Beyond the Rubicon: a Continuum Approach to Investigating the Impact of ADHD Like Characteristics on Everyday Executive Function in Children with Tourette Syndrome.

Child Neuropsychology

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Abstract

Across a range of neurodevelopmental conditions, a diagnosis of attention deficit hyperactivity disorder (ADHD) has been shown to be associated with executive function impairment. However, the DSM V emphasis upon viewing psychological characteristics as existing on a continuous distributed quantitative dimension has enabled the opportunity to consider the influence of sub-diagnostic or sub-referral levels of these psychological characteristics upon cognitive function. This study adopted a continuum approach to the consideration of this ADHD influence and examined the extent to which the difference in parental reported executive functions between children with Tourette syndrome (TS) or typically developing children could be mediated by a concurrent group difference in the possession of sub-referral levels of ADHD Like characteristics. A total of 146 children, 58 with reported TS diagnosis participated. Parental report measures of ecological executive functioning, the Child Executive Functioning Inventory, and the Vanderbilt ADHD Diagnostic Parent Rating Scale were employed. The analyses with a full sample and a sub-referral sample revealed significant group differences in most of the key measures, in addition these measures were highly correlated even when controlling for age and gender. A series of mediation analyses indicated that in all models, the ADHD Like measures significantly mediated the group difference in executive function. These results suggest that sub-referrals levels of ADHD Like characteristics continue to contribute to executive challenges in TS. Future intervention research targeting these executive functions should consider the presence of ADHD Like characteristics at sub-referral levels of possession.

Keywords: children; Tourette syndrome; ADHD Like characteristics; executive function; mediation analyses

Introduction

Tourette's Disorder or Tourette Syndrome (TS) diagnosis occurs when motor and vocal tics are exhibited at some period of the illness in the absence of any another medical condition (DSM-5, APA, 2013). Tics are specified as sudden, recurrent non-rhythmic motor actions or vocalizations. The onset of the tics should occur before the age of 18 years and have at least a one-year duration. TS has a prevalence rate of 0.3%-1% in children (Cavanna et al., 2020), with boys more likely to be affected, however, both boys and girls are likely to exhibit similar forms of tics. The impact of the disorder varies from social isolation to a great reluctance to go to school. The co-occurrence of an Attention Deficit Hyperactivity Disorder (ADHD) diagnosis can lead to a greater impact upon cognitive function.

The aim of this current research is to investigate challenges in everyday executive functioning (EF) in children with TS, and in particular by adopting a continuum or dimensional approach, investigate the impact of ADHD at levels of possession which typically would not warrant a referral for a clinical diagnosis (Wolraich et al., 2003). For a number of years, the dimensional approach has been employed to investigate autistic like traits (ALTs, Baron-Cohen et al., 2001; Constantino & Todd, 2003). These ALTs have been shown to be associated with underlying neural structures (Blanken et al., 2017) and more importantly for the current study, the possession of these ALTs at sub clinical levels are associated with challenges for the individual, both in cognition and in mental health (Ishizuka et al., 2022; Moses et al., 2022). It has been presumed that ADHD, much like ALTs, reflect a continuously distributed quantitative trait (Barkley, 2015; Greven et al., 2016; Heidbreder et al., 2015). In the general population, as with the ALTs, sub-referral levels of ADHD characteristics have been associated with challenges to cognition (Kildal et al., 2022) and to mental health (Vogel et al., 2018). In this paper these sub-referral level ADHD characteristics are labelled ADHD Like traits. ADHD Like characteristics in an individual are identified by the individual achieving high scores in the ADHD screening tool (the Vanderbilt ADHD Diagnostic Parent Rating Scale, VADPRS, Wolraich et al., 2003) but having a profile of scores which would not typically lead to a formal referral for further clinical investigation for a diagnosis of ADHD (see Method for further details).

Diamond (2013) referred to EF as a set of top-down cognitive processes important for working memory, concentration, attention, cognitive flexibility and inhibiting inappropriate actions or thoughts. Diamond documented how important EFs were to every-day activities: mental health, scholastic attainment, employment opportunities and public safety. Within a developmental neuropsychology context, EF challenges have been observed across a diverse range of neurodevelopmental conditions, e.g., in ASD, (Geurts et al., 2020); Down syndrome (Will et al., 2021); pediatric stroke (Rivella & Viterbori, 2021) and epilepsy (van den Berget al., 2021). Recent reviews of the TS research have summarised the neuropsychological profile of children with TS and have emphasised the executive challenges the children experience (Cavanna et al., 2020; Morand-Beaulieuet al., 2017). In this research, inhibitory processes were most reliably impaired (Yaniv et al., 2017). However, a major research confound associated with investigating these EF challenges has been the co-occurrence with other clinical conditions, in particular ADHD characteristics in participants with TS.

This association of EF challenges with the diagnosis of ADHD is identified in detail in the DSM V Diagnostic Criteria of ADHD. The behavioural characteristics identified included difficulty in sustaining attention, difficulty in maintaining sequential steps in complex tasks, and challenges in in inhibiting task irrelevant or extraneous stimuli, etc. Earlier, Barkley (1997) had provided a major theoretical account of the relationship between ADHD and EF efficacy. A key element of this theory was the role of inhibition which could influence behaviour in several ways: in cognitive tasks which require withholding of information; delayed responding; the stopping of inappropriate behaviours; and the resistance to external or task irrelevant events and information. Thus, challenges to EF and emotional regulation were important in this account of ADHD. Barkley (2010, 2015) has continued to suggest that challenges to EF and self-regulation are core phenotypic characteristics of individuals with ADHD.

A major challenge to research within a clinical context is the presence of co-occurrence of neuropsychological characteristics, e.g., Pliszka (2015) has suggested that between 67-80% of children and adults with ADHD possessed secondary clinical characteristics. Hirschtritt et al. (2015) suggested that ~57% of the TS population had 2 or more neuropsychological comorbidities. In a study by Wolicki et al. (2019), 27% of the TS sample had 5 or more co-occurring disorders. Rizzo et al. (2014) suggested that in 22% of their sample, TS was comorbid with an ADHD diagnosis. This high incidence of diagnosed ADHD co-occurrence has major implications for EF in children with TS. As discussed above, ADHD characteristics have been associated with challenged EF function (e.g., Barkley, 1997, 2014; DSM V; Feldman & Huang-Pollock, 2021), challenges in working memory and daily living (Irwin et al., 2020) and difficulties in education (DuPaul et al., 2021; Tamm et al., 2021). Research has suggested that these cognitive challenges are also present when ADHD co-occurs with other clinical conditions, e.g., with dyslexia (Czamara et al., 2013) and with ASD (Rosello et al., 2018). Importantly for the present study, Kibby et al. (2021) noted that executive dysfunction was a potential candidate for the core cognitive process in children with ADHD and reading disability. Across many neurodevelopmental research contexts, there have been issues associated with observations of differential or selective EF challenges. For example, some studies showing inhibition challenges, others not observing this. These issues of equivocal patterns of EF impairment may have been the consequence, in part, of comorbid characteristics varying between the samples employed in the studies, e.g., in the sex of the participant, and in part due to the nature of the EF protocol employed. Thus, some studies may have employed a recall retrieval context, others may have used a recognition procedure (Kasper et al.2012; Tarle et al., 2017).

This inconsistency in EF competency as a result of co-occurring characteristics has also been identified in TS research. Openneer et al. (2020) explicitly addressed this question in a sample of children with TS, where the groups were either, TS, ADHD, or possessed TS and (+) ADHD comorbidity. This research employed a range of EF tasks assessing inhibition in a go/no go task, cognitive flexibility or set shifting, and verbal working memory in the form of the forward and backward digit span. In addition, a processing speed measure, a simple reaction time (SRT) task, was employed. A key observation of the study was that the comorbid (TS+ADHD) group of children were relatively impaired in the go/no go inhibition task. There was also a strong, but non-significant effects in set switching accuracy (***d*** = *0.63*) and SRT (***d*** = *0.46*). There was no difference in the verbal working memory task (***d*** = *0.18*). Thus, the influence of comorbidity upon EF efficacy was quite variable, significantly affecting only the conventional go/no go task measure of inhibition (see also Will et al., 2021).

