Research Article

IMPAIRED EXECUTIVE FUNCTIONING IN PEDIATRIC TRICHOTILLOMANIA (HAIR PULLING DISORDER)

Christopher A. Flessner, Ph.D.,* Elle Brennan, B.A., Yolanda E. Murphy, B.S., and Sarah Francazio, B.A.

Background: *No neurocognitive examinations of pediatric trichotillomania (hair pulling disorder; HPD) have taken place. As a result, science's understanding of the underlying pathophysiology associated with HPD in youths is greatly lacking. The present study seeks to begin to address this gap in the literature via examination of executive functioning in a stimulant-free sample of children with HPD.* **Methods:** *Sixteen and 23 children between 9 and 17 years of age meeting DSM-5 diagnostic criteria for HPD or classified as a healthy control, respectively, were recruited (N = 39) to complete structured interviews, self-reports, and a subset of tests from the Cambridge Automatic Neurocognitive Test Assessment Battery (CANTAB) assessing cognitive flexibility/reversal learning (intradimensional/extradimensional; IED), working memory (spatial span; SSP), and planning and organization (Stocking of Cambridge; SOC).* **Results:** *Hierarchical regression analyses indicated that, after controlling for appropriate covariates, diagnostic status predicted impaired performance on both the IED (reversal learning only) and SOC (planning and organization) but failed to predict cognitive flexibility or working memory capacity. Correlational analyses revealed that pulling severity was strongly related to working memory capacity, while disparate relationships between pulling styles (automatic, focused pulling) were evident with respect to working memory and planning and organization.* **Conclusions:** *Children with HPD performed more poorly on tasks of executive functioning as compared to controls. Correlational analyses suggest potentially distinct pathophysiology underlying automatic and focused pulling warranting further research. Limitations and future areas of inquiry are discussed. Depression and Anxiety 33:219–228, 2016.*

Key words: *pediatric; children; trichotillomania; hair pulling; executive functioning*

INTRODUCTION

Trichotillomania, also known as hair pulling disorder (HPD), affects approximately 1–3% of the population in the United States,[1,2] though this may be an underestimate,[3] and commonly first develops during childhood (11–13 years).[4] HPD is associated with significant impairment in functioning across several domains including psychological (i.e., emotional distress, lack of confidence), economic (i.e., work disruption), medical (i.e., scarring, trichobezoars), social (i.e., embarrassment), and cognitive (i.e., impulsivity).[5,6] Though no longitudinal research exists within the HPD field, it is possible that such difficulties may per-

Department of Psychological Sciences, Kent State University, Kent, Ohio

*Corresponding to: Christopher A. Flessner, Department of Psychological Sciences, Kent State University, Kent, OH 44242.
E-mail: cflessne@kent.edu
Received for publication 6 March 2015; Revised 27 September 2015; Accepted 9 October 2015

DOI 10.1002/da.22450
Published online 18 November 2015 in Wiley Online Library (wileyonlinelibrary.com).

sist throughout one's lifetime and, in turn, negatively impact ones development. Despite this potential for negative sequelae, a paucity of research on pediatric HPD exists.

With the advent of the fifth version of the Diagnostic and Statistical Manual (DSM)[7], HPD is now classified alongside other obsessive compulsive and related disorders (OCRDs), including obsessive compulsive disorder (OCD), hoarding disorder (HD), pathological skin picking (PSP), and body dysmorphic disorder (BDD). Each of these disorders appears to share common traits such as repetitive or ritualistic behaviors, and some theorize that the cluster of disorders may share a common etiology.[8–13] However, HPD has previously been classified as an impulse control disorder,[14] which may also capture important facets to the disorder. To this end, HPD may exist along a continuum of impulsivity and compulsivity, with facets of the disorder falling under each description.[13,15,16] However, the underlying pathophysiology of the disorder remains poorly understood. This is particularly true as it relates to pediatric HPD which, to this point, has rarely been be studied. Addressing this limitation to prior research is the primary focus of the present investigation.

What literature exists within the realm of research on adults and children with OCRDs, such as HPD, suggests that these repetitive and ritualistic behaviors may be due to failures within the corticobasal ganglia circuitry.[13,17–20] From the perspective of adults with HPD, available research has revealed impaired performance in nonverbal memory, executive functioning, spatial processing, motor responses, and divided attention.[21–28] For example, Keuthen et al.[25] demonstrated that performance on tasks of executive functioning may be correlated with decreased ability to resist the impulse to pull, while Rettew et al.[27] hypothesized that worsened symptom severity and decreased response to psychopharmacological treatment may be indicative of poorer spatial processing. Adults with HPD have also been found to perform worse on a task of cognitive flexibility (e.g., the ability to switch attention from one task to another) as compared to healthy controls,[21] though still other studies have demonstrated contradictory findings within this domain of cognitive functioning.[23,29] Chamberlain et al.,[24] utilizing a comorbidity-free sample of adults with HPD, found deficits in spatial working memory but not within other cognitive domains (i.e., learning, affective processing, decision making, impulsivity). These findings suggest that deficits in executive functioning are likely to be characteristic of adults with HPD, yet the precise domain(s) (i.e., set shifting, planning and organization, working memory) in which this impairment exists is unclear. This is particularly true of pediatric HPD as only one prior study has examined cognitive functioning (i.e., motor inhibition) within this sample[30]—a fact that is particularly unsettling considering that the disorder most commonly first develops during late childhood and early adolescence.[4,31] The current study seeks to address this gap in the extant literature by examining executive functioning in youths diagnosed with HPD.

A secondary aim of the current study is to explore potential differences in executive functioning between two unique pulling styles identified in both children and adults with HPD.[32,33] Automatic pulling is characterized by pulling outside of one's conscious awareness, while focused pulling is described as being more intentional, directed, and possibly in response to negative affect or private experiences (i.e., pulling to reduce tension, depressive symptoms, etc.). Although the majority of patients (children and adults) who pull their hair experience both styles of pulling to varying extents, the functional differences between these styles of pulling suggest that disparate pathophysiological underpinning may be associated with each. Elucidation of this line of inquiry may ultimately lead to improvements in interventions designed for a specific style of pulling. For example, recent research examining attentional bias modification within anxious youths and cognitive remediation for attention-deficit hyperactivity disorder among children (i.e., treatment targeting executive functioning and attention problems) has demonstrated some preliminary efficacy in both improving the cognitive deficits associated with these disorders and improving illness severity.[34–36] Similar approaches, targeting those areas of cognitive functioning shown to be impaired among youths with HPD, may hold similar promise and advance the field's ability to successfully treat this sometimes debilitating disorder.

