TD group (F[1,22] = 6.84, p = .016, η_p^2 = .24), such that their mean performance was 0.55 *SD* units worse during their highest relative to lowest activity conditions (Cohen's d = -0.55).

For children with ADHD, variation in naturally occurring activity level also predicted working memory performance (p = .002), but in the opposite direction. In contrast to TD children, phonological WM performance among children with ADHD was significantly greater under the most active condition relative to the three lower activity level conditions (all p = .0001 to .03), which were not significantly different from each other (all p = .07 to .82). Trend analysis indicated an overall, positive linear relation between performance and activity level (F[1,28] = 5.75, p = .02, η_p^2 = .17), and to a lesser extent a quadratic effect (F[1,28] = 10.03, p = .004; $\Delta\ \eta_p^2$ = .09), wherein children with ADHD performed similarly to themselves under the lowest activity level conditions followed by a 41% increase in their performance under their highest activity level condition. The magnitude of performance differences between the least and most active conditions for children with ADHD was 0.59 *SD* units, which is nearly identical to the value obtained for the TD children, but in the opposite direction. Results are depicted in Table 2 and Figure 2a.

Task attention. The mixed-model ANOVA examining the association between children's activity level and attention revealed a significant main effect for group (p < .0001), wherein children with ADHD displayed less overall attentive behavior than TD children. However, the main effect for activity level indicated that children's attention did not change significantly with variations in activity level (p = .19), and the group by activity level interaction was also non-significant (p = .53), indicating that group differences in attention did not vary significantly as a function of variations in children's activity level.

Tier II: Heterogeneity in ADHD: Intra-individual activity level by performance patterns

Results revealed that the ADHD and TD groups differed significantly in their representation among the three RCI categories (χ^2 [2] = 9.76, *p* = .008). Specifically, children with ADHD were significantly more likely to exhibit reliably better phonological working memory performance when they were most active (48.3% Improved; $p < .05$) whereas significantly fewer TD children displayed reliable improvements (8.7% Improved; $p < .05$). In contrast, TD children were more than twice as likely as children with ADHD to be categorized as Deteriorated (ADHD = 17.2%, TD = 39.1%), although this difference did not reach statistical significance ($p = .13$). A similar proportion of children from both groups displayed no change (ADHD = 34.5%, TD = 52%; $p = .32$).

Exploratory analyses revealed no significant differences between the three ADHD subgroups or two TD groups [5], respectively, in terms of FSIQ, SES, age, ADHD/ODD symptoms, overall activity level, attention, or phonological working memory functioning (all p = .06 to .96).

Discussion

This is the first study to test competing model predictions regarding the complex interplay among activity level, working memory, and attention in children with ADHD and typically developing (TD) children. Children's accuracy and visual attention-to-task was examined during four phonological working memory tasks that were administered in counterbalanced order across four testing days and then sequenced hierarchically based on objective observations of each child's naturally occurring gross motor activity during each task. Sequencing the tasks in an ascending manner enabled us to investigate the extent to which intraindividual differences in gross motor

[5] The TD Improved subgroup was omitted from these analyses due to insufficient cell size; only two TD children were classified as Improved.

activity were associated with concurrent changes in attentive behavior and cognitive functioning, and to determine whether these relations were analogous for both groups of children. Overall, children with ADHD exhibited higher rates of gross motor activity and lower rates of attentive behavior relative to TD children under all conditions. These findings are consistent with a robust literature documenting ADHD-related hyperactivity and impaired attention across a diverse range of settings, contexts, demands, and measurement technologies (Abikoff et al., 2002; Imeraj et al., 2011).