The observation that comorbidity was associated with a differential pattern of EF efficacy provided further evidence of the heterogeneity of EF processes. This diversity in EF competence was explicitly examined by Miyake et al. (2000). Based upon lab-based cognitive task research they emphasised three key functions, mental set shifting, a feature of cognitive flexibility; inhibition of prepotent responses; monitoring and updating of information. They argued that although these functions were related, i.e., they had processes in common, they should also be viewed as discrete EF processes. Subsequent research led Miyake and Friedman (2012) to re-formulate this conceptualisation and emphasise, in a similar manner to Barkley (1997), that inhibitory processes were the common, the core, EF processes. Miyake and Friedman also suggested that although this conceptualisation was derived from lab-based observations these cognitive measures could predict clinical and everyday behaviours. Thus, Young et al. (2009) in a twin study found that behavioural disinhibition found in ADHD and Conduct Disorder was reliably associated with cognitive task measures of response inhibition, working memory updating and task set shifting. It should be noted that the identification and conceptualisation and measurement of the precise inhibitory challenges in ADHD is an ongoing process (see Alderson et al., 2007; Lijffijt et al., 2005). This in part, is likely to be due to the generic challenges in conceptualising inhibitory processes (e.g., Barkley, 1997; Friedman & Miyake, 2004; Oberauer & Lin, 2017; Venables et al., 2018; Yaniv et al., 2017)

The Young et al. (2009) study looked at inhibition within a social context and highlighted the interface between cognitive control and externalising behaviour (Donati et al., 2021). An interface which shows critical development in the adolescent phase of child development (Casey et al, 2019; Kilford et al., 2016). This consideration of EF efficacy within a social and motivational context has been labelled ‘hot EF’ (Welsh & Peterson, 2014; Zelaso & Carlson, 2012) or ‘affective control’ (Schweizer et al., 2020). These constructs relate to the use of EF within contexts which possess motivational or emotional demands, as in more typical everyday contexts. This differentiation of EF into hot and cold characteristics has also been associated with the development of research tools employed to assess EF, in particular measures which attempt to assess EF efficacy within everyday behaviour. Lab-based cognitive tasks looking at performance have been employed to assess ‘cold’ EF efficacy and self or third party-report questionnaires assessing EF within more naturalistic contexts, the ‘hot’ EF context (Gioia et al., 2000). Gioia et al. developed the Behavior Rating Inventory of Executive Function (BRIEF) rating scale for children aged 5-18 in order to directly assess EF in such contexts. Other measures such as the Childhood Executive Functioning Inventory (CHEXI) and the Teenage Executive Functioning Inventory (TEXI) have also been developed to identify EF efficacy in daily life contexts (Thorell et al., 2020; Thorell & Nyberg, 2008; see also Toplak et al., 2013).

Within an ADHD context Hovik et al. (2017) employed the BRIEF measure to contrast EF profiles in different neuropsychological groups: TS, ADHD-C (inattention and hyperactivity combined) ADHD-I (solely inattention), ASD and Typically Developing (TD) children. Children with TS, in contrast with the TD group, demonstrated challenges in Emotional Control, Initiation, Working Memory, and Planning & Organisation. The children also received a parental rating for ADHD characteristics, and it should be noted that the TS group had significantly elevated levels of inattention and hyperactivity/impulsivity in comparison to the TD group. The TS group differed in emotional control and inhibition from the ADHD-C group, showing more problems in emotional control but lower difficulties in inhibition. In contrast with the ADHD -I group of children, the TS group had more issues with Emotional Control, but less with Planning & Organisation. However, it should be noted that the TS group sample was small, and some TS children had comorbid disorders.

The Hovik et al. (2017) and Openneer et al. (2020) studies discussed above have relevance for the rationale of the current study. Both studies were interested in EF efficacy, with the latter using cognitive task measures and the former using a parental report EF measure. The Openneer et al. procedure contrasted the presence or absence of ADHD characteristics in discrete groups of TS with healthy controls and an ADHD group who had TS characteristics. The Hovik et al. study employed a comparison of TS and ADHD groups. Both studies employed parental rating scales for ADHD (which produced a continuous score) and reported these scores for each of their groups. These studies made use of groups of children who had received formal clinical diagnoses, a categorical approach, however, a consideration of group differences in the ADHD continuous measure is also informative. In the Openneer et al. study the TS group without a diagnosis of ADHD still had elevated

ADHD scores in comparison to the healthy control group (~***d*** = *-1.5*). This was also the case in the Hovik et al. (2017) study where both inattention and hyperactivity scores were elevated in the TS Group (~***d*** = *-1.88* and ~***d*** = -2.28 respectively, albeit with a minority of children with TS possessing an ADHD diagnosis) in comparison to the control group. It could be argued that the presence of significantly elevated ADHD characteristics in these two groups acted as a contributory factor to the subsequent patterns of performance in the Openneer et al. tasks and the Hovik et al. EF parental ratings. Within a dimensional or continuum approach, and with the assumption that observations with clinical groups can underpin hypotheses about performance throughout the continuum (Hamilton et al., 2018), one could predict that the possession of high, but sub-referral level of ADHD Like scores will be associated with EF challenges and be a significant source of the difference between the TS and TD groups, even at sub-referral levels of ADHD Like trait possession.

This current study represents an important extension of the Openneer et al. (2020) and the Hovik et al. (2017) studies. These studies confirmed the suggestion that children TS who possessed clinically diagnosed levels of ADHD characteristics had challenged EF efficacy, measured in cognitive task and EF rating contexts respectively. This study adds two elements to these earlier observations, by employing mediation analyses this study can demonstrate *how* the presence of ADHD characteristics can account for TS and TD (typically developing) group differences in EF (Hayes, 2018). Secondly, the identification of high levels of ADHD-like traits and challenged EF efficacy has important implications for mental health (Ameis et al., 2022; Vogel et al., 2018). This would enable this group of children to receive support which could help ameliorate the associated mental health difficulties.

Consequently, the aim of this current research, using an ecological EF measure, the CHEXI (Thorell & Nyberg, 2008), is to contrast EF rating scores in children with TS with a typically developing (TD) group of children whilst explicitly considering the contribution of sub-referral levels of ADHD Like characteristics to any observed group EF differences. The CHEXI has four EF sub-scales: Working Memory, Planning, Regulation, and Inhibition. Working Memory and Planning when combined provide an overall Working memory score. The Regulation and Inhibition scores in this scale when combined provide an overall Inhibition Score. In addition, the Vanderbilt ADHD Diagnostic Parent Rating Scale (VADPRS, Wolraich et al., 2003) was employed to identify individual differences in Inattention and Hyperactivity/Impulsivity characteristics in both groups of children. Wolraich et al. considered this parental tool as an effective means to assist, at an initial phase, the diagnostic process of children with ADHD. Whilst the questionnaire items were initially derived from the DSM IV publication it has been argued to satisfactorily satisfy the DSM 5 criteria in a recent standardisation study (Anderson et al., 2022).

Using mediation analyses (PROCESS, Hayes, 2018) it will be possible to identify the extent to which Inattention, Hyperactivity/Impulsivity, and/or Inattention + Hyperactivity/Impulsivity combined ADHD Like characteristics contribute to the relationships between group status and Working Memory, Planning, Regulation, and Inhibition. This enables an opportunity to look at the contribution of high, but sub-referral levels of ADHD Like traits on EF efficacy in TS vis a vis the TD group. Given the gender ratio differences in TS and ADHD in comparison to a TD group and given the observation of improvement in working memory and executive resources in this age group (Hamilton et al., 2003), both age and gender will be employed as covariates in the analyses.

The research discussed in the Introduction on ADHD comorbidity and EF impairment would lead to the prediction that ADHD Like measures, even at sub-diagnostic or sub-referral levels of intensity, will be significant mediators of any group differences in EF efficacy. However, given the complex pattern of EF challenges associated with ADHD co-occurrence, no specific EF process predictions will be made.

Method

Participants

Participants were opportunistically recruited from a self-selected sample of respondents through TS support groups and on social media platforms. An advertisement and recruitment poster stated the following inclusion criteria: for parents of children aged between 5 – 12 years old with or without a diagnosis of TS. Parental response identified children with and without a diagnosis for TS. A total of 62 respondents were excluded for not meeting the age range requirement or for not completing the measures. The final full sample for the study was: TS Group, $n = 58$, mean age = *10.03 (1.28)*, range 8-12 yrs., TD Group, $n = 88$, mean age = *10.14 (1.58)*, range 5-12 yrs. The age difference between the two groups was not significant, $F(1, 144) = 0.17, p > .05$. In the TS group, there were 12 girls and 46 boys, in the TD group there were 38 girls, and 50 boys. The Chi Square analysis revealed a significant association between group and gender, $X^2 = 7.85, p < .05$. A sub sample of participants was determined by the scoring on the Vanderbilt ADHD Diagnostic Parent Rating Scale (VADPRS, Wolraich et al., 2003), where a score of 2 or 3 on six of the 9 inattention questions and/or a score of 2 or 3 on six of the 9 hyperactivity and impulsivity items was recorded. This led to sub-referral sample size of n = 97, with 41 girls, with 71 TD children and 26 TS children not meeting or exceeding the ADHD referral criteria. A comparison of age across the two subgroups, TD and TS, indicated a non-significant difference, TD = *10.2 (1.57)*, TS = *9.96 (1.47)*, $F(1, 96) = 0.49, p > .05$. Again, Chi Square analyses revealed a significant association between Gender and Group, $X^2 = 5.36, p < .05$. The TS subgroup had 6 girls and 20 boys, the TD subgroup had 35 girls and 36 boys.