Presently, science's understanding of the underlying pathophysiology and cognitive functioning associated with HPD in youths is lacking, despite that such research—replicated and completed by multiple groups—may significantly inform future therapeutic and, ultimately, preventive treatments (i.e., identification of cognitive markers predictive of future development of HPD or other related clinical phenomena). Prior research has revealed mixed findings with respect to specific cognitive domains of impairment in adults with HPD, yet these findings do suggest deficits with respect to executive functioning (broadly defined). Most germane to the current study, several of these prior studies have demonstrated impairments in relation to both working memory and cognitive flexibility among adults exhibiting HPD or other OCRDs via administration of tasks from the Cambridge Automated Neurocognitive Test Assessment Battery (CANTAB).[24,37,38] Based upon this prior work, we hypothesize that children with HPD will perform poorer on selected tasks of executive functioning, as tested by the CANTAB (i.e., intra/extradimensional set shifting, Stockings of Cambridge, spatial span) than healthy controls. In an exploratory aim, we seek to explore whether worse pulling symptomatology is correlated with poorer executive functioning as well as whether particular pulling styles are more strongly correlated with particularly facets to cognitive functioning.

TABLE 1. Demographic characteristics from the present sample ($N = 39$)

	HPD ($N = 16$)	HC ($N = 23$)
Gender (female)	9 (56.3%)	10 (43.4%)
Age	M = 11.3 years; SD = 1.7 years	M = 12.3 years; SD = 2.6 years
Race (Caucasian)	15 (93.8%)	20 (87.0%)
Household income (modal)	$60,000+	$60,000+
Comorbid diagnoses		
GAD	4	—
Social anxiety	1	—
Specific phobia	1	—
ADHD	5	—
Oppositional defiant disorder	2	—

MATERIALS AND METHODS

PARTICIPANTS

This study was approved by the Kent State University Institutional Review Board. Participants were recruited throughout Northeast Ohio via newspaper advertisements, fliers, and letters to pediatricians and schools as part of a larger ongoing study examining biological and psychosocial risk factors for pediatric anxiety and related problems. For the purposes of the current study, participants were included in all subsequent analyses if the child (1) was between 9 and 17 years of age, (2) met DSM-5 diagnostic criteria for HPD or did not meet diagnostic criteria for any psychiatric condition (healthy controls), (3) reported English as their primary language, (4) did not meet diagnostic criteria for a mood disorder, psychotic disorders, or autism spectrum disorder—as these disorders may have influenced performance on cognitive tasks described herein, (5) was not currently taking stimulant medication, and (6) completed all study measures (described below). For these analyses, a total of 16 children met DSM-5 criteria for a diagnosis of HPD while 23 were considered healthy controls—as assessed via clinical diagnostic interview. Demographic characteristics of this sample are presented in Table 1.

MEASURES

Kiddie-Schedule for Affective Disorders and Schizophrenia—Present and Lifetime Version (K-SADS-PL).[39] The K-SADS-PL is a structured interview designed to be administered to children and their parents. The interview possesses excellent interrater reliability and good concurrent validity.[40]

Anxiety Disorders Interview Schedule for DSM-IV: Child Version (ADIS-C).[41] The ADIS-C is a structured interview of pediatric psychopathology administered, for purposes of this study, jointly to the child and at least one parent. Prior research has documented the ADIS-C's reliability and validity.[42–44] Reflecting a minor change in study design (i.e., decision to switch to the ADIS-C, rather than K-SADS-PL, as our diagnostic measure of choice), some participants in this study were assessed using the ADIS-C.

Trichotillomania Diagnostic Interview (TDI).[6] The TDI is a semistructured interview designed to assess symptoms related to HPD and has been used extensively as a diagnostic measure of HPD in research focusing on children and adults with the disorder.[45] At the onset of this study, proposed changes to the diagnostic criteria for HPD had not yet taken place (i.e., removal of tension reduction and pleasure, gratification, or relief criterion for HPD). Although DSM-5 diagnostic criteria were used for entrance into the study described herein, all participants included in the present analyses would have also met DSM-IV-TR diagnostic criteria.

Trichotillomania Scale for Children—Child Version (TSC-C).[46,47] The TSC-C is a 12-item self-report measure of pediatric HPD severity. Prior research has documented this scale's strong internal consistency, test–retest reliability, and concurrent validity with other measures of HPD severity.[47] TSC-C total score was used as our measure of pulling severity.

Milwaukee Inventory for Styles of Trichotillomania—Child Version (MIST-C).[33] The MIST-C is a 25-item self-report measure designed to assess two disparate styles of pulling, automatic and focused pulling, resulting in two separate subscales. Each item is rated on a scale from 0 to 9. Higher scores indicate increasingly focused or automatic pulling. Both the focused ($\alpha = .90$) and automatic ($\alpha = .83$) scales have demonstrated strong psychometric properties in prior research.[33]

Child Behavior Check List (CBCL).[48] The CBCL assesses aspects of social skills, school functioning, and emotional and behavioral problems per parental report. The 112-item measure results in total, internalizing, and externalizing scores, as well as sub-scale scores. The CBCL has demonstrated a strong body of research supporting its reliable and valid use for children and adolescents.[49–51] Previous research has demonstrated that the CBCL and its subscales may demonstrate clinical utility as a diagnostic screening tool.[52,53] For the purposes of this study the attention and depressed/withdrawn subscale scores were utilized as dimensional measures of attention-deficit hyperactivity disorder (ADHD) (i.e., inattention, impulsivity) and depression-related symptoms, respectively—as has been recommended by other researchers.[54]

CAMBRIDGE AUTOMATIC NEURCOGNITIVE ASSESSMENT BATTERY (CANTAB)

The CANTAB is an assessment battery utilizing touch-screen technology to assess various domains of cognitive functioning. Below is a list of the CANTAB tasks that assess domains of executive functioning relevant to the present study.

