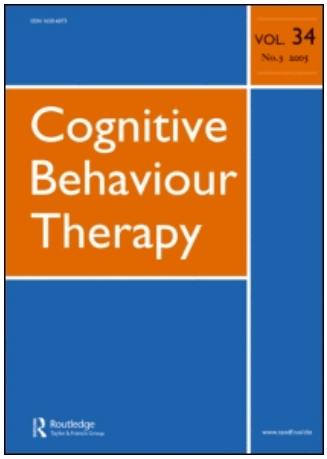

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Pediatric Trichotillomania: Descriptive Psychopathology and an Open Trial of Cognitive Behavioral Therapy

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Abstract. In study 1, 46 children and adolescents with trichotillomania who sought treatment at 2 specialty outpatient clinics were assessed. Most children reported pulling hair from multiple sites on the body, presented with readily visible alopecia, reported spending 30–60 minutes per day pulling or thinking about pulling, and reported experiencing significant distress about their symptoms. Most were described by their parents as having significant problems in school functioning. Few children met criteria for obsessive-compulsive disorder or tic disorder. Child and family rates of other forms of psychopathology were high. In study 2, 22 of these children were enrolled in an open trial of individual cognitive behavioral therapy with particular attention to relapse prevention. Trichotillomania severity decreased significantly and 77% of children were classified as treatment responders at post-treatment and 64% at 6-month follow-up. *Key words: trichotillomania; cognitive behavioral therapy; behavior therapy; children; hair-pulling*

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Trichotillomania (TTM), a chronic impulse control disorder characterized by repetitive pulling out of one's own hair and resulting alopecia, appears to be more common in young people than was previously believed. Although early reports (e.g. Mannino & Delgado, 1969) described child and adolescent TTM as a very rare condition, more recent studies using epidemiological and student samples of older adolescents and young adults estimate the prevalence of clinically significant pulling to range between 1% and 3.5% (Christenson, Pyle, & Mitchell, 1991; King, Zohar et al., 1995; Rothbaum, Shaw, Morris, & Ninan, 1993). The prevalence of TTM among younger children remains unknown.

DSM-IV-TR (American Psychiatric Association, 2000) specifies that patients with TTM experience an increasing sense of tension immediately before pulling or when attempting to resist the behavior, as well as pleasure, gratification or relief when pulling. However, these criteria have been challenged in light of the fact that many patients with other signs of TTM deny these symptoms, often reporting that pulling takes place outside awareness (e.g. Christenson, MacKenzie, & Mitchell, 1991; King, Zohar et al., 1995). The frequency of these symptoms in pediatric TTM samples is not fully known, due in part to the small samples used in previous studies. Studies with samples of children and adolescents have found the tension-relief cycle in

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1/10 (10%; Reeve, Bernstein, & Christenson, 1992), 4/11 (36%; Hanna, 1997) and 11/15 (73%; King, Scahill et al., 1995) of participants.

Psychiatric comorbidity appears to be quite common among adults with TTM, particularly mood, anxiety, substance use and personality disorders (Christenson, 1995; Christenson, Chernoff-Clementz, & Clementz, 1992; Christenson, MacKenzie, & Mitchell, 1991; Schlosser, Black, Blum, & Goldstein, 1994). To date, only 3 studies with very small samples (ns=10–15) have been published on the comorbidity of TTM in pediatric clinical samples (Hanna, 1997; King, Scahill et al., 1995; Reeve et al., 1992), with estimates of 60–70% of children meeting criteria for at least 1 comorbid psychiatric disorder. Among these, anxiety and depressive disorders were diagnosed most frequently (36–70%), followed by disruptive behavior disorders (27–47%). Perhaps inconsistent with the view that TTM is part of an ''obsessive-compulsive spectrum'' (e.g. Hollander et al., 1996), comparatively low rates of obsessive-compulsive disorder (OCD; 0–13%) and tic disorders (9–13%) were reported in these samples.

