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Executive function in fluency and recall measures among children with Tourette syndrome or ADHD

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Abstract

This study assessed two relevant aspects of executive dysfunction in children with either Tourette syndrome (TS) or ADHD. Process variables derived from existing neuropsychological measures were used to clarify the executive function construct. Clustering of responses on measures of verbal fluency, figural fluency, and verbal learning was examined to assess strategic response organization. Rule breaks, intrusions, and repetition errors were recorded to assess inhibition errors. No significant differences were found among the three groups (TS, ADHD, and controls) on tasks of response organization (clustering). In our sample, both the ADHD and the TS groups were largely free from executive function impairment, and their performance on the fluency and list learning tasks was in the average range. There was a significant group difference on one of the disinhibition variables, with both TS and ADHD groups showing significantly more intrusions on verbal list learning trials than controls. When more traditional total score variables were analyzed among the three groups, there were no significant differences; however, analysis of effect size revealed medium-to-large effect sizes for Letter Word Fluency total score differences (ADHD *vs.* controls), and for Semantic Word Fluency total score differences (ADHD *vs.* TS), with the ADHD group having weaker performance in both comparisons. Results provide some support for the use and analysis of process variables—particularly those related to inhibition and intrusion errors, in addition to the total score variables when assessing executive function deficits in children with ADHD and TS. While group differences may be found, children with uncomplicated TS should not routinely be considered to have significant executive function impairments, and when deficits are found, they may be attributable to other comorbid disorders. (*JINS*, 2001, *7*, 102–111.)

Keywords: Tourette syndrome, Executive function, Attention deficit/hyperactivity disorder, Verbal fluency, Verbal learning

INTRODUCTION

Research involving neurobehavioral functioning in children with Tourette syndrome (TS) highlights the need to consider the presence of Attention-Deficit/Hyperactivity Disorder (ADHD), as children with TS have comorbid ADHD in 50–60% of the cases (Spencer et al., 1998; Yeates & Bornstein, 1994). When these disorders have been studied separately, psychoeducational findings among children with TS or ADHD have not uncovered impairments in general intellectual functioning (Singer et al., 1994); nevertheless, there has been consistent evidence of executive function (EF) deficits in both these disorders with behaviors implicating prefrontal/subcortical system involvement (Denckla & Reiss, 1997).

Evidence from imaging studies clarifies some of these behavioral differences in children with TS and ADHD. Quantitative magnetic resonance imaging (MRI) studies have found that children with TS have a shift away from normal left-larger-than-right asymmetry of the putamen and lenticular regions of the basal ganglia (Peterson et al., 1993; Singer et al., 1993), as well as a larger-than-usual corpus callosum

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(Baumgardner et al., 1996). Among children with ADHD, a convergence of structural MRI evidence shows abnormality in striatal systems, including reduced size of the left caudate (Hynd et al., 1993), right caudate (Castellanos et al., 1994), right globus pallidus (Castellanos et al., 1996), and smaller left globus pallidus (Alyward et al., 1996). Decreased size of frontal regions has also been a consistent finding in ADHD, including bilateral frontal volumes (Hynd et al., 1990), right anterior frontal volumes (Castellanos, et al., 1996), right anterior superior white matter (Filipek et al., 1997), and right dorsolateral prefrontal cortex (Hill et al., 1999). In contrast to the evidence of larger corpus callosum in children with TS, those with ADHD have been found to have decreased size of the corpus callosum relative to controls (Baumgardner et al., 1996).

Functional imaging methods also point to frontal-striatal differences in TS and ADHD. TS groups have been shown to have hypoperfusion of basal ganglia and frontal lobes (Hall et al., 1991) and decreased blood flow to left lenticular regions (Riddle et al., 1992) on single photon emission computed tomography (SPECT), as well as bilateral increase in glucose utilization in basal ganglia and frontal regions (Baxter & Guze, 1993) on positron emission tomography (PET). Using functional MRI, Vaidya and colleagues (Vaidya et al., 1998) found greater frontal activation along with reduced striatal activation in unmedicated ADHD children performing a “go–no-go” task, compared with controls. Of importance is the fact that there has been an overrepresentation of males in many of these studies, and the differences observed may be due, in part, to hormonal factors instrumental in the development of brain asymmetries in males. When more proportionate numbers of boys and girls are in the studies, there are fewer differences between clinical groups and controls, particularly when comparing TS-only groups to controls (Schuerholz et al., 1998).

Because disruption in the development of frontal-striatal circuits has been implicated in both TS and ADHD, further investigation of these systems is warranted in understanding the EF deficits in these disorders. The striatum includes three major subdivisions (i.e., caudate, putamen, and ventral striatum) and is distinguished by circuits that share the frontal lobes, globus pallidus, and thalamus (Alexander et al., 1986; Cummings, 1993). These circuits link specific regions of the frontal lobes to subcortical structures, and provide the framework for understanding the neurobiological relationship between TS and its comorbid disorders (Singer, 1994). In order to link frontal-striatal circuits to behaviors associated with EF, Heilman and colleagues (Heilman et al., 1991; Heilman, 1994) described the striatum as acting to gate sensation into two systems: “how” (organization and praxis) and “when” (i.e., response inhibition).