Stockings of Cambridge Task (SOC). The SOC is a computerized test of spatial planning providing a measure of frontal lobe function. Within spatial planning, participants are presented with a display of colored balls in the top half of a screen and are instructed to copy or follow (see below) this pattern with similar balls in the bottom of the screen. As the task progresses, the number of moves necessary to copy the pattern increases from 2 to 5. Spatial planning is measured by the time taken to complete the pattern and the number of moves required. In total, this task involves four phases. In the initial and third phases, participants are instructed to move the balls in the bottom arrangement to match the pattern of balls in the top arrangement (copy). In the second and fourth phases of the task, the participant is instructed to move the balls in the bottom arrangement in a manner corresponding to the pattern of moves made in the top arrangement (follow). The relevant outcomes related to SOC include mean initial and mean subsequent think time. Initial think time represents the difference in time taken to select the first ball for the same problem under the copy and follow conditions (i.e., time taken to plan the problem's solution). Subsequent think time represents the difference in time between selecting the first ball and completing the problem for the same problem under the two conditions (copy and follow) and dividing this result by the number of moves made.

Spatial Span Task (SSP). The SSP is a computerized version of the Corsi Blocks task, which assesses working memory capacity and is sensitive to frontal lobe functioning. In this task, participants are presented with a pattern of nine boxes individually changing color one after the other, in a variable sequence. Upon hearing a tone, subjects are required to select the boxes in the same order in which they changed. As the task progresses, the number of changing stimuli increases from two to nine boxes. As such, the primary outcome measure for SSP is

the longest sequence length (i.e., spatial span length) the participant was able to complete.

Intradimensional/Extradimensional Task (IDED). The IDED is a computerized analogue of the Wisconsin Card Sorting test and is a test of set shifting/cognitive flexibility and reversal learning. In this task, the examinee is presented with two images. Each image contains a color-filled shape and white lines. The examinee chooses one of the images and receives feedback as to whether they were correct or incorrect, based on an unknown rule. The examinee is then presented with two new images, required to choose the correct image based on the feedback received in the previous trial, and again receives feedback as to their correctness. The examinee is considered to have established the rule after six consecutive correct responses (i.e., one block). Upon adequate demonstration of knowledge of the rule, the computer changes the rule requiring the examinee to mentally "switch" to the novel rule and respond according to feedback. At Block 6, the intradimensional set shift occurs. In Blocks 1 to 6, the rule is based on the pink, color-filled shape. Which pink shape is correct varies between blocks, but it is always based on the shape dimension. At Block 6, a novel set of shapes is presented and the examinee must apply the previous rule of shape to the novel shapes. At Block 7, the examinee must again apply the previously learned rule of shape but now must choose the shape that, in Block 6, was previously incorrect (i.e., intradimensional reversal learning). Block 8 constitutes the extradimensional shift stage. At Block 8, the examinee again is presented with novel shapes and lines, however, unlike in previous blocks, the rule depends on the line dimension rather than the shape dimension. Examinees are required to switch from the shape dimension rule they previously adhered to in the previous blocks, and apply a new line dimension rule. A final test block appears after Block 8 to test acquisition of the new line dimension rule. In this block, examinees are rewarded for choosing the pattern with the line that was previously incorrect in the preceding block (Block 9; i.e., extradimensional reversal learning). The primary outcomes of interests for the IED are total errors in Blocks 6 (intradimensional set shift), 7 (intradimensional reversal learning), 8 (extradimensional set shift), and 9 (extradimensional reversal learning).

PROCEDURES

Potential participants contact the lab and were provided with an overview of the methods, procedures, and aims of the study. A brief phone screen was conducted to determine the child's potential eligibility for the study. Potentially eligible children and one biological parent (i.e., the parent reported to spend the most time with the child) completed measures mailed in advance to their homes including demographic questionnaire (i.e., child age, presence/absence of perinatal events at birth, etc.) and several self- and parent-report measures. Upon arriving for their scheduled assessment parental consent and child consent/assent were obtained. A research assistant (RA), trained in the proper administration of the K-SADS-PL and ADIS by the first author (C.A.F.), administered the (semi) structured interview, and TDI jointly to the parent and child. Finally, all eligible children were asked to complete a neurocognitive test battery from the CANTAB including the SOC, SSP, and IDED.

Training of Evaluators. Prior to conducting diagnostic interviews, all study evaluators undertook identical and rigorous didactic (i.e., readings related to diagnostic interviews administered, viewing of previously administered interviews) and, subsequently, experiential training. With respect to experiential training, evaluators first observed administration of the K-SADS or ADIS-C by an expert in such administrations and licensed clinical psychologist (first author). Next, the evaluator was observed administering their own interview to a child and his/her parent(s)—with the first author present in the room and providing appropriate follow-up questions as necessary. Finally, the evaluator administered an interview while the first author observed from behind a one-way mirror. The evaluator met with the first author to discuss each assessment and conferred appropriate diagnoses during weekly supervision meetings.

Power Analyses and Data Analytic Plan. Power analyses were conducted based upon prior research examining executive functioning among adult HPD patients, as no prior research has examined cognitive flexibility, reversal learning, working memory, or planning and organization among a pediatric population. A review of these findings revealed effect sizes ranging from 0.26 to 1.05 (e.g., Grant et al.[29]; Bohne et al.[21]; Keuthen et al.[25]; Chamberlain et al.[24]) with an average effect size of 0.75. Consequently, a sample size of 34—using hierarchical regression—is necessary to detect a significant effect, if present. Thus, the sample size utilized for purposes of this study ($N = 39$) is sufficient to examine our primary aim.