The analysis of children's naturally occurring gross motor activity revealed an opposing pattern of relations with working memory performance between the two groups. Overall, higher rates of gross motor activity positively predicted phonological working memory performance for children with ADHD (Δd = 0.59) but not for TD children (Δd = -0.55). This finding was particularly noteworthy due to the magnitude of performance differences and accompanying stability of attention for children with ADHD. Specifically, the positive association with working memory performance exhibited by children with ADHD was sufficiently robust to preclude detection of their working memory deficits relative to TD children under the two highest activity level conditions (i.e., Δd from -1.28 and -1.43 to -0.52 and -0.36). Importantly, this diminution in between-group differences as a function of higher activity level was not attributable to set size or administration order, and occurred in the absence of concomitant changes in task-oriented attention. Juxtaposing each group's optimal cognitive performance based on activity level revealed large magnitude, albeit attenuated, between-group differences (d = -0.88; Figure 2a), suggesting that higher rates of naturally occurring gross motor activity may at best be associated with improvements in, but not normalization of, phonological working memory functioning for children with ADHD.

Collectively, the magnitude of ADHD children's phonological working memory deficits was consistent with those reported in experimental investigations (Bolden et al., 2012; Rapport, Alderson et al., 2008) and meta-analytic reviews (Kasper et al., 2012) and extends this literature by demonstrating that working memory impairment may vary considerably as a function of gross motor activity. The positive, linear relation between gross motor activity and working memory performance observed in children with ADHD, however, was inconsistent with extant theoretical models that describe hyperactivity as omnipresent (Barkley, 1997; Porrino et al., 1983) or unrelated cross-sectionally to setting-specific cognitive demands (Halperin et al., 2008; Porrino et al., 1983).

The current results suggest a positive link between hyperactivity and task performance for children with ADHD. An examination of individual differences in the pattern of activity-related working memory improvements revealed that this group-level effect was driven by approximately 50% of children with ADHD performing reliably better when most motorically active, relative to 17% who performed reliably worse when most active. Interestingly, this finding converges with other evidence suggesting that external stimulation may lead to optimal arousal and performance improvement in a similar proportion of children with ADHD (Pelham et al., 2011). Taken together, this finding is consistent with the well-documented neurocognitive and behavioral heterogeneity in ADHD and TD populations (Fair et al., 2012), and implies that gross motor movement may be more likely to facilitate than impair cognitive functioning for a large proportion of children with ADHD.

A parsimonious explanation for these findings is suggested by the neuroimaging and cognitive science literatures. Comprehensive meta-analytic reviews (Dickstein et al., 2006) and experimental investigations are highly consistent in documenting widely distributed hypoactivity in frontal/prefrontal cortical regions in children with ADHD while engaged in academic (Mann et al., 1992) and cognitive (Dickstein et al., 2006; El-Sayed et al., 2002) activities that place demands on working memory and other executive functions. From this perspective, excess gross motor activity exhibited by children with ADHD while engaged in cognitively demanding tasks may reflect central nervous system (CNS) arousal-regulating compensatory behavior (Rapport et al., 2009). That is, children with ADHD may up-regulate their gross motor activity as compensatory behavior to augment CNS arousal during tasks that challenge their underdeveloped neurocognitive functions and/or require sustained neural activity. Observational and actigraph study results are congruent with this explanation. Elevated rates of gross motor activity in children with ADHD are uniformly observed during academic seat-work activities (Abikoff et al., 2002) and challenging executive function tasks (Alderson et al., 2012), whereas their activity level is more similar to that of non-ADHD children during activities with minimal working memory demands such as lunch, recess, physical education (Porrino et al., 1983), and computer drawing (Rapport et al., 2009). No study to date, however, has experimentally manipulated activity level while concomitantly assessing physiological arousal to directly examine this hypothesized mechanism linking increased activity level with improved cognitive performance for children with ADHD. Physiological measurement appears promising for understanding why children with ADHD in the current study exhibited intra-individual, day-to-day differences in their observed activity level despite relatively stable central executive working memory task demands across sessions. In particular, recent research indicates within and between day fluctuations in physiological arousal for children with ADHD (Imeraj et al., 2011) that would be expected to elicit different levels of compensatory gross motor movement to the extent that movement facilitates arousal to augment task performance (Rapport et al., 2009).