Materials

The Childhood Executive Functioning Inventory (Thorell & Nyberg, 2008) was employed to measure parental ratings of executive functions. The Inhibition component, with 6 items, had an internal reliability in this sample of $\alpha = .90$ in this sample. The Regulation component, with 5 items, had an internal reliability of $\alpha = .94$. The combined Inhibition and Regulation components, Inhibitory Control, had a reliability of $\alpha = .95$. The Planning and Organization component, with 4 items, had an internal reliability in this sample of $\alpha = .89$. The Working Memory component, with 9 items, had an internal reliability in this sample of $\alpha = .94$. Finally, the overall Working memory component (Planning and Working Memory) had $\alpha = .96$. All items were randomly presented in the Qualtrics survey and were rated on a five-point scale from one (Definitely not true) to five (Definitely true), completed by the parent to state how well each statement accurately described their child, and thus provided a parental-report measure of these various components of executive function. Possible scores range from 24 to 120, higher scores on all 4 components indicated more challenges with the child's EF competences.

The Vanderbilt ADHD Diagnostic Parent Rating Scale (VADPRS, Wolraich et al., 2003) was employed to identify variability in Inattention and Hyperactivity/Impulsivity characteristics. The Inattention component, with 9 items, had an internal reliability in this sample of $\alpha = .94$; the Hyperactivity/Impulsivity component, with 9 items, had an internal reliability in this sample of $\alpha = .92$. The reliability for the full combined scale, Inattention + Hyperactivity/Impulsivity was $\alpha = .96$. All items were randomly presented in the survey and were rated on a four-point scale with 0 (Never) to 3 (Very Often). The diagnostic criteria for referral based on these scores was identified above. Scores in both subscales were added together to indicate ADHD Like Combined traits based on the parent's ratings of their child's behaviour in both components over the past six months. Higher scores on both measures indicated higher ADHD Like trait possession.

Procedure

The Ethics Committee of the Department of Psychology, Northumbria University approved the research ethics submission from which these data were obtained. Parental consent was required for participation in the study. The materials and procedures used in this study adhered to the principles of the Declaration of Helsinki. Participants were recruited from a number of social media web sites and asked to complete the on-line questionnaires via the Qualtrics platform.

Results

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The first statistical consideration focused upon the variable descriptive statistics in the full and in the sub-referral samples. Table 1 (see below) indicates the full sample (bold) TD Group and TS Group mean ratings for all the key measures. In all of the measures the mean performance of the TS Group was higher, which indicated more challenges in these measures. Across all of the measures with a full sample the effect sizes, ***d***, were large. This pattern was largely reflected in the sub-referral groups (not bold) where several large effect sizes associated with group difference persisted.

Insert Table 1 here.

Therefore, even when the sample is constrained to children with sub-referral level ADHD-Like scores, there remains several large effects sizes associated with the Group differences in the executive function and ADHD-Like rating scores.

In order to establish whether the group differences in Table 1 were significant, two MANCOVA analyses were carried out initially on the full sample and subsequently on the sub-referral sample, on the four CHEXI processes and then on the two VADPRS components. Age and gender were controlled in both analyses.

In the full sample, using Wilk's lambda, the MANCOVA with the CHEXI measures revealed a significant effect associated with Group, $\lambda = 0.76, F(4, 139) = 10.82, p < .001$, partial $\eta^2 = .237$, Box's $M = 15.15, p = .145$. Subsequent ANCOVA analyses showed a significant Group effect with CHEXI Inhibition, $F(1, 142) = 32.55, p < .001$, *partial* $\eta^2 = .187$; CHEXI Regulation, $F(1, 142) = 28.55, p < .001$, *partial* $\eta^2 = .167$; CHEXI Planning, $F(1, 142) = 27.191, p < .001$, *partial* $\eta^2 = .161$; and CHEXI Working Memory, $F(1, 142) =$

39.15, $p < .001$, partial $\eta^2 = .216$. Using Wilk's lambda, the MANCOVA with the VADPRS revealed a significant effect associated with Group, *$\lambda = 0.81$, $F(4, 139) = 16.12$, $p < .001$*, partial $\eta^2 = .186$, Box's $M = 3.02$, $p = .145$. Subsequent ANOVA analyses also showed a significant Group effect with VADPRS Inattention, *$F(1, 142) = 30.06$, $p < .001$, partial $\eta^2 = .175$*; VADPRS Hyperactive, *$F(1, 142) = 27.33$, $p < .001$, partial $\eta^2 = .161$.*

In the sub-referral sample analyses, using Wilk's lambda, the MANCOVA with the CHEXI measures revealed a significant effect associated with Group, *$\lambda = 0.876$, $F(4, 90) = 3.18$, $p = .017$*, partial $\eta^2 = .124$, Box's $M = 10.32$, $p = .471$. Subsequent ANCOVA analyses showed a significant Group effect with CHEXI Inhibition, *$F(1, 93) = 12.67$, $p < .001$, partial $\eta^2 = .120$*; CHEXI Regulation, *$F(1, 93) = 7.50$, $p = .007$, partial $\eta^2 = .075$*; CHEXI Planning, *$F(1, 93) = 4.95$, $p = .029$, partial $\eta^2 = .051$*; and CHEXI Working Memory, *$F(1, 93) = 6.72$, $p = .011$, partial $\eta^2 = .067$*. Using Wilk's lambda, the MANCOVA with the VADPRS revealed a marginally non-significant effect associated with Group, *$\lambda = 0.94$, $F(2, 92) = 2.88$, $p = .061$*, partial $\eta^2 = .059$, Box's $M = 1.42$, $p = .712$. Subsequent ANOVA analyses also showed a significant Group effect with VADPRS Inattention, *$F(1 ,93) = 5.56$, $p = .021$, partial $\eta^2 = .056$*; VADPRS Hyperactive, *$F(1, 93) = 3.96$, $p = .050$, partial $\eta^2 = .041$.*

The MANCOVA analyses indicated that in the full sample, all of the group differences in both CHEXI and the VADPRS measures were significant. This was the case for the majority of subgroup analyses. Thus, in these measures, the TS Group showed significantly more challenges, in both the full and sub-referral group analyses.

To establish whether these measures were correlated with one another partial correlation analyses (controlling for Age and Gender) were carried out. The partial correlations indicated strong and significant relationships within and between the CHEXI and

VADPRS measures. This was the case for the both the full sample and sub-referral sample (see Table 2).

Insert Table 2 here.

The results shown in Table 2 indicate partial correlations controlling for age and gender for the full sample (bold) shown above the diagonal. Shown below the diagonal in Table 2 is the partial correlation coefficients for the sub-referral sample. All partial correlations were highly significant.

The mediation analyses, employing global (summary) measures considered the mediation effects of the VADPRS Inattention, VADPRS Hyperactivity, and VADPRS Combined measures on the difference in Overall Working Memory (Planning + WM) and the Overall Inhibition (Inhibition + Regulation) scores between the TD and TS Groups (see Figure 1). The mediation analyses looked at the contribution of the ADHD Like trait characteristics to the TD and TS Group differences in the CHEXI measures. Following a dimensional approach, The Inattention, Hyperactivity- impulsivity and the Inattention/Hyperactivity-Impulsivity combined ADHD rating scores were employed as mediators in these analyses (Hayes, 2018).

Insert Figure 1 here.

The first analyses considered the mediation effects of the VADPRS Inattention, Hyperactivity, and Combined measures on the difference in global Working Memory (Planning + WM) and the global Inhibition (Inhibition + Regulation) scores between the TD and TS Groups in the full sample (see Figure 1).