A priori statistical analyses exhibited no statistically significant differences between the HPD and healthy control groups in relation to important demographic characteristics including age, gender, socioeconomic status, and race. Assumptions germane to the analyses conducted were examined and, if necessary, appropriate analytic modifications (i.e., transformation of outcomes variables to achieve normality; see Table 2) were performed. Preliminary analyses examining the relationship between relevant executive functioning outcomes and potential control variables—guided by findings from prior research among pediatric populations—were conducted for use in subsequent regression analyses including child age, gender, ADHD-related symptoms (i.e., scores on the CBCL attention scale), depressive symptoms (i.e., scores on the CBCL depression scale), anxiety symptoms (i.e., scores on the CBCL anxious-depressed scale), and report of perinatal events (yes/no) at child birth. When appropriate ($P \leq .01$), these covariates were controlled for in subsequent regression analyses. For example, child age was used as a control variable for only those regression analyses in which preliminary data suggested that it would be a relevant control variable.

A series of regression models were tested to examine whether a diagnosis of HPD was predictive of neurocognitive performance on the SSP, IED, and SOC. For each regression, appropriate control variables, when applicable and based upon our preliminary analyses (see above), were included in Step 1 and our dummy coded diagnostic variable (0 = healthy control, 1 = HPD) was entered into Step 2. Further, a series of correlational analyses were conducted to examine whether performance on the outcomes assessed via our regression analyses (see Measures section) were associated with HPD severity or pulling styles. As this study represents the first such examination of executive functioning within pediatric HPD, an α level of .05 was used to determine statistical significance for all analyses—unless otherwise noted.

RESULTS

Table 2 presents raw scores for participant performance on each of the outcome measures utilized for the purpose of this study. Further, findings from each of the regression models tested are presented in Table 3. What follows is an overview of findings in relation to the data presented in this latter table.

COGNITIVE FLEXIBILITY (IDED, BLOCKS 6 AND 8)

Results of our hierarchical regression analyses revealed that HPD status failed to predict performance on either total errors in Block 6 (intradimensional set shifting; $\beta = -.02$, $P = .913$), in isolation, or Block 8

TABLE 2. Performance (raw score) for participants with respect to SOC, SSP, and ID/ED outcomes on CANTAB

	HPD ($N = 16$)	HC ($N = 23$)
Set shifting		
Errors Block 6	0.50 (0.63)	0.52 (0.59)
Errors Block 8	7.81 (9.68)	7.48 (8.33)
Reversal learning		
Errors Block 7	1.81 (1.68)[a]	1.04 (0.37)
Errors Block 9	4.25 (8.21)	3.30 (7.30)
Working memory		
Spatial span length	5.56 (1.50)	6.35 (1.50)
Planning and organization		
Problems solved in minimum moves	7.06 (1.84)	7.83 (1.78)
Mean initial think time 2 moves[a]	1235.88 ms (999.16)[a]	2276.43 ms (2580.7)
Mean initial think time 3 moves	3768.34 ms (2035.11)	3477.76 ms (2422.11)
Mean initial think time 4 moves[a]	3648.70 ms (3686.78)	4754.98 ms (3348.64)
Mean initial think time 5 moves	3622.02 ms (2625.25)	7695.12 ms (6963.12)
Mean subsequent think time 2 moves[a]	166.71 ms (336.76)	33.68 ms (109.61)
Mean subsequent think time 3 moves[a]	933.94 ms (1304.97)	232.31 ms (508.67)
Mean subsequent think time 4 moves[a]	1278.22 ms (1197.07)	1172.56 ms (988.18)
Mean subsequent think time 5 moves[a]	1041.31 ms (1630.47)	791.13 ms (662.77)

Note: Neurocognitive task outcome demarcated by a superscript letter "a" indicates that data for this outcome did not meet the assumption of normality and was, in turn, transformed for data analytic purposes. Data reported on in this table are raw scores for all outcomes.
[a]Diagnostic status predicted the respective outcome of interest or demonstrated a trend towards significance.

(extradimensional set shifting; $\beta = -.06$, $P = .713$), after controlling for child age.

REVERSAL LEARNING (IDED, BLOCKS 7 AND 9)

Contrary to findings in relation to set shifting, results revealed that HPD status was a statistically significant predictor of total errors in Block 7 (intradimensional reversal learning; $\beta = .29$, $P = .049$) and yielding a moderate effect size, after controlling for the presence/absence of perinatal events. However, HPD status failed to predict total errors in Block 9 (extradimensional reversal learning; $\beta = -.07$, $P = .661$).

WORKING MEMORY (SSP)

Hierarchical regression analyses revealed that HPD status failed to predict performance on spatial span length (intradimensional set shifting; $\beta = -.11$, $P = .394$), after controlling for child age.

PLANNING AND ORGANIZATION (SOC)

Finally, a series of hierarchical regression analyses was conducted to examine whether HPD status predicted performance on relevant outcomes from the SOC. These analyses revealed that HPD status was a statistically significant predictor of mean initial thinking time 5 moves ($\beta = -.34$, $P = .032$), yielding a moderate effect size, and suggesting that children with HPD take longer to perform this component of the SOC. What is more, after controlling for child gender and depressive symptoms, results demonstrated a trend toward statistical significant in HPD status' ability to predict mean initial thinking time 2 moves ($\beta = -.24$, $P = .091$), yielding a small to moderate effect size. Additionally, analyses revealed that HPD status failed to predict performance on any of the remaining SOC outcomes.

RELATIONSHIP BETWEEN PERFORMANCE ON COGNITIVE TASKS, PULLING SEVERITY, AND PULLING STYLES

A strong, negative correlation was found between hair pulling severity—as assessed via the TSC-Child Total Score—and spatial span length ($r = -.58$, $P = .022$) suggesting that poorer performance on this task is associated with greater pulling severity. No additional, statistically significant relationships were found between performance on IED or SOC outcomes and pulling severity. Though, as demonstrated in Table 4, several correlations, particularly with respect to performance on the SOC, exhibited moderately strong though statistically nonsignificant correlations.