When juxtaposed with the pattern of results for children with ADHD, the typically developing group’s pattern of modestly worse performance when most active was puzzling, and appeared inconsistent with studies suggesting potential benefits of diverse physical movements ranging from gum chewing (Onyper et al., 2011) to doodling (Andrade, 2010). Upon closer inspection, however, this interaction effect appears consistent with the *optimal stimulation* model that explains hyperactivity in terms of the Yerkes-Dodson (1908) ‘inverted U’ relation between arousal and performance (Zentall & Zentall, 1983). That is, cortically underaroused children such as those with ADHD may benefit from increasing cortical arousal via physical movement, whereas similar increases in motor movement may result in over-arousal and adversely impact performance for children without ADHD. By extension, children with underdeveloped neurocognitive abilities – such as children with ADHD (Kasper et al., 2012) – may need to up-regulate their gross motor activity more frequently and in response to lower cognitive demands than TD children to optimize their opportunities for task success (Rapport et al., 2009). This hypothesis is consistent with the current observations, wherein TD children performed optimally when moving 43-57% of the time, and comparatively less well when moving 67-78%. In contrast, children with ADHD performed optimally when moving 81-89% of the time, and comparatively less well when moving 60-72% (i.e., 34% difference concerning activity frequency linked with optimal task performance for both groups).

Limitations

Several caveats merit consideration when considering the present findings, despite methodological refinements including the stringent inclusion criteria and objective observations of activity level, attentive behavior, and working memory performance. Importantly, the current study relied on observations of children’s naturally occurring activity level during counterbalanced tasks administered across four separate testing days. As such, our *a posteriori* sequencing of performance and attentive behavior as a function of ascending activity level was non-experimental, and causal attributions cannot be drawn. Carefully controlled provocation and rarefaction studies, and investigations of movement topography, are needed to corroborate and extend the current findings. In addition, the large magnitude between-group differences may be related to our stringent inclusion criteria for both groups, and may be attenuated to the extent that future studies include a less severe

ADHD sample and/or a clinical comparison group known to exhibit deficits in attention, gross motor activity, and/or working memory. Independent replication with larger samples that include females, older and younger children, and other ADHD subtypes/presentations is needed to address the extent to which our results generalize to the larger ADHD population. It will also be important to examine the extent to which activity level is positively related to performance on other neurocognitive tasks or with alternate indicators of movement, whether these relations are observed when examining movement intensity (e.g., actigraphs) rather than frequency, and more importantly, whether *in situ* movement translates to improved academic attainment in classroom settings.

Clinical and Research Implications

Collectively, the present findings suggest that higher rates of gross motor movement within the context of attentive behavior are associated positively with phonological working memory performance for children with ADHD, whereas this link is somewhat negative for typically developing children. These results suggest a need for increased specificity when defining ‘hyperactive’ behavior, and if confirmed experimentally, call for caution to avoid overcorrecting gross motor movement that may be functional for some children. That is, the current results provide initial support for incorporating devices or techniques into classrooms that accommodate movement while minimizing its disruptive nature (e.g., activity balls, stationary bikes while reading) to the extent that movement facilitates task-relevant arousal necessary to support phonological working memory processes that are critical for completing myriad academic tasks (Sarver et al., 2012). We might speculate also that this positive association may help explain the robust decreases in gross motor activity associated with psychostimulants (van der Oord et al., 2008), such that excess movement is compensatory but less effective than psychostimulants for up-regulating the chronic cortical underarousal associated with ADHD. Importantly, these hypotheses were not tested in the current study and remain highly speculative; direct tests are needed to examine these issues and clarify the role of excess gross motor activity in ADHD.