In all these full sample mediation analyses the ADHD Like traits significantly mediated the TD and TS Group relationship with both the CHEXI Working Memory and Inhibition/Regulation global scores. Therefore, a significant contribution to the EF differences between the groups was accounted for by the concurrent group differences in the VADPRS measures. Note though that the direct effect of Group upon the CHEXI measures remained significant in some of the models. Thus, the ADHD Like traits did not account for all of these global EF group differences. Further mediation analyses on the full sample were carried out to identify the mediation effect of the ADHD Like characteristics in explaining the TD and TS Group differences in each of the CHEXI components (see Table 3). This was carried out for the full sample (bold) and sub-referral sample.

Insert Table 3 here.

Table 3 demonstrates that in all the models with the full sample, the ADHD Like measures significantly mediated the TD and TS group difference across all four CHEXI components: Working Memory, Planning, Inhibition, and Regulation. In some cases this mediation led to the direct relationship, *c'*, between Group and the outcome variable, the CHEXI measure, becoming non-significant.

A final set of mediation analyses was carried out on the sub sample whose VADPRS score profile would not warrant a clinical referral. These findings are reported in Table 4.

Insert Table 4 here

These findings indicate that in all of these sub-referral sample mediation analyses the ADHD like measures were significant mediators of the Group – EF relationship. In a small number of models the direct effect remained significant.

In summary, the ANCOVA results suggest that TD and the TS group differences in executive function efficacy *remain* when the sample is constrained to TD and TS children with sub-referral levels of ADHD Like ratings. Consequently, these high, but sub-referral levels of ADHD characteristics are still associated with impaired executive functioning. The importance of recognising sub-referral levels of ADHD Like characteristics is supported by the mediation analyses which indicate that each of the three ADHD Like measures significantly mediate the relationship between Group and each of the executive functions in both the full sample and sub-referral samples. Consequently, the importance of the ADHD Like characteristic mediation remains important at levels of possession below the conventional diagnostic referral levels.

Discussion

The aim of this research was to identify the extent to which ADHD Like traits in children with a TS diagnosis could account for any challenges in everyday EF performance when employing an ecological measure of EF efficacy, the Child Executive Functioning Inventory. More importantly, what further made this research novel was the explicit consideration of the impact of sub-referral ADHD Like trait possession. This is the group of children who received VADPRS rating scores which were relatively high but below the Wolraich et al. (2003) referral criteria for further ADHD diagnostic consideration. Therefore, a dimensional or continuum approach to the importance of ADHD Like characteristics was emphasised in this study.

Evidence for the presence of these ADHD Like characteristics in both groups in the present study is also supported in Table 1 where despite mean differences, the range of ADHD Like scores shows substantial overlap between the TD and TS groups. A pattern which is not inconsistent with a dimensional approach. Table 1 also indicates that the TS group had a greater challenge across most of the EF (and ADHD Like) measures, and this was supported by the two MANCOVA analyses in the full and sub-referral group samples. However, the key analyses were the mediation analyses, as these analyses directly examined the extent to which the group differences in EF efficacy could be accounted for by group differences in the possession of ADHD characteristics.

Generally, the pattern of results was unequivocal, the group difference in EF challenges was reliably mediated by the ADHD Like characteristics. In all the models, both in the full sample and in the sub-referral sample, the ADHD Like trait mediation contribution was significant, for all the individual CHEXI components. In many of the models the mediation effect of the ADHD Like traits led to a non-significant direct effect, *c′*, in the Group to CHEXI outcome variable relationship, suggesting that a dominant factor in explaining the initial group difference for these EF processes was the greater possession of ADHD Like characteristics in the TS group. This was the case in both the full sample *and* the sub-referral sample analyses.

This pattern of mediation findings, in combination with the high correlations noted in Table 2, could point towards the suggestion that EF and ADHD Like characteristics share common cognitive characteristics (Drechsler et al., 2018; Linder et al., 2010). In many ways this overlap is an inevitable consequence of the EF methodology employed, both Linder et al., and Drechsler et al., employed the BRIEF (Gioia et al, 2000) parental report tool to assess EF. Since both questionnaires are assessing behaviour in an ecological context, it is not too unsurprising that some elements of the questionnaire may assess similar behaviours.

Whilst there are methodological reasons why the EF and VADPRS are so highly correlated (see also the comments below on a 'negative halo effect' below) there are also theoretical reasons why the EF and VADPRS measures are strongly correlated. Barkley (1997, 2015) has argued that ADHD is essentially an EF disorder. Should this be the case it is understandable that the EF and VADPRS measures would strongly correlate. These observations suggest that although EF and ADHD Like features are understandably associated, they may not be isomorphic with one another, an observation supporting the conclusions of Linder et al., (2010) and of Drechsler et al., (2018). However, these results also suggest that the degree of overlap between EF and ADHD Like characteristics in children with TS may be greater that observed with the use of task-based EF measures (Openneer et al., 2020, however, see negative halo effect discussion below). It should be noted that the identification of the degree of overlap between EF and ADHD measures is an ongoing research question (e.g., Al-Yagon & Borenstein, 2022).

This comprehensive degree of mediation by sub-referral levels of ADHD Like traits, identified in Table 4, where high but sub-referral levels of ADHD Like traits consistently account for group differences in EF has important implications for cognitive interventions in TS This implies that any intervention undertaken to reduce the observed cognitive challenges in children with TS needs to take into consideration the possession of ADHD Like traits, even when these traits may be at a sub-referral level (Agnew-Blais & Michelini, 2023)..

Limitations

The current findings need to be replicated, and to be extended to different measures of EF and ADHD Like traits. Both other ecologically driven measures such as the BRIEF (Gioia et al., 2000) and task-based EF could be concurrently employed, to enable a direct consideration of the impact of ADHD Like traits with both formats of EF assessment. The use of cognitive task performance alongside the use of rating scales provides complimentary information about EF efficacy in everyday behaviour (Gioia et al., 2000; Salthouse, 2012; Toplak et al., 2013). By employing measures of internalisation and externalisation alongside EF performance and rating measures one could also account for some of the discrepancies between performance and rating EF efficacies (Williams et al., 2022). The Williams et al. study highlights some of the ways in which rating judgments may be biased (see also Denckla's , 2002, observation of halo effects in participant rating judgments). Other measures of ADHD Like traits in the general population such as the Conners Revised Parent Rating Scale (Conners et al., 1998) could be employed to ensure the current pattern of significant mediation is replicable across a range of ADHD Like rating measures.

A further issue with the current research is the use of on-line sampling procedure employed to recruit participant families. The use of TS web forums could potentially not only recruit children with a diagnosis of TS but enable a parent/carer to also rate the child's biological sibling(s) who may not have a TS diagnosis. This possibility means that the apparent group difference observed in the present study may be an underestimate of the true difference in cognitive challenge between the TD and sub-referral TS groups. Additionally, the diagnostic status of the children was derived entirely from parental reporting, a common method employed in the initial phase of many diagnostic processes. Future studies could bring a formal diagnostic indicator process to the research along with other clinical characteristics pertinent to the cognitive performance, e.g., age of onset of the TS, severity of tics etc. This approach could produce a more clinically nuanced, idiosyncratic view of the nature of the TS features association with EF challenge.

In addition, some individuals in the group of children who met the referral criteria may **not** go on to receive a formal ADHD diagnosis. Consequently, the present study should be considered in terms of sample categorisation based upon VADPRS ratings below and above the referral criteria rather than children with or without an ADHD diagnosis. Future research may want to consider in detail in children the impact of VADPRS scores which meet the referral criteria, but the child does or does not receive a subsequent formal ADHD diagnosis.

An additional putative limitation of the current study is the use of CHEXI and VADPRS scales to measure executive function and ADHD Like characteristics. Like most measures in psychological science at any point in time, measures which appear to be validated in the extant literature will upon the subsequent publication of empirical and theoretical observations be subject to methodological refinement. Thus, it could be argued that these current measures could merely reflect concentration difficulties, impulsivity, disorganization, etc. This is particularly the case when the research literature has suggested that ADHD characteristics are so commonly found in a range of neurodevelopment contexts (see Pliszka, 2015, Figure 5.1). Co-occurring observations such as these have led Pliszka to seriously question the concept of clinical comorbidity, instead advocating a new paradigm that emphasises major psychological dimensions such as inattention, emotional regulation, executive function which would have transdiagnostic implications (see also Krakowski et al., 2022; Viadya et al., 2020). Initial candidates for these primary transdiagnostic dimensions have been the *c* factor (Abramovitch et al., 2021) and the *d* factor (Zhang et al., 2022). Future research within TS could also incorporate these generic measures.