With respect to pulling styles, no statistically significant relationships or trends toward significance were found between cognitive outcomes and focused pulling scores from the MIST-C, though small to moderate, nonsignificant, negative correlations were found with respect to reversal learning (total errors Block 7 and 9, respectively) and select facets to planning and organization (i.e., mean subsequent and initial think time 2 moves). Contrarily, a strong, negative relationship was found between working memory performance (spatial span length) and automatic pulling (Spearman's rho = $-.52$, $P = .045$), suggesting that children exhibiting increasingly automatic (habitual) pulling demonstrated decreased performance on this task. As well, several weak to moderate to strong, nonsignificant, positive correlations were found with respect to select facets to planning and organization (i.e., mean initial think time 2, 3, and

TABLE 3. Summary of (hierarchical) regression analyses for SSP, ID/ED, and SOC

SST outcome	*B*	*SE (B)*	*β*	R^2	$F/\Delta F$	ΔR^2
Cognitive flexibility						
Total errors Block 6				.00	0.012	—
Diagnostic status	−0.02	0.20	−.02	—	—	—
Total errors Block 8						
Step 1				.11*	4.70	—
Child age	−0.20	0.09	−.34*	—	—	—
Step 2				.12	0.14	.00
Child age	−0.20	0.09	−.35*			
Diagnostic status	−0.16	0.43	−.06			
Reversal learning						
Total errors Block 7						
Step 1				.20**	9.31	—
Perinatal events	0.14	0.05	.45**	—	—	—
Step 2				.28**	4.14*	.08
Perinatal events	0.13	0.04	.41**			
Diagnostic status	0.08	0.04	.29*	—	—	—
Total errors Block 9				.01	0.20	—
Diagnostic status	−0.04	0.08	−.07	—	—	—
Working memory						
Spatial span Length						
Step 1				.51***	38.00	—
Child age	0.47	0.08	.71***	—	—	—
Step 2				.52***	0.75	.01
Child age	0.47	0.08	.69***			
Diagnostic status	−0.32	0.37	−.10	—	—	—
Planning and organization						
Problems solved minimum moves				0.04	1.69	—
Diagnostic status	−0.76	0.59	−.21	—	—	—
Mean initial think time 2 moves						
Step 1				.30***	7.88	—
Gender	−18.28	6.05	−.43**	—	—	—
Depressive symptoms	0.52	0.29	.26*	—	—	—
Step 2				.36***	3.03^{T}	.06
Gender	−16.90	5.94	−.40**	—	—	—
Depressive symptoms	0.54	0.28	.27	—	—	—
Diagnostic status	−10.22	5.87	−.24^{T}	—	—	—
Mean initial think time 3 moves						
Step 1				.13^{T}	2.61	—
Depressive symptoms	−22.60	54.03	−.11	—	—	—
ADHD symptoms	−43.91	42.60	−.26	—	—	—
Step 2				.15	1.16	
Depressive symptoms	2.00	58.55	.01	—	—	—
ADHD symptoms	−68.93	48.46	−.42	—	—	—
Diagnostic status	859.72	799.42	.19	—	—	—
Mean initial think time 4 moves						
Step 1				.20*	4.44	—
Depressive symptoms	−0.58	0.34	−.26^{T}	—	—	—
Gender	−20.17	7.26	−.43**	—	—	—
Step 2				.23*	1.26	.03
Depressive symptoms	−0.56	0.34	−.25	—	—	—
Gender	−19.08	7.30	−.40*	—	—	—
Diagnostic status	−8.11	7.22	−.17	—	—	—
Mean initial think time 5 moves						
Diagnostic status	−4073.10	1830.67	−.34*	.12*	4.95	—
Mean subsequent think time 2 moves						
Step 1				.19**	8.51	—
Perinatal events	0.93	0.32	.43**	—	—	—
Step 2				.21*	4.70	
Perinatal events	0.89	0.32	.42**	—	—	—

(*Continued*)

TABLE 3. Continued

SST outcome	*B*	*SE (B)*	*β*	R^2	$F/\Delta F$	ΔR^2
Diagnostic status	0.29	0.30	.14	—	—	—
Mean subsequent think time 3 moves						
Step 1				.19**	8.78	—
ADHD symptoms	0.62	0.21	.44**	—	—	—
Step 2				.23*	1.58	—
ADHD symptoms	0.54	0.22	.38*	—	—	–
Diagnostic status	7.48	5.95	.19	—	—	—
Mean subsequent think time 4 moves						
Step 1				.11*	4.43	—
Child age	−2.21	1.05	−.33*	—	—	—
Step 2				.11	0.21	—
Child age	−2.32	1.09	−.34*	—	—	—
Diagnostic status	−2.30	5.07	−.07	—	—	—
Mean subsequent think time 5 moves						
Step 1				.08T	3.24	—
Depressive symptoms	−0.45	0.25	−.28T	—	—	—
Step 2				.08	0.02	.00
Diagnostic status	−0.71	5.44	−.02	—	—	—

*$P \leq .05$.
**$P \leq .01$.
***$P \leq .001$.
TTrend towards statistical significance as defined by $P \leq .10$.

4 moves, mean subsequent think time 2 moves). Fisher's *z*-test revealed a statistically significant difference between correlations representative of the relationship between automatic and focused pulling scores and mean initiate think time 2 moves ($P < .05$). Likely due to our small sample size, no additional statistically significant relationships or differences between correlations were found.

DISCUSSION

The current study sought to examine executive functioning in a sample of youths with HPD. To our knowledge, this is the first study to investigate executive performance within such a sample and, as such, provides novel insights in relation to potential neurocognitive aspects of this sometimes debilitating disorder. In

TABLE 4. Spearman (nonparametric) correlations between neurocognitive measures, pulling severity and pulling styles (*N*= 15)

		Pulling styles	
	TSC-C total score	MIST-C: automatic	MIST-C: focused
Cognitive flexibility/reversal learning			
Total errors Block 6	−.09	−.42	−.28
Total errors Block 7	0.01	.01	−.22
Total errors Block 8	.31	.25	.16
Total errors Block 9	−.05	−.17	−.31
Working memory			
Spatial span length	−.59*	−.52*	−.17
Planning and organization			
Problems solved in minimum moves	.34	.48T	.41
Mean initial think time 2 moves	.11	.42	−.38
Mean initial think time 3 moves	.28	.20	.31
Mean initial think time 4 moves	.33	.33	.02
Mean initial think time 5 moves	.10	−.06	.03
Mean subsequent think time 2 moves	.06	.23	−.23
Mean subsequent think time 3 moves	.11	.06	−.08
Mean subsequent think time 4 moves	.11	.03	−.01
Mean subsequent think time 5 moves	.21	.03	−.12

Note: TSC-C, Trichotillomania Scale for Children-Child Report.
*$P < .05$.
TTrend toward statistical significance as defined by $P \leq .10$.

corroboration with proposed hypotheses, HPD youth demonstrated significant deficits in several areas of executive functioning performance including reversal learning and planning and organization, though performance on tasks of neither cognitive flexibility nor working memory differentiated children with HPD from healthy controls. What follows is a brief review of these findings and their implications for science's understanding as it relates to the etiology and treatment of pediatric HPD.