In studies of convenience samples, TTM appears to be associated with substantial impairment and reductions in quality of life. The majority of adult TTM patients report problems such as impaired social functioning, negative affect, interference with grooming behaviors, impaired recreational activity, work productivity, and physical illness or symptoms caused by pulling (Diefenbach, Tolin, Hannan, Crocetto, & Worhunsky, 2005; Keuthen et al., 2002). Many TTM patients spend considerable time concealing areas of alopecia (Swedo & Leonard, 1992), and experience guilt, shame and low self-esteem (Diefenbach, Tolin, Hannan et al., 2005). Among children and adolescents, the presence of hair-pulling appears to elicit negative peer evaluations (Boudjouk, Woods, Miltenberger, & Long, 2000), although the impact of TTM on quality of life has not been well documented in pediatric samples.

Effective and efficient treatment is viewed as critical for children and adolescents with TTM in order to minimize the likely adverse effects of pulling and alopecia on socialization and social identity. To date, however, no studies have examined the efficacy of pharmacological or cognitive behavioral interventions for pediatric TTM. Data from controlled studies of adults suggest that selective serotonin reuptake inhibitor (SSRI) antidepressant pharmacotherapy is not effective (Christenson, MacKenzie, Mitchell, & Callies, 1991; Streichenwein & Thornby, 1995); data for the serotonin reuptake inhibitor (SRI) antidepressant clomipramine are mixed (Ninan, Rothbaum, Marsteller, Knight, & Eccard, 2000; Swedo, Lenane, & Leonard, 1993). Furthermore, the use of clomipramine in children raises safety concerns (e.g. cardiac risk) to a greater extent than do the SSRIs (Emslie & Judge, 2000; Hazell, O'Connell, Heathcote, & Henry, 2002).

A small number of controlled trials suggest that cognitive behavioral therapy (CBT) can be efficacious for adults with TTM (Azrin, Nunn, & Frantz, 1980; Diefenbach, Tolin, Hannan, Maltby, & Crocetto, 2006; Ninan et al., 2000; van Minnen, Hoogduin, Keijsers, Hellenbrand, & Hendriks, 2003). Ninan et al. (2000) found CBT superior to both clomipramine and placebo at post-treatment. Van Minnen et al. (2003) reported a similar pattern of results comparing CBT with the SSRI antidepressant fluoxetine. In an early trial, Azrin et al. (1980) reported that habit reversal training, a component of CBT, was more effective than was negative practice. Diefenbach et al. (2006) found group CBT to be superior to supportive therapy, although treatment gains were fairly modest.

The significant problem of relapse following CBT, which is certainly a common phenomenon reported clinically, was highlighted in an open trial of CBT incorporating habit reversal (Lerner, Franklin, Meadows, Hembree, & Foa, 1998), with the majority of treatment responders losing this designation at extended follow-up. Similar findings have emerged from studies of adults receiving group CBT (Diefenbach et al., 2006; Mouton & Stanley, 1996).

The present article describes 2 studies of TTM in a pediatric sample. The primary aim of study 1 is to provide descriptive information about children and adolescents seeking treatment for TTM, using a larger sample than has been investigated previously (King, Scahill et al., 1995; Reeve et al., 1992; Tay,

Levy, & Metry, 2004). Study 1 also expands upon previous findings by using standardized TTM interviews and clinician rating scales to describe the phenomenology and severity of pediatric hair-pulling. Family history of mental disorder and signs of educational and social impairment are also examined. The primary aim of study 2 is to describe the results of a preliminary open trial of CBT for children and adolescents with TTM. The treatment protocol was largely based on standard interventions used for adults with TTM (Lerner et al., 1998; Ninan et al., 2000; van Minnen et al., 2003), but to our knowledge this is the first systematic application of such principles to a pediatric sample (see Roblek, Detweiler, Fearing, & Albano, 1999; Vitulano, King, Scahill, & Cohen, 1992, for case reports). To address concerns about relapse, existing protocols were modified to incorporate a treatment tapering phase in which relapse prevention strategies (McKay, 1997; McKay, Todaro, Neziroglu, & Yaryura-Tobias, 1996) were emphasized. It was anticipated that CBT would lead to reductions in TTM severity, and that treatment gains would be maintained over a 6-month follow-up period.