Conclusion

The current study adopted a dimensional approach to the confounding presence of ADHD Like traits in EF efficacy and investigated the extent to which these characteristics could account for TS and TD group differences in an everyday measure of EF, the CHEXI measure. The findings suggested that group differences were present in all the key EF and ADHD Like measures, these measures also reliably correlated with one another, even when age and gender were controlled. A pattern of findings which was observed in both the full sample and sub-referral sample. The presence of ADHD Like characteristics demonstrated significant mediation in all of the models assessed; however importantly, the present findings suggested that in all of the sub-referral sample analyses the mediation analyses indicated significant mediation by the VADPRS Inattention, Hyperactivity, and the VADPRS Combined measures. Thus, the negative impact of ADHD Like traits upon EF function in children with TS is not limited to children with a formal ADHD diagnosis.

References

Abramovitch, A., Short, T., & Schweiger, A. (2021). The C Factor: Cognitive dysfunction as a transdiagnostic dimension in psychopathology. *Clin Psychol Rev, 86*, 102007. doi:10.1016/j.cpr.2021.102007

Agnew-Blais, J., & Michelini, G. (2023). Taking stock of the present and looking to the future of ADHD research: a commentary on Sonuga-Barke et al. *Journal of Child Psychology and Psychiatry*, 64(4), 533-536. https://doi.org/https://doi.org/10.1111/jcpp.13758 Alderson, R. M., Rapport, M. D., & Kofler, M. J. (2007). Attention-deficit/hyperactivity disorder and behavioral inhibition: a meta-analytic review of the stop-signal paradigm. *J Abnorm Child Psychol, 35*(5), 745-758. doi:10.1007/s10802-007-9131-6Al-Yagon, M., & Borenstein, T. (2022). Adolescents' executive functions: Links to inattention, hyperactivity-impulsivity, trait mindfulness, and attachment relationships with fathers and mothers. *Res Dev Disabil, 124*, 104212. doi:10.1016/j.ridd.2022.104212

Ameis, S. H., Haltigan, J. D., Lyon, R. E., Sawyer, A., Mirenda, P., Kerns, C. M., . . . Pathways in, A. S. D. S. T. (2022). Middle-childhood executive functioning mediates associations between early-childhood autism symptoms and adolescent mental health, academic and functional outcomes in autistic children. *J Child Psychol Psychiatry, 63*(5), 553-562. doi:10.1111/jcpp.13493

Anderson, N. P., Feldman, J. A., Kolko, D. J., Pilkonis, P. A., & Lindhiem, O. (2022). National Norms for the Vanderbilt ADHD Diagnostic Parent Rating Scale in Children. *J Pediatr Psychol, 47*(6), 652-661. doi:10.1093/jpepsy/jsab132

Barkley, R. A. (1997). Behavioral inhibition, sustained attention, and executive functions: constructing a unifying theory of ADHD. *Psychol Bull, 121*(1), 65-94. doi:10.1037/0033-2909.121.1.65

Barkley, R. A. (2010). Differential diagnosis of adults with ADHD: the role of executive function and self-regulation. *J Clin Psychiatry, 71*(7), e17. doi:10.4088/JCP.9066tx1c

Barkley, R. A. (2015). Executive Functioning and Self-Regulation Viewed as an Extended Phenotype: Implications of the theory for ADHD and its Treatment. In R. A. Barkley (Ed.), *Attention-Deficit Hyperactivity Disorder* (pp. 405-434). London: The Guildford Press. (Reprinted from: 4th).

Baron-Cohen, S., Wheelwright, S., Skinner, R., Martin, J., & Clubley, E. (2001). The autism-spectrum quotient (AQ): evidence from Asperger syndrome/high-functioning autism, males and females, scientists and mathematicians. *J Autism Dev Disord, 31*(1), 5-17. doi:10.1023/a:1005653411471

Blanken, L. M. E., Muetzel, R. L., Jaddoe, V. W. V., Verhulst, F. C., van der Lugt, A., Tiemeier, H., & White, T. (2017). White matter microstructure in children with autistic traits. *Psychiatry Res Neuroimaging, 263*, 127-134. doi:10.1016/j.pscychresns.2017.03.015

Casey, B. J., Heller, A. S., Gee, D. G., & Cohen, A. O. (2019). Development of the emotional brain. *Neurosci Lett, 693*, 29-34. doi:10.1016/j.neulet.2017.11.055

Cavanna, A. E., Ganos, C., Hartmann, A., Martino, D., Pringsheim, T., & Seri, S. (2020). The cognitive neuropsychiatry of Tourette syndrome. *Cogn Neuropsychiatry, 25*(4), 254-268. doi:10.1080/13546805.2020.1760812

Conners, C. K., Sitarenios, G., Parker, J. D., & Epstein, J. N. (1998). The revised Conners' Parent Rating Scale (CPRS-R): factor structure, reliability, and criterion validity. *J Abnorm Child Psychol, 26*(4), 257-268. doi:10.1023/a:1022602400621

Constantino, J. N., & Todd, R. D. (2003). Autistic traits in the general population: a twin study. *Arch Gen Psychiatry, 60*(5), 524-530. doi:10.1001/archpsyc.60.5.524

Czamara, D., Tiesler, C. M., Kohlbock, G., Berdel, D., Hoffmann, B., Bauer, C. P., . . . Heinrich, J. (2013). Children with ADHD symptoms have a higher risk for reading, spelling and math difficulties in the GINIplus and LISAplus cohort studies. *PLoS One, 8*(5), e63859. doi:10.1371/journal.pone.0063859

Denckla, M. B. (2002). The Behavior Rating Inventory of Executive Function: commentary. *Child Neuropsychol, 8*(4), 304-306. doi:10.1076/chin.8.4.304.13512

Diamond, A. (2013). Executive functions. *Annu Rev Psychol, 64*, 135-168. doi:10.1146/annurev-psych-113011-143750

Donati, G., Meaburn, E., & Dumontheil, I. (2021). Internalising and externalising in early adolescence predict later executive function, not the other way around: a cross-lagged panel analysis. *Cogn Emot, 35*(5), 986-998. doi:10.1080/02699931.2021.1918644

Drechsler, R., Zulauf Logoz, M., Walitza, S., & Steinhausen, H. C. (2018). The Relations Between Temperament, Character, and Executive Functions in Children With ADHD and Clinical Controls. *J Atten Disord, 22*(8), 764-775. doi:10.1177/1087054715583356

DuPaul, G. J., Gormley, M. J., Anastopoulos, A. D., Weyandt, L. L., Labban, J., Sass, A. J., . . . Postler, K. B. (2021). Academic Trajectories of College Students with and without ADHD: Predictors of Four-Year Outcomes. *J Clin Child Adolesc Psychol, 50*(6), 828-843. doi:10.1080/15374416.2020.1867990

Feldman, J. S., & Huang-Pollock, C. (2021). A New Spin on Spatial Cognition in ADHD: A Diffusion Model Decomposition of Mental Rotation. *J Int Neuropsychol Soc, 27*(5), 472-483. doi:10.1017/S1355617720001198

Friedman, N. P., & Miyake, A. (2004). The relations among inhibition and interference control functions: a latent-variable analysis. *J Exp Psychol Gen, 133*(1), 101-135. doi:10.1037/0096-3445.133.1.101

Geurts, H. M., Pol, S. E., Lobbestael, J., & Simons, C. J. P. (2020). Executive Functioning in 60+ Autistic Males: The Discrepancy Between Experienced Challenges and Cognitive Performance. *J Autism Dev Disord, 50*(4), 1380-1390. doi:10.1007/s10803-020-04368-9

Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). Behavior Rating Inventory of Executive Function. *Child Neuropsychology, 6*(3), 235-238.

Greven, C. U., Merwood, A., van der Meer, J. M., Haworth, C. M., Rommelse, N., & Buitelaar, J. K. (2016). The opposite end of the attention deficit hyperactivity disorder continuum: genetic and environmental aetiologies of extremely low ADHD traits. *J Child Psychol Psychiatry, 57*(4), 523-531. doi:10.1111/jcpp.12475

Hamilton, C., Coates, R., & Heffernan, T. (2003). What develops in visuo-spatial working memory development? *European Journal of Cognitive Psychology, 15*(1), 43-69. doi:10.1080/09541440303597

Hamilton, C. J., Mammarella, I. C., & Giofre, D. (2018). Autistic-like traits in children are associated with enhanced performance in a qualitative visual working memory task. *Autism Res, 11*(11), 1494-1499. doi:10.1002/aur.2028

Hayes, A. F. (2018). *Introduction to Mediation, Moderation, and Conditional Process Analysis A Regression-Based Approach 2nd Edition*. New York: Guilford Press.