Neuropsychological investigations of children with TS have shown differences, relative to controls, in systems related to Heilman’s concept of the “how” action of the striatum. In particular, children with TS demonstrate selected deficits in EF including slow and variable reaction time on continuous performance tests (Harris et al., 1995; Shucard et al., 1997) and reduced letter-word fluency (Schuerholz et al., 1996; Sutherland et al., 1982). This reduced output in TS has not been found to be related to motor slowing, but rather to mental slowing or “bradyphrenia,” which has long been considered a feature of subcortical dysfunction and associated with disorders such as Parkinson’s disease. Since motor slowing was not responsible for slowed productivity in children with TS, it was argued that the frontal-striatal circuits responsible for the “how” function, particularly those involving the striatum and dorsolateral aspects of the prefrontal cortex, may be deficient (Schuerholz et al., 1997). Indeed, investigations with adults have demonstrated that damage to systems involving the dorsolateral prefrontal cortex leads to a dysexecutive syndrome involving deficits in planning, organization, and judgment, with specific neuropsychological deficits on tasks of fluency (Duffy & Campbell, 1994; Mega & Cummings, 1994).

ADHD has been associated with EF deficits involving both the “how” and the “when”striatal-frontal systems. Compared to controls, children with ADHD demonstrate not only slow and variable reaction times on continuous performance tests, but also increased commission errors (Harris et al., 1995; Levy & Hobbes, 1997). Recent research cites evidence of organization and planning deficits in children with ADHD, as measured by tasks involving concurrent memory search and output (Rey Osterrieth Complex Figure, Letter Word Fluency), flexibility of thinking (Wisconsin Card Sorting Test; Pineda et al., 1999), and strategic recall on new learning tasks (Cornoldi et al., 1999). Similar deficits in children with ADHD were found on the organization score of the Rey Osterrieth Complex Figure, but not on the Wisconsin Card Sorting Test or the Letter Word Fluency (Reader et al., 1994). Children with ADHD also showed increased levels of choreiform movements, relative to controls (Schuerholz et al., 1997).

A potential drawback among existing clinical measures of EF, particularly those involving organized search and efficient production of responses (e.g., fluency, recall, and learning measures), is the interpretation of the total outcome score, without directly assessing the response organization strategies (i.e., “how”) used to arrive at a score, or the interfering behaviors (“when”) which may impede output. Carefully observing these process variables is particularly salient in TS and ADHD, and inefficient use of strategies and disinhibition can be considered markers for executive dysfunction in children with a wide range of neurodevelopmental disorders (Barkley, 1997).

Measures of fluency (verbal, design, and figural) are commonly used in clinical practice with children and adults, and are thought to represent behavioral demands involving organized memory search and sustained production (i.e., the “how” system). Verbal fluency measures are further divided into letter and semantic tasks, and poor performance on letter fluency relative to semantic fluency is considered evidence for executive dysfunction (Denckla, 1994). Design and figural fluency tasks have been developed as nonverbal analogs to the verbal fluency measures (DeMakis &

Harrison, 1997) and investigated in a variety of clinical populations (Lezak, 1995). Using PET, Elfgren and Risberg (1998) found increased left frontal activation during a letter fluency task and bilateral frontal activation during a design fluency measure, suggesting differences in cortical areas engaged under the different task demands. On verbal fluency and learning measures, healthy individuals have been shown to spontaneously use phonemic "clustering" (i.e., grouping words by initial sound blends or words with similar phoneme patterns) significantly more than individuals with known frontal lobe dysfunction (Troyer et al., 1997). Similarly, reduced semantic clustering (i.e., grouping words by category) has been found in adults with individuals with closed head injury (Levin & Goldstein, 1986), documented frontal lobe lesions (Gershberg & Shimamura, 1995), Alzheimer's disease (Troyer et al., 1998), and schizophrenia (Robert et al., 1998); however, similar patterns with children have been less clear (Beebe et al., 2000). While reduced total output in design fluency has been documented in patients with obsessive-compulsive traits (Mataix-Cols et al., 1999), high functioning autism (Turner, 1999), and traumatic brain injury (Varney et al., 1996), little work has been done to clarify the process aspects of the deficits found in these groups. Process variables involving intrusions and commission errors (i.e., the "when" system) are more commonly considered in studies of fluency and learning. These more direct observations of performance have been used to clarify the behavioral strengths and weaknesses in children with both ADHD and learning disabilities (Denckla, 1996). On list learning recall trials, abnormally high rates of free recall intrusions have been found in individuals with left frontal cortex lesions (Parkin et al., 1996), reading disorders (DeBeni et al., 1998), and in polydrug abusers (Heishman et al., 1999).