Contrary to past research within adult HPD populations,[24] current findings regarding SOC performance indicate impaired planning and organization among HPD youths compared to healthy controls. Interestingly, prior research within pediatric OCD samples indicates SOC performance as highly sensitive to frontostriatal dysfunction.[55] Thus the current data may implicate frontostriatal impairment as a critical factor for understanding the pathophysiology of HPD in youths. Results also indicated significant differences between correlations representative of the relationship between automatic and focused pulling scores and planning. Although requiring replication and extension among a large sample, this may point to important differences in risk factors contributing to the onset and/or maintenance of these disparate pulling styles. As such, future research examining neurological functioning within and across disparate pulling styles is warranted. What is more, past research within pediatric OCD samples suggests that poor response to treatment (e.g., cognitive behavioral therapy, CBT) may be related to executive functioning impairment (i.e., planning organization).[56] Although disparate disorders, the documented overlap between OCD and HPD suggests that, in addition to replication and extension of the findings described herein, future treatment studies may wish to examine the relationships between executive functioning performance (i.e., planning and organization) and treatment within pediatric HPD. Such lines of inquiry may prove beneficial for improving the efficacy and potential portability (i.e., development of cognitive enhancement interventions) of extant therapeutic interventions for pediatric HPD, particularly given practitioners general lack of knowledge and expertise in treating this understudied and misunderstood disorder.[57]

With respect to spatial span performance, the present study's findings suggest that diagnostic status—HPD or healthy control—failed to predict working memory capacity. Such findings are in contrast to prior research indicating decreased spatial span length among adult HPD samples[24] and imply potentially important differences within neurocognitive functioning (i.e., working memory) among youths and adults with HPD. Despite the absence of significant working memory impairment, findings from the current study demonstrate significant negative correlations between hair pulling severity and automatic pulling and spatial span length, in which poorer performance on this task was associated with greater pulling severity and increasingly automatic (habitual) pulling, respectively. Such impairments may further indicate executive functioning deficits specific to particular pulling styles in that working memory impairment may be a critical factor in the etiology and/or maintenance of automatic pulling, while discordantly playing lesser of or no role in the pathophysiology of focused pulling. Future research may wish to reexamine differences in working memory outcomes within a larger sample ($N = 100+$) of youths with HPD representing various degrees of pulling styles. Such lines of inquiry may, in turn, greatly enhance our pathophysiological understanding of HPD and inform advances in treatment.

Contrary to proposed hypotheses, results of the current study were nonsignificant in relation to cognitive flexibility outcomes; however, results did indicate impaired reversal learning amongst HPD youths. Collectively, these findings both complement, as well as contradict previous adult literature demonstrating lack of impairment in both reversal learning[24] and attention shifting[8,25] among adults diagnosed with HPD. Results herein suggest that unique cognitive profiles may exist among youths and adults with HPD, in which children with HPD exhibit difficulty learning to reverse their maladaptive behaviors (e.g., hair pulling) exclusively within complex situations. Such findings may implicate distinct brain mechanisms involved in the pathophysiology of HPD across the lifespan. From a phenotypic perspective, proposed difficulties in behavior reversal in the presence of complex stimuli may help explain pulling persistence in youths with HPD. In particular, when faced with aversive stimuli (e.g., negative events, emotions, etc.) youths may experience difficulty shifting their attention from automatic or learned maladaptive behaviors (i.e. hair pulling) or alternatively may experience difficulty generalizing positive coping strategies to such complex negative situations. As such, reversal learning impairments may indicate mechanisms upon which cognitive-behavior therapy (CBT; i.e., habit reversal training, stimulus control, etc.)[58] works to decrease hair pulling in children. Given the paucity of executive functioning research in pediatric HPD, additional studies are necessary to corroborate results within the reversal learning domain (i.e., large samples, multiple comparison groups, etc.). Similarly, the examination of executive functioning performance/changes pre- to posttreatment as potential mechanisms of changes in psychosocial (i.e., habit reversal training) interventions for pediatric HPD and related concerns appears warranted.

Though findings of the current study provide novel insights into neurocognitive correlates of pediatric HPD, there are several limitations to be considered. First, neurocognitive performance of each domain was measured utilizing only one form of assessment. Although the CANTAB battery has been used in previous pediatric studies[59,60] assessing neurocognitive functioning, further research may wish to include additional measures (i.e., paper and pencil, additional computerized tasks, etc.) that may corroborate and extend findings presented herein. A second limitation, common to previous HPD

research, is the homogeneity of our utilized sample. Future research should consider replication of study procedures in a diversified (e.g., ethnicity, socioeconomic status, gender, etc.) sample to determine possible similarities and differences in outcome. Lastly, study findings suggest that differences between youths with HPD and healthy controls, as well as differences between pulling styles, may indicate critical etiological and/or maintenance factors within pediatric HPD. Though such hypotheses are plausible, limitations of cross sectional study design impede confirmation of these suggestions. As such, contingent on replication of current study findings, future longitudinal research may examine differences in executive functioning (i.e., planning and organization and working memory) throughout HPD development as well as during the course of psychosocial and pharmacological interventions. Despite these limitations, the results of this study provide much needed information regarding potential underlying mechanisms of pediatric HPD. As such, continued research in this domain can help enhance our development of a more comprehensive understanding of this disorder.

Conflict of interest. None of the authors have any conflicts of interests or financial gains to disclose.