Heidbreder, R. (2015). ADHD symptomatology is best conceptualized as a spectrum: a dimensional versus unitary approach to diagnosis. *Atten Defic Hyperact Disord, 7*(4), 249-269. doi:10.1007/s12402-015-0171-4

Hirschtritt, M. E., Lee, P. C., Pauls, D. L., Dion, Y., Grados, M. A., Illmann, C., . . . Tourette Syndrome Association International Consortium for, G. (2015). Lifetime prevalence, age of risk, and genetic relationships of comorbid psychiatric disorders in Tourette syndrome. *JAMA Psychiatry, 72*(4), 325-333. doi:10.1001/jamapsychiatry.2014.2650

Hovik, K. T., Egeland, J., Isquith, P. K., Gioia, G., Skogli, E. W., Andersen, P. N., & Oie, M. (2017). Distinct Patterns of Everyday Executive Function Problems Distinguish Children With Tourette Syndrome From Children With ADHD or Autism Spectrum Disorders. *J Atten Disord, 21*(10), 811-823. doi:10.1177/1087054714550336

Irwin, L. N., Groves, N. B., Soto, E. F., & Kofler, M. J. (2020). Is There a Functional Relation Between Set Shifting and Hyperactivity in Children with Attention-Deficit/Hyperactivity Disorder (ADHD)? *J Int Neuropsychol Soc, 26*(10), 1019-1027. doi:10.1017/S1355617720000545

Ishizuka, K., Ishiguro, T., Nomura, N., & Inada, T. (2022). Autistic traits as predictors of persistent depression. *Eur Arch Psychiatry Clin Neurosci, 272*(2), 211-216. doi:10.1007/s00406-021-01292-6

Kibby, M. Y., Newsham, G., Imre, Z., & Schlak, J. E. (2021). Is executive dysfunction a potential contributor to the comorbidity between basic reading disability and attention-deficit/hyperactivity disorder? *Child Neuropsychol, 27*(7), 888-910. doi:10.1080/09297049.2021.1908532

Kildal, E., Hassel, B., & Bjornebekk, A. (2022). ADHD symptoms and use of anabolic androgenic steroids among male weightlifters. *Sci Rep, 12*(1), 9479. doi:10.1038/s41598-022-12977-w

Kilford, E. J., Garrett, E., & Blakemore, S. J. (2016). The development of social cognition in adolescence: An integrated perspective. *Neurosci Biobehav Rev, 70*, 106-120. doi:10.1016/j.neubiorev.2016.08.016

Krakowski, A. D., Cost, K. T., Szatmari, P., Anagnostou, E., Crosbie, J., Schachar, R., . . . Barnett-Tapia, C. (2022). Characterizing the ASD-ADHD phenotype: measurement structure and invariance in a clinical sample. *J Child Psychol Psychiatry*. doi:10.1111/jcpp.13609

Lijffijt, M., Kenemans, J. L., Verbaten, M. N., & van Engeland, H. (2005). A meta-analytic review of stopping performance in attention-deficit/hyperactivity disorder: deficient inhibitory motor control? *J Abnorm Psychol, 114*(2), 216-222. doi:10.1037/0021-843X.114.2.216

Linder, N., Kroyzer, N., Maeir, A., Wertman-Elad, R., & Pollak, Y. (2010). Do ADHD and executive dysfunctions, measured by the hebrew version of Behavioral Rating Inventory of Executive Functions (BRIEF), completely overlap? *Child Neuropsychol, 16*(5), 494-502. doi:10.1080/09297041003781884

Miyake, A., & Friedman, N. P. (2012). The Nature and Organization of Individual Differences in Executive Functions: Four General Conclusions. *Curr Dir Psychol Sci, 21*(1), 8-14. doi:10.1177/0963721411429458

Miyake, A., Friedman, N. P., Emerson, M. J., Witzki, A. H., Howerter, A., & Wager, T. D. (2000). The unity and diversity of executive functions and their contributions to complex "Frontal Lobe" tasks: a latent variable analysis. *Cogn Psychol, 41*(1), 49-100. doi:10.1006/cogp.1999.0734

Morand-Beaulieu, S., Leclerc, J. B., Valois, P., Lavoie, M. E., O'Connor, K. P., & Gauthier, B. (2017). A Review of the Neuropsychological Dimensions of Tourette Syndrome. *Brain Sci, 7*(8). doi:10.3390/brainsci7080106

Moses, M., Tiego, J., Demontis, D., Bragi Walters, G., Stefansson, H., Stefansson, K., . . . Bellgrove, M. A. (2022). Working memory and reaction time variability mediate the relationship between polygenic risk and ADHD traits in a general population sample. *Mol Psychiatry*. doi:10.1038/s41380-022-01775-5

Oberauer, K., & Lin, H. Y. (2017). An interference model of visual working memory. *Psychol Rev, 124*(1), 21-59. doi:10.1037/rev0000044

Openneer, T. J. C., Forde, N. J., Akkermans, S. E. A., Naaijen, J., Buitelaar, J. K., Hoekstra, P. J., & Dietrich, A. (2020). Executive function in children with Tourette syndrome and attention-deficit/hyperactivity disorder: Cross-disorder or unique impairments? *Cortex, 124*, 176-187. doi:10.1016/j.cortex.2019.11.007

Pliszka, S. R. (2015). Comorbid Psychiatric Disorders in Children with ADHD. In R. A. Barkley (Ed.), *Attention-Deficit Hyperactivity Disorder* (pp. 140-168). London: The Guildford Press. (Reprinted from: 4th).

Rivella, C., & Viterbori, P. (2021). [Formula: see text] Executive function following pediatric stroke. A systematic review. *Child Neuropsychol, 27*(2), 209-231. doi:10.1080/09297049.2020.1820472

Rizzo, R., Gulisano, M., Pellico, A., Cali, P. V., & Curatolo, P. (2014). Tourette syndrome and comorbid conditions: a spectrum of different severities and complexities. *J Child Neurol, 29*(10), 1383-1389. doi:10.1177/0883073814534317

Roberts, W., Milich, R., & Barkley, R. A. (2015). Primary Symptoms, Diagnostic Criteria, Subtyping, and Prevalance of ADHD. In R. A. Barkley (Ed.), *Attention-Deficit Hyperactivity Disorder* (pp. 51-80). London: The Guildford Press. (Reprinted from: 4th).

Rosello, B., Berenguer, C., Baixauli, I., Colomer, C., & Miranda, A. (2018). ADHD symptoms and learning behaviors in children with ASD without intellectual disability. A mediation analysis of executive functions. *PLoS One, 13*(11), e0207286. doi:10.1371/journal.pone.0207286

Salthouse, T. (2012). Consequences of age-related cognitive declines. *Annu Rev Psychol, 63*, 201-226. doi:10.1146/annurev-psych-120710-100328

Schweizer, S., Gotlib, I. H., & Blakemore, S. J. (2020). The role of affective control in emotion regulation during adolescence. *Emotion, 20*(1), 80-86. doi:10.1037/emo0000695

Tamm, L., Loren, R. E. A., Peugh, J., & Ciesielski, H. A. (2021). The Association of Executive Functioning With Academic, Behavior, and Social Performance Ratings in Children With ADHD. *J Learn Disabil, 54*(2), 124-138. doi:10.1177/0022219420961338

Thorell, L. B., Lazarevic, N., Milovanovic, I., & Bugarski Ignjatovic, V. (2020). Psychometric properties of the Teenage Executive Functioning Inventory (TEXI): A freely available questionnaire for assessing deficits in working memory and inhibition among adolescents. *Child Neuropsychol, 26*(6), 857-864. doi:10.1080/09297049.2020.1726885

Thorell, L. B., & Nyberg, L. (2008). The childhood executive functioning inventory (CHEXI): a new rating instrument for parents and teachers. *Dev Neuropsychol, 33*(4), 536-552. doi:10.1080/87565640802101516

Toplak, M. E., West, R. F., & Stanovich, K. E. (2013). Practitioner review: do performance-based measures and ratings of executive function assess the same construct? *J Child Psychol Psychiatry, 54*(2), 131-143. doi:10.1111/jcpp.12001

van den Berg, L., de Weerd, A. W., Reuvekamp, M. H. F., & van der Meere, J. J. (2021). Associating executive dysfunction with behavioral and socioemotional problems in children with epilepsy. A systematic review. *Child Neuropsychol, 27*(5), 661-708. doi:10.1080/09297049.2021.1888906

Venables, N. C., Foell, J., Yancey, J. R., Kane, M. J., Engle, R. W., & Patrick, C. J. (2018). Quantifying Inhibitory Control as Externalizing Proneness: A Cross-Domain Model. *Clinical Psychological Science, 6*(4), 561-580.