The present investigation sought to clarify the EF differences in children with either ADHD or TS (without ADHD), and to expand the operational definition of EF in these groups to include Heilman's concepts of the "how" (organization) and "when" (inhibition) actions of the striatum and associated prefrontal systems. Specifically, two hypotheses were made. First, given the previous neuropsychological and imaging findings, it was hypothesized that children with TS would show deficits, relative to controls, on measures of response preparation ("how"), due to slowing in organized search and retrieval, while children with ADHD would show deficits, relative to controls in both response preparation ("how") and disinhibition ("when") measures. Second, it was hypothesized that group differences (i.e., effect sizes) among the three groups (TS, ADHD, and controls) would be more pronounced when comparing process variables (e.g., clustering, intrusions) on fluency and recall tasks, than group differences on the more traditional total-score-outcome variables. Third, because both organization and inhibition are affected, it was hypothesized that children with ADHD would show more overall EF deficits, relative to controls, than children with uncomplicated TS on measures of fluency and list learning.

METHODS

Participants

Seventy-four children (45 boys, 29 girls) were research participants for this study. These children were participants in a larger study (Neurodevelopmental Pathways to Learning Disabilities, NS-25806) at the Kennedy Krieger Institute. Children were included in the study if they were between the ages of 6 and 16, free from a history of seizures, head injury, or other neurologic illness. All participants had Full Scale IQ (WISC-III or WAIS-R) of 80 or above (range 82–146). The sample was predominantly Caucasian (93%) and right-handed (93%). Three diagnostic groups were formed for the analyses. There were a total of 21 children in the ADHD-only group, 25 in the TS-only group, and 28 controls. Children with comorbid TS and ADHD were excluded from the current study. None of the children in the TS-only group had comorbid Obsessive Compulsive Disorder (OCD), while only one of the children in the ADHD group had been diagnosed with OCD.

Diagnosis of TS was made by a pediatric neurologist and director of the Johns Hopkins Tourette Syndrome Clinic (HSS), on the basis of the Tourette Syndrome Classification Group (1993) criteria. In order to be included in the TS group, children had to manifest all the following symptoms: (1) onset of tic symptoms before age 21; (2) multiple motor tics; (3) one or more vocal tics; (4) tic frequency which changes over time; (5) duration of tic symptoms greater than 1 year; (6) tics not secondary to other medical conditions; and (7) tics are witnessed by a reliable observer. Overall tic severity was reported to be mild to moderate in the TS-group sample, although individual measurement of tic severity was not obtained. Diagnosis of ADHD was made after participants met the following criteria: (1) identification and referral by professionals (psychologists, psychiatrists, pediatricians, neurologists) in the local community as having a current diagnosis of ADHD; (2) independent DSM IV diagnosis of ADHD (any type) based on interview with a licensed psychologist or child neurologist; and (3) parent rating of 2 or higher (on a 4-point Likert Scale ranging from 0 to 3) for six of nine items assessing inattention and/or six of nine items assessing hyperactivity/impulsivity on the ADHD Rating Scale (DuPaul, 1991; DuPaul et al., 1998). The ADHD group included eight children with Predominantly Inattentive and 13 children with either Hyperactive-Impulsive or Combined Type ADHD, as defined by pattern of caregiver responses on the ADHD Rating Scale. Control group participants included unaffected siblings from Fragile X, Neurofibromatosis Type 1, and Turner Syndrome projects studied at the Kennedy Krieger Institute Learning Disabilities Research Center ($n = 13$), as well as participants recruited as controls ($n = 15$). None of the participants in any of the groups were on stimulant or tic-suppressing medication at the time of testing. All participants were assessed for presence of learning disabilities in reading and mathematics. For the present study, a learning disability was defined as a 1.5 standard deviation discrepancy between WISC-III Full Scale IQ (FSIQ) and achievement on the Reading or Math Composite from the Wechsler Individual Achievement Test (WIAT; Wechsler, 1992). Only two participants in the sample had this discrepancy. One child with ADHD had a discrepancy of 24 points between Math Composite score and FSIQ; however, the child's Math Composite was still in the average range for age (standard score = 98). One child in the TS group had a 23-point discrepancy between Reading Composite and FSIQ, but the Reading Composite standard score was 101. Since both children had all achievement scores at least in the average range, they were retained in the sample.