A critical next step will be assessing the relation between hyperactivity in ADHD and visuospatial working memory given evidence that even minor motor movements such as pointing (Brooks, 1968), arm movement (Lawrence et al., 2001) and finger tapping (Della Sala et al., 1999) may interfere with visuospatial working memory by disrupting the visuospatial system's location-based rehearsal processes (Awh & Jonides, 2001) or interrupting feedback loops between pre-frontally mediated systems responsible for allocating attention and maintenance of spatial information (Chafee & Goldman-Rakic, 2000). This potential duality – wherein increased movement may facilitate phonological but impair visuospatial working memory performance – deserves systematic investigation given its implications for ADHD classroom management. If confirmed, this pattern would suggest that some forms of movement should be reinforced during academic tasks that rely predominantly on the phonological system (e.g., reading and in-seat work) but discouraged during scholastic activities that depend more heavily on the visuospatial system (e.g., math; Sarver et al., 2012). Given that many activities require both phonological and visuospatial processes for optimal performance, however, careful consideration of the intensity, duration, and topography of children's motor movements will be necessary to determine how, and how much, these children can move to optimize the phonological system without detriment to the visuospatial system.

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Table 1. Sample and demographic variables

Variable	ADHD		TD			
	M	*SD*	*M*	*SD*	*F* (1, 50)	*d*
Age	9.22	1.05	10.18	1.31	8.53**	0.81
FSIQ	104.83	12.54	108.83	11.51	1.40	0.33
SES	48.79	11.70	52.91	10.19	1.78	0.37
ADHD Problems						
CBCL	71.86	7.58	54.17	7.54	70.24***	2.30
TRF	66.41	7.28	53.50	4.82	51.88***	2.04
ADHD Symptom Severity						
CSI-Parent	78.14	9.53	49.17	11.48	98.89***	2.76
CSI-Teacher	65.31	14.76	48.73	7.61	23.04***	1.39

Note: ADHD = attention-deficit/hyperactivity disorder; CBCL = Child Behavior Checklist *T*-scores; CSI = Child Symptom Inventory severity *T*-scores; FSIQ = Full Scale Intelligence Quotient; SES = socioeconomic status; TD = typically developing children; TRF = Teacher Report Form.
** $p \leq .01$, *** $p \leq .001$

Table 2. Phonological working memory performance and attentive behavior as a function of diagnostic group and activity level

	Activity Level Condition						
	1	**2**	**3**	**4**	**Group Composite**		Set Size
	M (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SD*)	*M* (*SE*)	*F*	Contrasts
Phonological working memory performance (% stimuli correct) [a]							
ADHD	64.05 (25.37)	54.59 (25.15)	64.70 (23.23)	76.80 (17.29)	65.23 (2.17)	5.71**	4>1=2=3
TD	89.74 (10.05)	85.05 (14.79)	76.99 (19.32)	83.67 (15.23)	83.61 (2.44)	3.00*	3<1=2=4; 3=2; 3=4
Activity Level Composite	75.26 (23.96)	68.11 (25.99)	70.99 (21.32)	79.39 (16.51)	--	3.47*	
Group Composite *F*	20.91***	26.43***	3.65	1.64	30.80***		
Group Contrasts	TD > ADHD	TD > ADHD	TD = ADHD	TD = ADHD	TD > ADHD		
Attentive behavior (% of task oriented) [b]							
ADHD	77.23 (20.12)	73.19 (21.22)	70.77 (21.59)	78.91 (16.41)		--	--
TD	97.31 (4.73)	96.44 (4.81)	90.78 (10.38)	93.29 (7.87)		--	--
Activity Level Composite	86.36 (17.90)	82.62 (20.47)	80.18 (21.80)	79.38 (21.61)	--	2.09	1=2=3=4
Group Composite *F*	--	--	--	--	30.73***		
Group Contrasts	--	--	--	--	TD > ADHD		

Note: Within-group and between-group *F* values/set size contrasts are not presented for attentive behavior due to the non-significant omnibus interaction effect. ADHD = attention-deficit/hyperactivity disorder; SD = standard deviation; SE = standard error; TD = typically developing children.