REFERENCES

1. Christenson GA, Pyle RL, Mitchell JE. Estimated lifetime prevalence of trichotillomania in college students. J Clin Psychiatry 1991;52(10):415–417.
2. Huynh M, Gavino AC, Magid M. Trichotillomania. Semin Cutan Med Surgery 2013;32(2):88–94.
3. Hajcak G, Franklin ME, Simons RF, Keuthen NJ. Hair-pulling and skin picking in relation to affective distress and obsessive-compulsive symptoms. J Psychopathol Behav Assess 2006;28(3):177–185.
4. Swedo SE, Rapoport JL. Annotation: trichotillomania. J Child Psychol Psychiatry 1991;32(3):401–409.
5. Harrison JP, Franklin ME. Pediatric trichotillomania. Curr Psychiatry Rep 2012;14(3):188–196.
6. Rothbaum BO, Ninan PT. The assessment of trichotillomania. Behav Res Ther 1994;32(6):651–662.
7. American Psychiatric Association, Diagnostic and statistical manual of mental disorders. 5th ed., Washington, DC: bookpointUS; 2013.
8. Chamberlain SR, Fineberg NA, Blackwell AD, Robbins TW, Sahakian BJ. Motor inhibition and cognitive flexibility in obsessive-compulsive disorder and trichotillomania. Am J Psychiatry 2006;163(7):1282–1284.
9. Christenson GA, Crow SJ. The characterization and treatment of trichotillomania. J Clin Psychiatry 1996;57(Suppl 8):42–47; discussion 48-49.
10. Diefenbach GJ, Reitman D, Williamson DA. Trichotillomania: a challenge to research and practice. Clin Psychol Rev 2000;20(3):289–309.
11. Grant JE, Levine L, Kim D, Potenza MN. Impulse control disorders in adult psychiatric inpatients. Am J Psychiatry 2005;162(11):2184–2188.
12. Hollander E, Rosen J. Impulsivity. J Psychopharmacol 2000;14(2 Suppl 1):S39–S44.
13. Stein DJ, Lochner C. Obsessive-compulsive spectrum disorders: a multidimensional approach. Psychiatr Clin North Am 2006;29(2):343–351.
14. American Psychiatric Association, Diagnostic and Statistical Manual of Mental Disorders. 4th ed., text revision. Washington, DC: American Psychiatric Association; 2000.
15. Flessner CA, Knopik VS, McGeary J. Hair pulling disorder (trichotillomania): genes, neurobiology, and a model for understanding impulsivity and compulsivity. Psychiatry Res 2012;199(3):151–158.
16. Lochner C, Seedat S, du Toit PL, et al. Obsessive-compulsive disorder and trichotillomania: a phenomenological comparison. BMC Psychiatry 2005;5:2.
17. Graybiel AM, Rauch SL. Toward a neurobiology of obsessive-compulsive disorder. Neuron 2000;28:343–347.
18. Rosenberg DR, Keshavan MS. Toward a neurodevelopmental model of obsessive–compulsive disorder. Biol Psychiatry 1998;43(9):623–640.
19. Rosenberg DR, MacMaster FP, Mirza Y, Easter PC, Buhagiar CJ. Neurobiology, neuropsychology and neuroimaging of child and adolescent obsessive-compulsive disorder. In: Storch E GG, Murphy T, ed. Handbook of Child and Adolescent Obsessive-Compulsive Disorder. Mahwah, NJ: Lawrence Erlbaum Associates; 2007.
20. Stein DJ, Hollander E, Cohen L, Simeon D, Aronowitz B. Serotonergic responsivity in trichotillomania: neuroendocrine effects of m-chlorophenylpiperazine. Biol Psychiatry 1995;37(6):414–416.
21. Bohne A, Savage CR, Deckersbach T, et al. Visuospatial abilities, memory, and executive functioning in trichotillomania and obsessive-compulsive disorder. J Clin Exp Neuropsychol 2005;27(4):385–399.
22. Bohne A, Savage CR, Deckersbach T, Keuthen NJ, Wilhelm S. Motor inhibition in trichotillomania and obsessive-compulsive disorder. J Psychiatr Res 2008;42(2):141–150.
23. Chamberlain SR, Fineberg N, Blackwell AD, Robbins TW, Sahakian BJ. Motor inhibition and cognitive flexibility in obsessive-compulsive disorder and trichotillomania. Am J Psychiatry 2006;163:1282–1284.
24. Chamberlain SR, Fineberg NA, Blackwell AD, Clark L, Robbins TW, Sahakian BJ. A neuropsychological comparison of obsessive-compulsive disorder and trichotillomania. Neuropsychologia 2007;45(4):654–662.
25. Keuthen NJ, Savage CR, O'Sullivan RL, et al. Neuropsychological functioning in trichotillomania. Biol Psychiatry 1996;39(8):747–749.
26. Stanley MA, Hannay HJ, Breckenridge JK. The neuropsychology of trichotillomania. J Anxiety Disord 1997;11(5):473–488.
27. Rettew DC, Cheslow DL, Rapoport JL, et al. Neuropsychological test performance in trichotillomania: a further link with obsessive-compulsive disorder. J Anxiety Disord 1991;5(3):225–235.
28. Odlaug BL, Chamberlain SR, Grant JE. Motor inhibition and cognitive flexibility in pathologic skin picking. Prog Neuropsychopharmacol Biol Psychiatry 2010;34(1):208–211.
29. Grant JE, Odlaug BL, Chamberlain SR. A cognitive comparison of pathological skin picking and trichotillomania. J Psychiatr Res 2011;45(12):1634–1638.
30. Brennan E, Francazio S, Gunstad J, Flessner C. Inhibitory control in pediatric trichotillomania (hair pulling disorder): the importance of controlling for age and symptoms of inattention and hyperactivity. Child Psychiatry Hum Dev in press.
31. Christenson G. Trichotillomania - from prevalence to comorbidity. Psychiatr Times 1995;12(9):44–48.