Among the ADHD subjects, there were no significant differences between children with Inattentive ADHD and those with Hyperactive or Combined ADHD on FSIQ $[F(1,19) = .01$, $p = .93]$ or age $[F(1,19) = .32$, $p = .58]$. Among control subjects, there were no significant differences between subjects recruited as controls and control subjects who were unaffected siblings of other research participants when compared on FSIQ $[F(1,26) = .04$, $p = .85]$, age $[F(1,26) = .003$, $p = .96]$, or Child Behavior Checklist (CBCL; Achenbach, 1991) Attention Problems score $[F(1,26) = .01$, $p = .92]$. When comparing clinical groups (i.e., ADHD, TS, and controls), there were no significant differences between group differences in Full Scale IQ $[F(2,71) = 1.2$, $p < .31]$ or age $[F(2,71) = 1.6$, $p < .22]$. There was an expected between-group difference on the CBCL Attention Problems score $[F(2,70) = 69.7$, $p < .00001]$, with *post-hoc* tests (Tukey) revealing significantly greater scores for the ADHD group than either TS group ($p < .00001$) or the control group ($p < .00001$). The TS group had significantly higher CBCL ratings than the control group ($p < .005$), although their mean score on this scale (55.6) was well within the average range. Means for demographics, FSIQ, and CBCL are among the three groups are presented in Table 1.

Table 1. Mean scores for FSIQ and distribution of age and gender[a]

	ADHD	TS-only	Control	Total
N	21	25	28	74
Mean Age (years)	11.7	10.2	10.8	10.9
FSIQ	109.5	109.2	114.0	111.1
% Caucasian	90	92	96	93
% Right Handed	81	100	96	93
# with LD	1	1	0	2
Gender				
Girls	7	6	16	29
Boys	14	19	12	45
CBCL[b]	68.5	55.6	50.6	57.4

[a]*Note. FSIQ: WISC-III or WAIS-R Full Scale IQ; CBCL*: Achenbach Child Behavior Checklist Attention Problem T-Score.

[b]ANOVA $p < .000001$. *Post-hoc* tests (Tukey HSD) reveal that ADHD > TS ($p < .0001$), ADHD > control ($p < .0001$), and TS > control ($p < .005$).

Neuropsychological Assessment

Neuropsychological variables were selected from among measures administered as part of a larger project, with respect to two domains of EF: response organization and inhibition errors. The variables were chosen to highlight available process variables available in commonly used neuropsychological tests. To assess response organization, the following scores were obtained: (1) California Verbal Learning Test for Children (CVLT-C; Delis et al., 1994) semantic clustering (raw score total trials 1–5); (2) Ruff Figural Fluency (FF) Test (Ruff et al., 1994) total figures produced by using sequential strategies; (3) Semantic Word Fluency (SWF; animals/foods) total words in the two trials related by semantic clustering; and (4) Letter Word Fluency (LWF) total words in the three trials related by phonemic clustering. The LWF trials used the letters F, A, and S for stimuli. Measures of clustering on both the word fluency tasks were obtained using the method described by Robert et al. (1998). Measures of inhibition error included: (1) CVLT-C intrusions (raw score total, trials 1–5); (2) Semantic Word Fluency total errors (# intrusions + # repetitions); (3) Letter Word Fluency total errors (# intrusions + # repetitions); and (4) Figural Fluency errors (# rule breaks + # repetitions).*

All participants in the study completed the measures as part of a battery of psychoeducational and neuropsychological testing. Evaluators were blind to subject diagnosis, and were asked to comment on the level of cooperation of the participants. None of the children in the study were noted to have tic behavior or hyperactivity which interfered with test validity. In a small number of cases, children from the

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**Calculation of Word Fluency Clustering*: Calculation of cluster scores was made using an adaptation of the procedures described by Robert et al. (1998).

Semantic Clustering: Clusters were defined as groups of contiguous words belonging to the same semantic subcategory, such as farm animals, birds, fish, or sea mammals; or related food groups. Associations were considered semantic clusters only when at least three consecutive words were semantically related. One exception to the rule is that a semantic cluster was scored for the words "cat" and "dog" produced simultaneously.

Example: *dog*, *cat*, monkey, *hen*, *rooster*, *goose*, whale, *fly*, *cockroach*, *beetle*, snake, *pigeon*, *seagull*, *owl*, *canary*. # words related by cluster = 12.

Letter Word Clustering: Clusters were defined as groups of contiguous words that begin with the same two letters (e.g., fry, friend, frantic) or that differed by only a vowel sound (e.g., sat, set, sit). In the case of contiguous homophones (e.g., son, sun), two words were sufficient to score a formal cluster.

Example: sip, *sat*, *sap*, *sand*, *sack*, still, *spot*, *spit*, *spill*, soak, snow, *so*, *sew*, salad. # words related by cluster = 9.

Calculation of Figural Fluency Clustering: The clustering score in Figural Fluency was obtained by counting the number of figures contained in different strategies. Two distinct strategy types were used—rotational and quantitative (Vic & Ruff, 1988). To be considered a strategy, at least three consecutive figures must be systematically rotated or quantitatively changed, and figures contained in each strategy continue the established pattern. Repetitions and errors were not included in any strategies.