Vogel, S. W. N., Ten Have, M., Bijlenga, D., de Graaf, R., Beekman, A. T. F., & Kooij, J. J. S. (2018). Distribution of ADHD symptoms, and associated comorbidity, exposure to risk factors and disability: Results from a general population study. *Psychiatry Res, 267*, 256-265. doi:10.1016/j.psychres.2018.06.017

Vogel, S. W. N., Ten Have, M., Bijlenga, D., de Graaf, R., Beekman, A. T. F., & Kooij, J. J. S. (2018). Distribution of ADHD symptoms, and associated comorbidity, exposure to risk factors and disability: Results from a general population study. *Psychiatry Res, 267*, 256-265. doi:10.1016/j.psychres.2018.06.017

Welsh, M., & Peterson, E. (2014). Issues in the conceptualization and assessment of hot executive functions in childhood. *J Int Neuropsychol Soc, 20*(2), 152-156. doi:10.1017/S1355617713001379

Will, E. A., Schworer, E. K., & Esbensen, A. J. (2021). [Formula: see text] The role of distinct executive functions on adaptive behavior in children and adolescents with Down syndrome. *Child Neuropsychol, 27*(8), 1054-1072. doi:10.1080/09297049.2021.1917531

Williams, K. L., Holmes, J., Farina, F., Vedechkina, M., team, C., & Bennett, M. P. (2022). Inconsistencies between Subjective Reports of Cognitive Difficulties and Performance on Cognitive Tests are Associated with Elevated Internalising and Externalising Symptoms in Children with Learning-related Problems. *Res Child Adolesc Psychopathol*. doi:10.1007/s10802-022-00930-4

Wolicki, S. B., Bitsko, R. H., Danielson, M. L., Holbrook, J. R., Zablotsky, B., Walkup, J. T., . . . Mink, J. W. (2019). Children with Tourette Syndrome in the United States: Parent-Reported Diagnosis, Co-Occurring Disorders, Severity, and Influence of Activities on Tics. *J Dev Behav Pediatr, 40*(6), 407-414. doi:10.1097/DBP.0000000000000667

Wolraich, M. L., Lambert, W., Doffing, M. A., Bickman, L., Simmons, T., & Worley, K. (2003). Psychometric properties of the Vanderbilt ADHD diagnostic parent rating scale in a referred population. *J Pediatr Psychol, 28*(8), 559-567. doi:10.1093/jpepsy/jsg046

Yaniv, A., Benaroya-Milshtein, N., Steinberg, T., Ruhrrman, D., Apter, A., & Lavidor, M. (2017). Specific executive control impairments in Tourette syndrome: The role of response inhibition. *Res Dev Disabil, 61*, 1-10. doi:10.1016/j.ridd.2016.12.007

Young, S. E., Friedman, N. P., Miyake, A., Willcutt, E. G., Corley, R. P., Haberstick, B. C., & Hewitt, J. K. (2009). Behavioral disinhibition: liability for externalizing spectrum disorders and its genetic and environmental relation to response inhibition across adolescence. *J Abnorm Psychol, 118*(1), 117-130. doi:10.1037/a0014657

Zelazo, P. D., & Carlson, S. M. (2012). Hot and cool executive function in childhood and adolescence: Development and plasticity. *Child Development Perspectives, 6*(4), 354-360.

Zhang, H., Miyake, A., Osborne, J., Shah, P., & Jonides, J. (2022). The d Factor: General Distractibility and Its Relations to ADHD Symptoms and Hyperfocus. PsyArXiv. https://psyarxiv.com/5nzs2/

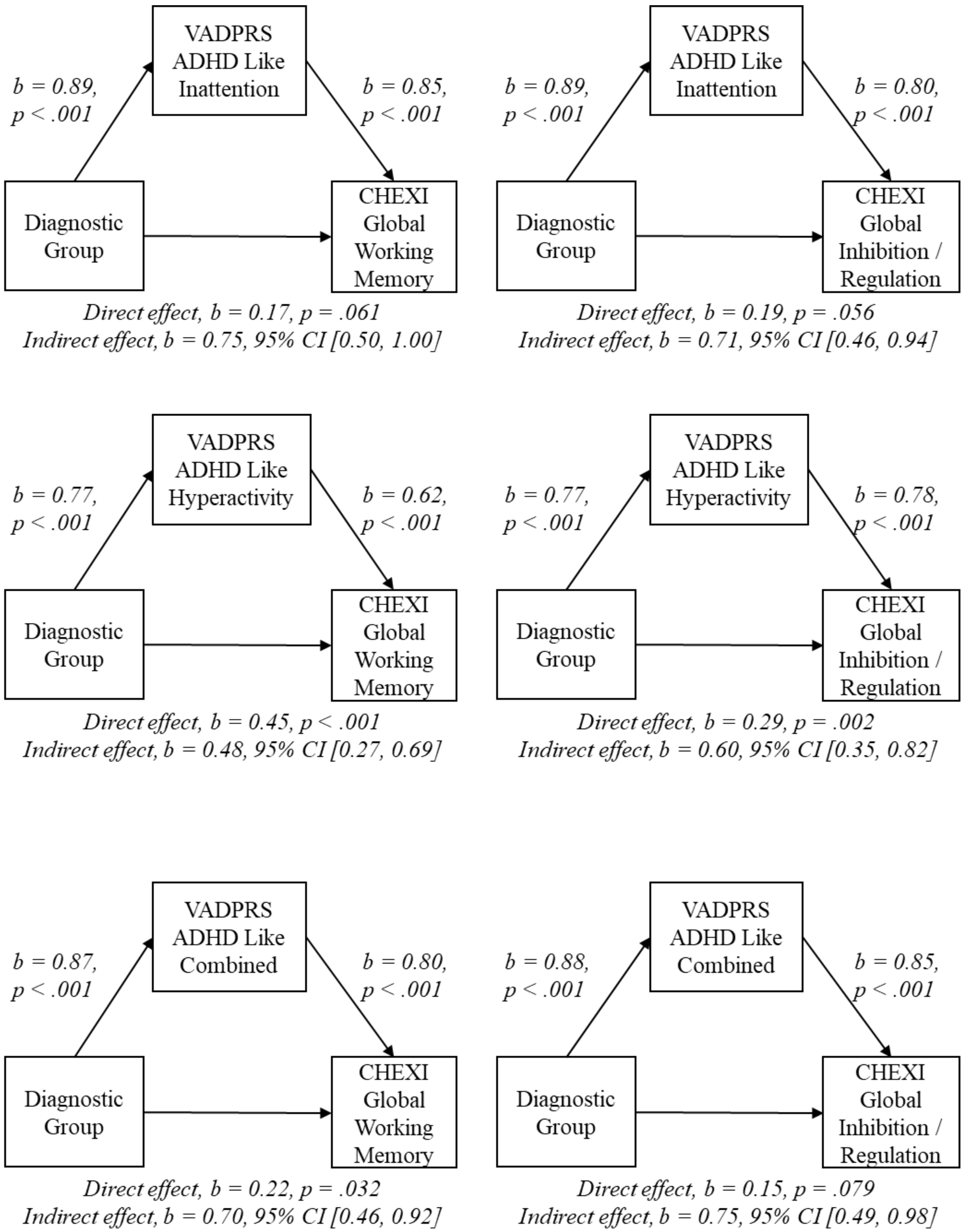

Figure 1. Full sample VADPRS mediation analyses of the Group – Combined (Global) CHEXI relationship

Note. CHEXI = Child Executive Functioning Inventory; VADPRS = Vanderbilt ADHD Diagnostic Parent Rating Scale

Table 1

Typical Development (TD) Group and Tourette Syndrome (TS) Group Descriptive Statistics as a Function of the Sample Characteristics

Measure	TD Group		TS Group		Effect Size
	Mean	SD	Mean	SD	***d***
CHEXI Overall Inhibition/ Regulation	***2.67***	***0.90***	***3.66***	***0.87***	***1.12***
	2.35	*0.66*	*2.96*	*0.73*	*0.90*
CHEXI Overall Working Memory	***2.28***	***0.84***	***3.28***	***0.97***	***1.12***
	2.03	*.66*	*2.48*	*.68*	*0.68*
CHEXI Inhibition	***2.45***	***0.90***	***3.43***	***0.92***	***1.08***
	2.15	*0.63*	*2.75*	*0.70*	*0.92*
CHEXI Regulation	***2.93***	***1.02***	***3.95***	***0.98***	***1.02***
	2.60	*0.81*	*3.22*	*0.98*	*0.72*
CHEXI Planning	***2.31***	***1.01***	***3.33***	***1.07***	***0.99***
	2.01	*0.63*	*2.51*	*0.82*	*0.73*
CHEXI Working Memory	***2.27***	***0.79***	***3.25***	***0.96***	***1.14***
	2.04	*0.63*	*2.47*	*0.66*	*0.67*
VADPRS Combined	***1.87***	***0.59***	***2.56***	***0.71***	***1.08***
	0.66	*0.42*	*0.93*	*0.36*	*0.67*
VADPRS Inattention	***1.95***	***0.64***	***2.69***	***0.76***	***1.02***
	0.73	*0.46*	*1.01*	*0.38*	*0.64*
VADPRS Hyperactivity / Impulsivity	***1.79***	***0.63***	***2.43***	***0.75***	***0.94***
	0.59	*0.45*	*0.84*	*0.41*	*0.57*

Note. CHEXI = Child Executive Functioning Inventory; VADPRS = Vanderbilt ADHD Diagnostic Parent Rating ScaleBold data = Full Sample; Non-bold = Sub-Referral Level Sample

Table 2.