32. Flessner CA, Woods DW, Franklin M, Cashin SE, Keuthen NJ, Board TLCSA. The Milwaukee Inventory of Trichotillomania - Adult Version (MIST-A): development of an instrument for the assessment of "focused" and "automatic" hair pulling. J Psychopathol Behav Assess 2008;30:20–30.
33. Flessner CA, Woods DW, Franklin ME, et al. The Milwaukee Inventory for Styles of Trichotillomania-Child Version (MIST-C): initial development and psychometric properties. Behav Modif 2007;31(6):896–918.
34. Klingberg T, Fernell E, Olesen PJ, et al. Computerized training of working memory in children with ADHD–a randomized, controlled trial. J Am Acad Child Adolesc Psychiatry 2005;44(2):177–186.
35. Klingberg T, Forssberg H, Westerberg H. Training of working memory in children with ADHD. J Clin Exp Neuropsychol 2002;24(6):781–791.
36. Eldar S, Apter A, Lotan D, et al. Attention bias modification treatment for pediatric anxiety disorders: a randomized controlled trial. Am J Psychiatry 2012;169(2):213–220.
37. Francazio SK, Flessner CA. Cognitive flexibility differentiates young adults exhibiting obsessive-compulsive behaviors from controls. Psychiatry Res 2015;228(2):185–190.
38. Odlaug BL, Chamberlain SR, Harvanko AM, Grant JE. Age at onset in trichotillomania:clinical variables and neurocognitive performance. Prim Care Companion CNS Disord 2012;14(4): doi:10.4088/PCC.12m01343
39. Kaufman J, Birmaher B, Brent D, et al. Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL): initial reliability and validity data. J Am Acad Child Adolesc Psychiatry 1997;36(7):980–988.
40. Ambrosini PJ. Historical development and present status of the schedule for affective disorders and schizophrenia for school-aged children (K-SADS). J Am Acad Child Adolesc Psychiatry 2000;39:49–58.
41. Silverman W, Albano A. The Anxiety Disorders Interview Schedule for Children–IV (Child and Parent Versions). San Antonio, TX: Psychological Corporation; 1996.
42. Silverman WK, Nelles WB. The Anxiety Disorders Interview Schedule for Children. J Am Acad Child Adolesc Psychiatry 1988;27(6):772–778.
43. Silverman WK, Saavedra LM, Pina AA. Test-retest reliability of anxiety symptoms and diagnoses with the Anxiety Disorders Interview Schedule for DSM-IV: child and parent versions. J Am Acad Child Adolesc Psychiatry 2001;40(8):937–944.
44. Wood JJ, Piacentini JC, Bergman RL, McCracken J, Barrios V. Concurrent validity of the anxiety disorders section of the Anxiety Disorders Interview Schedule for DSM-IV: child and parent versions. J Clin Child Adolesc Psychol 2002;31(3):335–342.
45. Diefenbach GJ, Tolin DF, Crocetto J, Maltby N, Hannan S. Assessment of trichotillomania: a psychometric evaluation of hair-pulling scales. J Psychopathol Behav Assess 2005;27(3):169–178.
46. Diefenbach GJ, Tolin DF, Franklin ME, Anderson ER. The Trichotillomania Scale for Children (TSC): a new self-report measure to assess pediatric hair pulling. Annual Meeting of the Association for Advancement of Behavior Therapy, Boston, MA, 2003.
47. Tolin DF, Diefenbach GJ, Flessner CA, et al. The trichotillomania scale for children: development and validation. Child Psychiatry Hum Dev 2008;39(3):331–349.
48. Achenbach TM. Integrative guide for the 1991 CBCL/4–18, YSR and TRF profiles. Burlington: Department of Psychiatry, University of Vermont; 1991.
49. Achenbach TM, Edelbrock CS. Manual for the Child Behavior Checklist and Revised Child Behavior Profile. Burlington, Vermont: University of Vermont, Department of Psychiatry; 1983.
50. Ivanova MY, Achenbach TM, Dumenci L, et al. Testing the 8-syndrome structure of the child behavior checklist in 30 societies. J Clin Child Adolesc Psychol 2007;36(3):405–417.
51. Dutra L, Campbell L, Westen D. Quantifying clinical judgment in the assessment of adolescent psychopathology: reliability, validity, and factor structure of the child behavior checklist for clinician report. J Clin Psychol 2004;60(1):65–85.
52. Jensen PS, Watanabe HK, Richters JE, et al. Scales, diagnoses, and child psychopathology: II. Comparing the CBCL and the DISC against external validators. J Abnorm Child Psychol 1996;24(2):151–168.
53. Rishel CW, Greeno C, Marcus SC, Shear MK, Anderson C. Use of the child behavior checklist as a diagnostic screening tool in community mental health. Res Social Work Prac 2005;15(3):195–203.
54. Lampert TL, Polanczyk G, Tramontina S, Mardini V, Rohde LA. Diagnostic performance of the CBCL-Attention Problem Scale as a screening measure in a sample of Brazilian children with ADHD. J Atten Disord 2004;8(2):63–71.
55. Huyser C, Veltman DJ, Wolters LH, de Haan E, Boer F. Functional magnetic resonance imaging during planning before and after cognitive-behavioral therapy in pediatric obsessive-compulsive disorder. J Am Acad Child Adolesc Psychiatry 2010;49(12):1238–1248.e1235.
56. Flessner CA, Allgair A, Garcia A, et al. The impact of neuropsychological functioning on treatment outcome in pediatric obsessive-compulsive disorder. Depress Anxiety 2010;27(4):365–371.
57. Flessner CA, Penzel F, Board TLC-SA, Keuthen NJ. Current treatment practices for children and adults with trichotillomania: consensus among the experts. Cogn Behav Pract 2010;17(3):290–300.
58. Franklin M, Edson AL, Ledley DA, Cahill SP. Behavior therapy for pediatric trichotillomania: a randomized controlled trial. Am Acad Child Adolesc Psychiatry 2011;50(8):763–771.
59. Dickstein DP, Treland JE, Snow J, et al. Neuropsychological performance in pediatric bipolar disorder. Biol Psychiatry 2004;55(1):32–39.
60. Rasmussen C, Soleimani M, Carroll A, Hodlevskyy O. Neuropsychological functioning in children with Tourette syndrome (TS). J Can Acad Child Adolesc Psychiatry 2009;18(4):307–315.