Scoring Procedures: All clustering scores for the fluency measures were scored twice to check scoring validity. Scorers were the first two authors (E.M.M. and C.W.K.), who were blind to subject diagnosis when scoring; however, in approximately 5% of the sample, hyperactivity and/or tic behavior was noted by the examiners.

Table 2. Correlations between response organization and disinhibition measures[a]

	1	2	3	4	5	6	7
1. CVLT-C Clus							
2. CVLT-C Int	−.23						
3. SWF Clus	**.25**	−.04					
4. SWF Err	−.03	−.06	.00				
5. LWF Clus	**.27**	−.12	**.27**	.02			
6. LWF Err	.00	.11	**.28**	.12	.16		
7. FF Clus	**.43**	−.16	**.46**	−.03	**.41**	.11	
8. FF Err	−.19	−.09	.05	.21	.03	.00	−.03

[a] *CVLT-C Int*: Intrusion errors trials 1–5; *CVLT-C Clus*: Semantic clustering raw score, trials 1–5; *SWF Err*: Semantic Word Fluency total errors; *SWF Clus*: Semantic Word Fluency number of words related by clustering; *LWF Err*: Letter Word Fluency total errors; *LWF Clus*: Letter Word Fluency total words related by phonemic clustering; *FF Err*: Figural Fluency total errors; and *FF Clus*: Figural Fluency total designs drawn within clusters of strategies. Correlations in boldface are significant at $p < .05$.

TS group demonstrated tic behavior during testing that may have cued the examiner about the child's diagnostic group.

RESULTS

Chi-square analyses were performed for gender between the ADHD, TS, and control groups. There was a significant between-group difference for gender ($\chi^2 = 6.5$, $p < .05$), indicating the different proportions of males to females between the three groups, with the ADHD and TS groups having more males and the control group having more females. However, there were no significant gender differences on FSIQ, age, or CBCL Attention Problems Scale. Comparisons between groups revealed no significant gender differences on any of the eight process variables (clustering or inhibition errors). For total score variables from the CVLT-C, SWF, LWF, and FF, the only significant gender difference was for SWF z-score [$t(72) = -2.5$, $p < .02$], with females performing better than males on that task. Using multivariate analysis of variance (MANOVA) assessing the interaction between the group and gender on the four total score variables, the overall multivariate interaction effect (Pillai's) between group and gender was not significant ($p = .55$).

A total of eight raw score process variables were used for the primary analyses. Correlations between the eight process variables are listed in Table 2. Overall, the intercorrelations between the process measures were low to moderate. Mean intercorrelation among the clustering variables was moderate (.30), while there was little correlation among the intrusion variables (mean = .01). In general, clustering variables were more correlated with other clustering variables than they were with intrusion variables derived from the same measure. For example, there were significant correlations between FF clustering and CVLT-C clustering (.43), SWF clustering (.46), and LWF clustering (.41), but not between FF clustering and FF errors (−.03). Similarly, there were significant correlations between LWF clustering and CVLT-C clustering (.27), SWF clustering (.27) and FF clustering (.46), but not between LWF clustering and LWF errors (.16). A similar pattern was not found when analyzing intercorrelations among the intrusion variables.

Because the eight derived process variables are not normally distributed, nonparametric statistics (Kruskal Wallace Tests) were used to compare group differences. The results are listed in Table 3. Bonferroni correction for number of comparisons was made in determining significance level (Bonferroni corrected $\alpha = .05/8$ or .00625). There were no significant group differences among the four response organization (clustering) variables. For the inhibition error variables, there was a significant group difference ($p = .002$) for CVLT-C intrusions. *Post-hoc* tests (Mann-Whitney U) for the CVLT-C intrusions revealed that both the ADHD group ($p = .002$) and the TS group ($p = .002$) had a greater number of intrusion errors than controls. There were no significant differences between the ADHD and TS groups on CVLT-C intrusions.

Table 3. Means and standard deviations for process variables[a]

	ADHD	TS	Control	χ^2	p
CVLT-C Clus	48.2 (9.6)	47.6 (12.0)	47.8 (10.0)	0.14	.93
SWF Clus	18.0 (8.2)	17.1 (7.7)	17.3 (9.5)	0.35	.84
LWF Clus	3.7 (3.8)	3.5 (3.7)	5.2 (4.9)	1.60	.44
FF Clus	5.2 (7.3)	7.6 (10.6)	13.1 (24.0)	0.25	.88
CVLT-C Int	2.5 (2.6)[a]	2.4 (2.5)[a]	0.7 (1.3)[b]	12.00	.002
SWF Err	1.7 (2.5)	1.2 (1.2)	1.3 (1.9)	0.89	.64
LWF Err	2.3 (2.0)	1.7 (1.7)	1.9 (1.9)	1.30	.51
FF Err	12.9 (19.6)	6.4 (7.6)	9.5 (20.0)	1.40	.50

[a] *CVLT-C Clus*: Semantic clustering raw score, trials 1–5; *SWF Clus*: Semantic Word Fluency number of words related by clustering; *LWF Clus*: Letter Word Fluency total words related by phonemic clustering; *FF Clus*: Figural Fluency total designs drawn within clusters of strategies; *CVLT-C Int*: Intrusion Errors trials 1–5; *SWF Err*: Semantic Word Fluency total errors; *LWF Err*: Letter Word Fluency total errors; *FF Err*: Figural Fluency total errors. χ^2: Kruskal Wallis Test; Standard deviations are listed in parentheses. Significance calculated based on a Bonferroni-corrected error rate (.05/8 = .00625); *Post-hoc* tests (Mann Whitney U): $a > b$ ($p = .002$).