* $p < .05$; ** $p \leq .01$; *** $p \leq .001$

[a] Phonological performance group x activity level interaction, F (3,150) = 4.54, $p < .005$

[b] Attentive behavior group x activity level interaction, F (3,150) = 1.38, $p = .25$

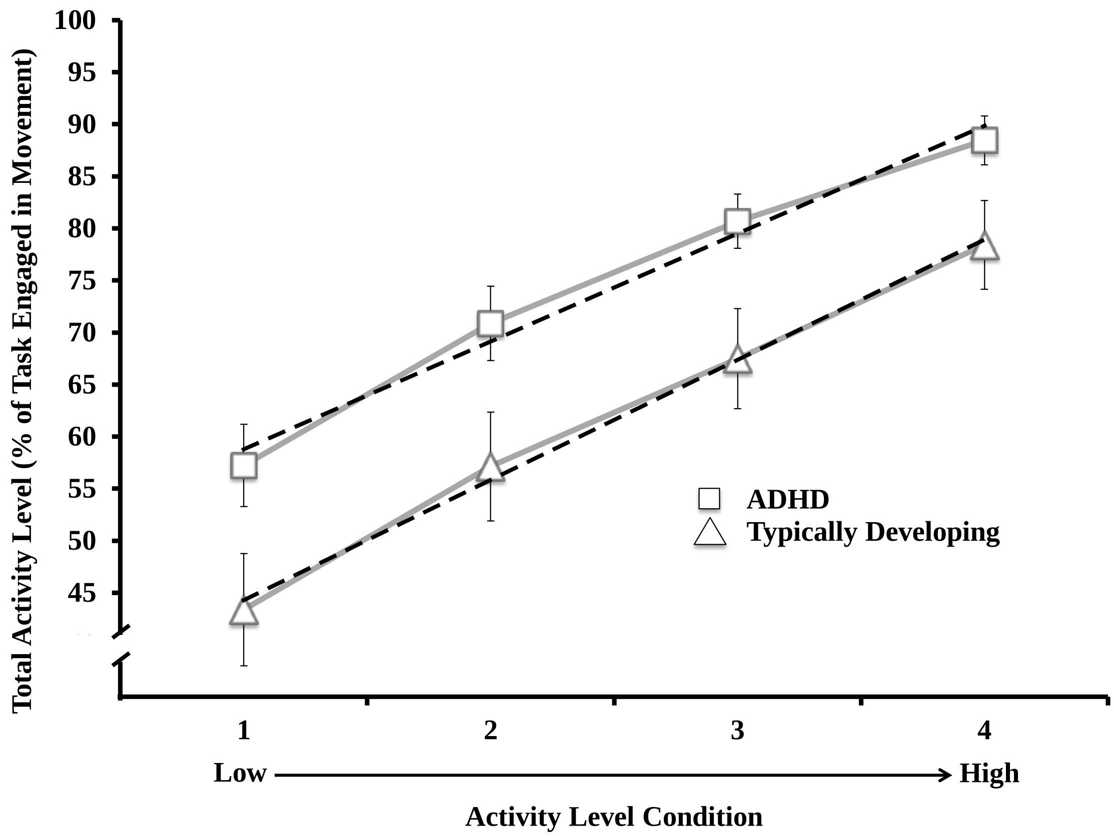

Figure 1. Gross motor activity during the four activity level conditions. Triangles represent typically developing children. Squares represent children with attention-deficit/hyperactivity disorder. Black lines reflect line of best fit. Error bars represent standard error. ADHD = attention-deficit/hyperactivity disorder; TD = typically developing.