Partial Correlations Controlling for Age and Gender between the Main Variables (n = 146)

	CHEXI Combined Inhibition & Regulation	CHEXI Combined Working Memory	CHEXI Inhibition	CHEXI Regulation	CHEXI Planning	CHEXI Working Memory	VADRS Combined	VADRS Inattention	VADRS Inhibition & Regulation
CHEXI Combined Inhibition/ Regulation		**.852**	**.949**	**.938**	**.823**	**.845**	**.896**	**.851**	**.842**
CHEXI Combined Working Memory	0.787		**.786**	**.824**	**.967**	**.992**	**.839**	**.884**	**.698**
CHEXI Inhibition	0.911	0.703		**.781**	**.753**	**.783**	**.859**	**.774**	**.852**
CHEXI Regulation	0.926	0.741	0.688		**.802**	**.814**	**.829**	**.834**	**.731**
CHEXI Planning	0.746	0.946	0.652	0.715		**.927**	**.807**	**.863**	**.658**
CHEXI Working Memory	0.774	0.985	0.699	0.722	0.875		**.834**	**.872**	**.700**
VADRS Combined	0.797	0.735	0.744	0.720	0.695	0.724		**.948**	**.943**
VADRS Inattention	0.759	0.756	0.665	0.725	0.741	0.731	0.923		**.786**
VADRS Hyperactivity / Impulsivity	0.710	0.598	0.707	0.602	0.539	0.604	0.921	0.700	

All correlations p < .001

Note. Above the diagonal in bold = full sample partial coefficients, below the diagonal = sub referral sample, partial coefficients controlling for Age and Gender

CHEXI = Child Executive Functioning Inventory;

VADPRS = Vanderbilt ADHD Diagnostic Parent Rating Scale

Table 3.

Mediation Models Identifying the Mediational Effect of the ADHD Like Measures Upon the Group – Child Executive Functioning Inventory Relationships with the Full Sample.

Model	Model Significance	*c′* (Direct) Predictor - Outcome	*a b* (Indirect) Mediation Effect
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI WM	*F(3, 142) = 15.71* *p < .001*	*Effect = 0.21* *p = .021*	*Effect = 0.73* *95% CI [0.45, 1.00]*
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI WM	*F(3, 142) = 15.71* *p < .001*	*Effect = 0.48* *p < .001*	*Effect = 0.48* *95% CI [0.27, 0.70]*
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI WM	*F(3, 142) = 15.71* *p < .001*	*Effect = 0.26* *p = .013*	*Effect = 0.68* *95% CI [0.42, 0.94]*
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI Planning	*F(3, 142) = 13.55* *p < .001*	*Effect = 0.07* *p = .512*	*Effect = 0.86* *95% CI [0.54, 1.16]*
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI Planning	*(3, 142) = 13.55* *p < .001*	*Effect = 0.36* *p = .010*	*Effect = 0.45* *95% CI [0.25, 0.67]*
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI Planning	*F(3, 142) = 13.55* *p < .001*	*Effect = 0.15* *p = .253*	*Effect = 0.78* *95% CI [0.49, 1.06]*
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI Inhibition	*F(3, 142) = 16.39* *p < .001*	*Effect = 0.24* *p = .047*	*Effect = 0.65* *95% CI [0.39, 0.91]*
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI Inhibition	*F(3, 142) = 16.39* *p < .001*	*Effect = 0.25* *p = .008*	*Effect = 0.61* *95% CI [0.38, 0.85]*
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI Inhibition	*F(3, 142) = 16.39* *p < .001*	*Effect = 0.15* *p = .123*	*Effect = 0.74* *95% CI [0.48, 1.00]*
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI Regulation	*F(3, 142) = 14.05* *p < .001*	*Effect = 0.13* *p = .270*	*Effect = 0.79* *95% CI [0.50, 1.07]*
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI Regulation	*(3, 142) = 14.05* *p < .001*	*Effect = 0.31* *p = .013*	*Effect = 0.52* *95% CI [0.32, 0.73]*
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI Regulation	*F(3, 142) = 14.05* *p < .001*	*Effect = 0.14* *p = .240*	*Effect = 0.78* *95% CI [0.51, 1.06]*

Note: VADPRS = Vanderbilt ADHD Diagnostic Parent rating scale*;* CHEXI = Child Executive Functioning Inventory; *Note:* CHEXI WM = Child Executive Functioning Inventory Working Memory

Table 4.

Mediation Models Identifying the Mediational Effect of the ADHD Like Measures Upon the Group – Child Executive Functioning Inventory Relationships with the Sub-Referral Sample.

Model	Model Significance	*c′* (Direct) Predictor - Outcome	*a b* (Indirect) Mediation Effect
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI WM	*F*(3, 93) = 3.06 *p* = .032	*Effect* = 0.14 *p* = .213	*Effect* = 0.26 *95% CI* [0.04, 0.48]
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI WM	*F*(3, 93) = 3.06 *p* = .032	*Effect* = 0.33 *p* = .089	*Effect* = 0.27 *95% CI* [0.02, 0.52]
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI WM	*F*(3, 93) = 3.06 *p* = .032	*Effect* = 0.14 *p* = .213	*Effect* = 0.25 *95% CI* [0.04, 0.47]
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI Planning	*F*(3, 93) = 3.90 *p* = .011	*Effect* = 0.09 *p* = .474	*Effect* = 0.32 *95% CI* [0.04, 0.57]
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI Planning	*F*(3, 93) = 3.90 *p* = .011	*Effect* = 0.27 *p* = .177	*Effect* = 0.23 *95% CI* [0.02, 0.47]
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI Planning	*F*(3, 93) = 3.90 *p* = .011	*Effect* = 0.11 *p* = .418	*Effect* = 0.30 *95% CI* [0.06, 0.54]
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI Inhibition	*F*(3, 93) = 6.98 *p* < .001	*Effect* = 0.31 *p* = .012	*Effect* = 0.23 *95% CI* [0.03, 0.42]
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI Inhibition	*F*(3, 93) = 6.98 *p* < .001	*Effect* = 0.47 *p* = .004	*Effect* = 0.30 *95% CI* [0.02, 0.60]
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI Inhibition	*F*(3, 93) = 6.98 *p* < .001	*Effect* = 0.28 *p* = .011	*Effect* = 0.26 *95% CI* [0.05, 0.48]
Predictor: Diagnostic Group Mediator: VADPRS Inattention Outcome: CHEXI Regulation	*F*(3, 93) = 3.73 *p* < .014	*Effect* = 0.22 *p* = .146	*Effect* = 0.26 *95% CI* [0.05, 0.48]
Predictor: Diagnostic Group Mediator: VADPRS Hyperactivity Outcome: CHEXI Inhibition	(3, 93) = 3.73 *p* < .014	*Effect* = 0.36 *p* = .061	*Effect* = 0.26 *95% CI* [0.02, 0.53]
Predictor: Diagnostic Group Mediator: VADPRS Combined Outcome: CHEXI Regulation	*F*(3, 93) = 3.73 *p* < .014	*Effect* = 0.22 *p* = .148	*Effect* = 0.34 *95% CI* [0.06, 0.65]

Note: VADPRS = Vanderbilt ADHD Diagnostic Parent rating scale*;* CHEXI = Child Executive Functioning Inventory; *Note:* CHEXI WM = Child Executive Functioning Inventory Working Memory