Table 4. Means and standard deviations for total scores on neuropsychological measures[a]

	ADHD	TS	Control	p	Power
CVLT-C Total T	50.5 (10.8)	53.4 (8.6)	51.3 (13.1)	.65	.11
SWF Total z	0.20 (0.7)	0.59 (0.8)	0.52 (0.8)	.18	.36
LWF Total z	−0.38 (1.0)	−0.19 (1.4)	0.35 (1.0)	.14	.40
FF Total*	54.1 (19.0)	57.6 (23.6)	59.2 (28.1)	.12	.08

[a] *CVLT-C Total T*: T-score for trials 1–5 total; *SWF Total z*: Semantic Fluency total correct *z*-score; *LWF Total z*: Letter Word Fluency total correct *z*-score; *FF Total*: Figural Fluency total correct raw score. *p*: Significance level for univariate ANOVA or ANCOVA*. For ANCOVA, age is used as covariate standard deviations in parentheses. Observed power computed using alpha = .05.

Total score performance on the three fluency (LWF, SWF, and FF) and one recall (CVLT-C) measure was compared among the three groups. Assumptions for parametric statistics were met for these variables. Multivariate analysis of variance (MANOVA) was used, comparing group differences among standard means for the following measures: (1) CVLT-C total correct T-score from trials 1–5; (2) SWF total correct *z*-score; and (3) LWF total correct *z*-score. Mean scores for these variables, along with power calculations for the analyses are listed in Table 4. There was no significant multivariate group effect (Pillai's) for these three variables ($p = .29$). Because means and standard deviations were not available for the Figural Fluency Test for this age range, an analysis of covariance was used (with age as covariate), comparing groups, for raw score totals (i.e., total number correct designs) on Figural Fluency. There was no significant group difference for Figural Fluency raw score totals. Despite nonsignificant group differences, there were trends for group differences for all three fluency variables. The failure to detect group differences among these variables may be due in part to the low statistical power of the analyses.

Effect-size calculations were derived for both process variables and total score variables, in order to provide estimates of population differences, and to clarify the relative contribution of each to understanding group differences. Effect sizes for group comparisons are listed in Table 5. Effect size is a standardized quantitative index that can represent the magnitude of change that one variable produces in another variable as reflected in the difference between two means (Cohen, 1988). Effect-size values were computed using the *d* statistic. Interpretation of the effect size *d* is based on a convention suggested by Cohen—.20 is considered a "small" effect size; .50 considered "medium," and .80 or greater, a "large" effect size. By computing and interpreting power, sample effect sizes are assumed to generalize to the population. In our sample, only the CVLT-C intrusions showed large effect sizes, both when comparing ADHD to controls ($d = .88$), and TS to controls ($d = .85$). A medium-to-large effect size was observed when comparing the ADHD group to controls on LWF total *z*-score ($d = .73$), suggesting that, despite nonsignificant statistical differences on ANOVA, a replication study with larger sample sizes is likely to detect a true group difference which exists in the population. On the SWF total *z*-score, a medium effect size (.52) was found for the ADHD *versus* TS group comparison, also suggesting that group differences may be detected in a replication with greater statistical power. Because the TS and ADHD groups had similar proportions of males, the SWF effect is not likely due to gender differences.

Analysis of effect sizes also allows for comparison of clustering variables (i.e., the "how" system) to inhibition error variables (i.e., the "when" system) for their ability to demonstrate group differences among children with ADHD or TS and controls. In our analyses, the clustering variables, across measures, demonstrated poor ability to detect group differences, with no effect sizes in the medium or large range (mean effect size = .13; range = −.08 to .45). In contrast, both the intrusion error variables (mean effect size = .25; range = −.22 to .88) and total score variables (mean effect size = .25; range = −.19 to .73) yielded some medium and large effect sizes, with more consistent patterns (control > TS > ADHD) of performance among the three groups on most of the measures.