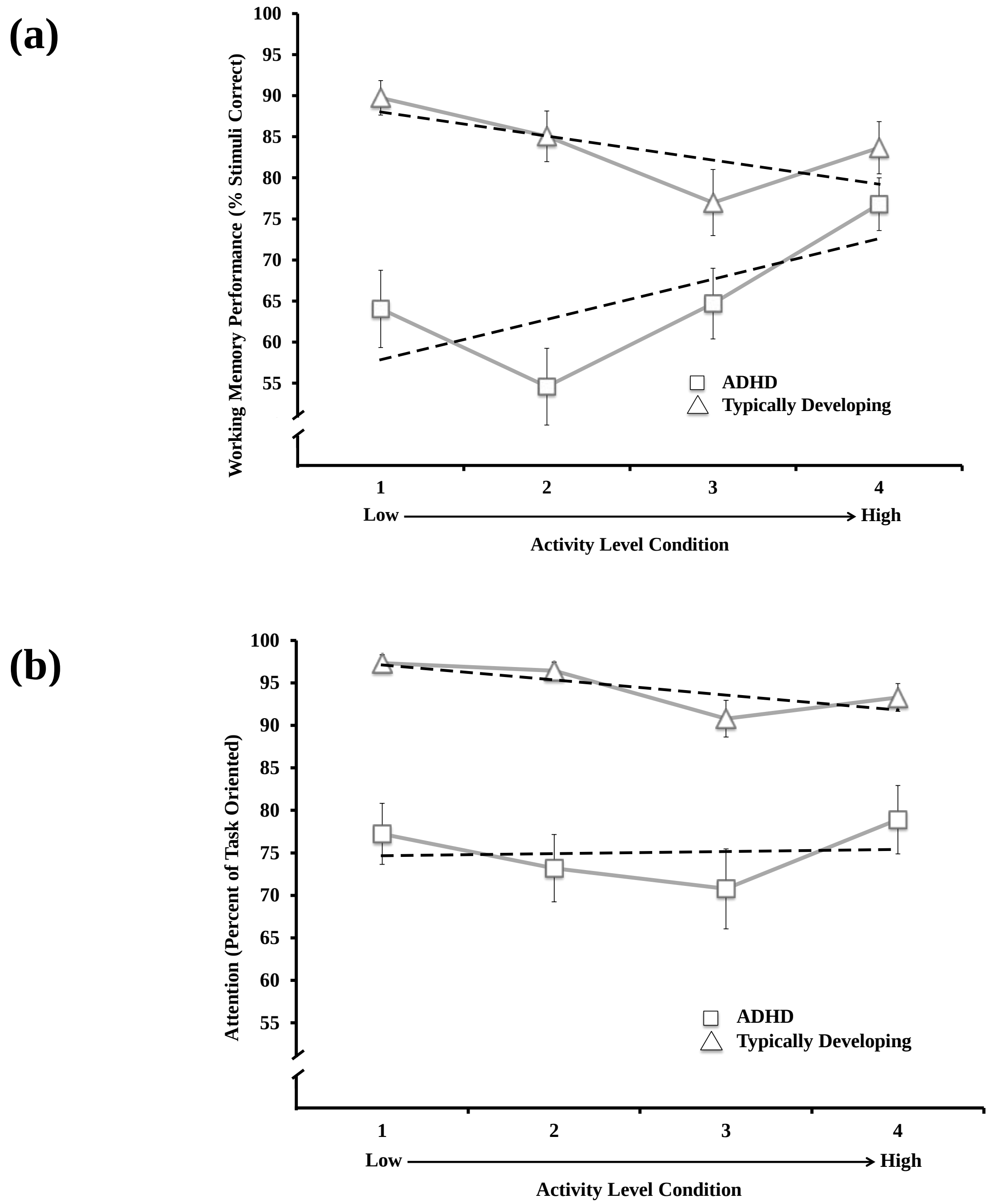

Figure 2. Relationship between naturally occurring differences in activity level and (a) phonological working memory performance and (b) attentive behavior. Black lines reflect line of best fit. Error bars represent standard error. ADHD= attention-deficit/hyperactivity disorder; TD= typically developing.