Table 5. Effect size (d) for comparisons between clinical groups and controls[a]

	ADHD vs Control	TS vs Control	ADHD vs TS
CVLT-C Clus	−.04	.02	−.06
CVLT-C Int	**.88**	**.85**	.04
CVLT-C Total T	.07	−.19	.30
SWF Clus	−.04	.02	−.08
SWF Err	.18	−.06	.25
SWF Total z	.42	.09	**.52**
LWF Clus	.34	.39	−.01
LWF Err	.21	−.11	.32
LWF Total z	**.73**	.44	.16
FF Clus	.45	.30	.26
FF Err	.17	−.20	.44
FF Total	.21	.06	.16

[a] *CVLT-C Clus*: Semantic clustering raw score, trials 1–5; *CVLT-C Int*: Intrusion errors trials 1–5; *CVLT-C Total T*: T-score for trials 1–5; *SWF Clus*: Semantic Word Fluency words related by clustering; *SWF Err*: Semantic Word Fluency total errors; *SWF Total z*: Semantic Fluency total correct *z*-score; *LWF Clus*: Letter Word Fluency total words related by phonemic clustering; *LWF Err*: Letter Word Fluency total errors; *LWF Total z*: Letter Word Fluency total *z*-score; *FF Clus*: Figural Fluency total designs drawn in clusters of strategies; *FF Err*: Figural Fluency total errors; and *FF Total*: Figural Fluency total correct raw score. Effect size d = (mean of clinical group − mean of comparison group)/pooled standard deviation of two groups). Medium and large effects in bold.

DISCUSSION

The present study represents an attempt to clarify the underlying mechanisms of action in existing neuropsychological measures of EF in children with ADHD or "pure" TS (i.e., without ADHD, OCD, or LD) who were not treated with medications at the time of assessment. Process variables selected to represent Heilman's (1994) concept of the "how" and "when" actions of the striatum were derived from existing measures of fluency and verbal learning, and compared with more traditional total-score-outcome measures. We have three primary findings. First, significant group differences were found on only one process variable (CVLT-C intrusions), with both the ADHD and TS groups having more intrusions than controls. Our hypotheses regarding deficits among children with TS or ADHD on measures of response organization (i.e., the "how" system) were not supported by the present data. In contrast, analysis of inhibition errors (i.e., the "when" system) was useful in demonstrating some clinical differences for both the ADHD and TS groups. Significant group differences on CVLT-C intrusions were found even when the total standard score variables were not significantly different. These findings provide some support for Barkley's (1997) beliefs about the salience of disinhibition as a process variable in models of EF for ADHD, and are consistent with prior hypotheses which asserted that the underlying mechanisms for tic behavior in TS may be related to a failure of an inhibitory system mediated by a central executive (Kane, 1994).

Second, our results provide some initial support for consideration of process variable constructs in addition to total score variables when interpreting EF performance *via* fluency and list learning measures. Although our groups did not differ on the clustering measures, there is evidence for the discriminant validity of the clustering/response organization construct across different neuropsychological tests. Because of the skewed distribution of these raw score variables, we were not able to use traditional factor analysis to statistically derive underlying constructs. However, in our analyses, there was often greater covariation among different measures of the same construct (i.e., clustering) than among process variables drawn from the same neuropsychological test (e.g., Figural Fluency). These results are generally consistent with the previous findings of a two-factor solution (orbitofrontal-inhibitory, dorsolateral-executive) among neuropsychological measures used to assess ADHD (Wozniak et al., 1998).

Third, our hypothesis that children with ADHD would show more "impairment" on EF measures than the TS-only group was not supported. Among the four neuropsychological tests used in the present investigation (CVLT-C, Letter Word Fluency, Semantic Word Fluency, and Figural Fluency), neither of our clinical groups showed performance outside the average range. Even when significant group differences or large effect sizes were obtained, mean scores for these groups remained in the average range, and as such the concept of EF "impairment" in our sample is not appropriate. One reason for the absence of impairment in the present measures is that the ADHD, TS, and control groups all had solidly average (in many cases high average or above) Full Scale IQ. Prior research from this center has suggested that EF deficits, as measured by performance on neuropsychological instruments, may not be detected in children with high average IQ and above (Mahone et al., 1999). In fact, among a different cohort of children with "pure" TS previously studied at our center, we found a higher-than-expected (i.e., compared to parents) FSIQ (i.e., mean = 117; Schuerholz et al., 1996). Full Scale IQ among the TS-only group in the present cohort was slightly lower (mean = 109), yet still at the high end of the average range. The apparent "drop" in mean IQ score among the TS groups can be explained, in part, by the administration of the WISC-III in the present study, compared with the WISC-R in the prior study. Reviews comparing the WISC-R to the WISC-III showed that Full Scale IQ scores on the WISC-III average 5–6 points lower than those on the WISC-R (Zimmerman & Woo-Sam, 1997). Nevertheless, generalizability of the present findings to groups with lower IQ should be made cautiously.

In the present investigation, we studied children with "pure" TS, based on prior findings of executive dysfunction in this group. Based in part on recommendations from prior literature reviews (Pennington & Ozonoff, 1996), we excluded children from our TS sample who had comorbid ADHD, LD, or OCD. By excluding children with these comorbid conditions, we limited the sample size, which in turn, decreased statistical power of the group comparisons. In recruitment from a large, hospital-based Tourette Syndrome Clinic, we found the incidence of comorbidities for children with TS to be higher than previously reported (Yeates & Bornstein, 1994), and the presence of a "pure" TS condition to be less common.

Based on research evidence involving the slowing referable to the basal ganglia in children with TS or ADHD (Denckla & Reiss, 1997), it was argued that the basal ganglia may account for the EF differences which persist despite higher intellectual functioning in these groups. Indeed, previous research (Schuerholz et al., 1996; Sutherland et al., 1982) found lower performance on Letter Word Fluency among children with TS, relative to controls, which was thought to be sensitive to cognitive slowing and related to "linguistic bradyphrenia." In our sample, the Letter Word Fluency Test showed a strong effect size for the ADHD *versus* control group comparison, and a moderate effect size for the TS *versus* control comparison, even with all scores in the average range. In our sample, many of whom had above average IQ, the possibility of "overgrowing" (i.e., the cortex maturating to dominate subcortical deficits) may in fact be the mechanism by which these children continue to perform normally on the clustering/response organization measures. In our sample, 48% of the ADHD, 48% of the TS, and 69% of the control group had FSIQ scores of 110 or greater. Nevertheless, the present findings provide only limited support for the notion that TS, in the absence of other comorbid conditions, is associated with clinical impairments in the response organization/clustering aspect of EF, as measured by slowing on verbal memory search or inefficient organizing on recall and fluency measures. These findings are consistent with those of the Ozonoff et al. (1994) study with a "pure" TS population, which found no consistent evidence of EF deficits. Indeed, the group differences in the Schuerholz et al. (1996) research may have been related to the higher incidence of OCD in their TS-only sample, or to the fact that their group included almost all males. With more diagnosed OCD in the prior sample, there may have been more deleterious effects of the cognitive rigidity seen in that disorder. Tic severity among our TS sample was mild, and there were no reported instances of overt tic behavior interfering with test performance (e.g., fluency measures). The relationship between tic severity, intelligence, and EF deficits among children with TS remains an empirical question.

Number of intrusion errors on the CVLT-C appears to be a strong and robust measure of EF among children with TS or ADHD. In fact, presence of intrusion errors at all on the initial five trials may be a marker for EF disturbance. In our sample, 76% of the ADHD and 72% of the TS groups had intrusion errors on the CVLT-C first five trials, compared to 39% of controls. These group differences for TS and ADHD occurred despite the fact that both groups had intact overall verbal intelligence and absence of language-based learning disabilities. There was also a trend for group differences on LWF total scores, with a large effect size, suggesting that in replication studies with larger samples, the group differences in the population may be highlighted and significant group differences may be detected. Taken together, the findings of difficulties with fluency and disinhibition during linguistic memory search tasks implicate relative inefficiencies in left (compared with right) hemisphere frontal-striatal systems, and is consistent with prior imaging studies showing left-sided basal ganglia involvement (Alyward et al., 1996; Hynd et al., 1993; Singer et al., 1993). Additional investigation, particularly using functional imaging with measures such as verbal fluency and list learning is needed to clarify the underlying brain mechanisms used and the actual demands of these tests among children with ADHD and/or TS.

Several issues were not directly addressed in the present study and also warrant further investigation in their relationship with behavioral sequelae of TS, ADHD, and other disorders involving executive dysfunction. First, although attempts were made though group selection to include more females than in prior studies, gender effects were not entirely eliminated *via* sample sizes. Nevertheless, despite different proportions of males in the clinical *versus* control groups, there were no gender differences for age, IQ, or CBCL Attention scale ratings in our samples, nor were there interactions between gender and group on neuropsychological outcome measures. As such it is unlikely that the group differences we obtained were primarily a function of gender differences in the population.

Children with both inattentive and hyperactive varieties of ADHD were included in the ADHD group. Given the potential interaction between ADHD type and EF variables (particularly those involving inhibition errors), this relationship may have influenced or contributed to the lack of group differences on some of the outcome measures. Future studies with larger samples should continue to address ADHD type as a predictor of EF differences. Comorbidities for learning disabilities were minimized through sample selection. Participants were recruited for ADHD and tic symptoms, not for learning problems. Future research which incorporates larger samples and additional neuropsychological measures, and which correlates behavioral measures with structural and functional imaging, will contribute substantially to our understanding and treatment of the brain systems involved in TS and ADHD. In particular, research aimed at validating the constructs of response organization and inhibition as subcategories of EF, through functional imaging, are indicated. Further clarification of gender differences, effects of IQ, and effects of pharmacotherapy (particularly pharmacological agents known to affect executive functions) in the development of executive functions in children are indicated.

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