Study 1: descriptive psychopathology

Method

Participants. Forty-six children and adolescents with TTM participated in the present study as part of an open trial and randomized controlled trial of CBT for pediatric TTM. Inclusion criteria were: age range 8–17 years (inclusive); primary diagnosis of TTM; and minimum symptom duration of 6 months. Exclusion criteria were a primary diagnosis other than TTM; current bipolar illness, developmental disorder, or thought disorder; or current psychotherapy. Comorbid conditions were allowed, provided that TTM was clearly the primary concern in the opinion of the assessor. The mean age of the sample was 12.6 (SD=2.9) years. Thirty-three (71.7%) of the participants were female. Thirty-six (78.3%) were Caucasian; the primary minority group was African-American (13.0%). As is customary in TTM studies (Franklin, Tolin, & Diefenbach, 2006), criteria B and C (increasing and decreasing tension) of the DSM-IV-TR criteria for TTM were optional, as these criteria have been found to exclude patients with clearly significant hair-pulling (Christenson, MacKenzie, & Mitchell, 1991; Hanna, 1997; Schlosser et al., 1994).

Measures of TTM. The diagnostic criteria for TTM were assessed via the Trichotillomania Diagnostic Interview (TDI) (Rothbaum & Ninan, 1994), which corresponds to the DSM-IV-TR criteria. Assessors rated the degree of TTM severity and impairment using the NIMH Trichotillomania Severity Scale (NIMH-TSS) and NIMH Trichotillomania Impairment Scale (NIMH-TIS) (Swedo, Rapoport, Leonard, Lenane, & Cheslow, 1989), 2 ratings derived from a semi-structured interview that assesses time spent pulling in the past week, time spent pulling the previous day, resistance to pulling, distress and interference. The NIMH-TSS shows adequate internal consistency, excellent inter-rater agreement, and adequate correlations with other TTM severity measures (Diefenbach, Tolin, Crocetto, Maltby, & Hannan, 2005). The single-item NIMH-TIS shows good correlations with interviewer, although not self-report, measures of TTM (Diefenbach, Tolin, Crocetto et al., 2005). Additional information about TTM severity was obtained using the Psychiatric Institute Trichotillomania Scale (PITS) (Winchel et al., 1992), a clinician-rated measure that assesses number of hair-pulling sites, duration of time spent pulling or thinking about pulling, frequency of resisting hair-pulling urges, interference, distress, and severity of hair loss. PITS items are scaled from 0 to 7, with a maximum total score of 42. The PITS shows excellent inter-rater reliability, but rather low internal consistency (Diefenbach, Tolin, Crocetto et al., 2005). Therefore, emphasis will be placed on descriptive hair-pulling data (e.g. age of onset) collected via the PITS rather than the total score.

Measure of family history and school functioning. Parents described children's early history, family history, and school performance on the Conners-March Developmental Questionnaire (CMDQ) (Conners & March, 1996), an open-ended history questionnaire. The psychometric properties of the CMDQ are not known. However, given the paucity of descriptive information about children and

adolescents with TTM, the CMDQ was included for exploratory purposes.

Measures of co-morbid psychopathology. Comorbid psychiatric diagnoses were ascertained using the Anxiety Disorders Interview Schedule for DSM-IV, Child Version (ADIS-C) (Silverman & Albano, 1996), a semi-structured diagnostic interview assessing anxiety, mood, externalizing, somatoform, psychotic, and substance-use disorders. Reliability for the various DSM-IV categories comprising the ADIS-C extends from good to excellent (Albano & Silverman, 1996; Silverman, Saavedra, & Pina, 2001). In addition, clinical ratings demonstrate strong interrater reliability and test-retest reliability (Brown, Di Nardo, Lehman, & Campbell, 2001). Comorbid psychopathology and behavioral problems were assessed further using the Conners Parent Rating Scale-Revised (CPRS-R) (Conners, 2000), a broad-based parent report with good internal reliability coefficients, high test-retest reliability, and effective discriminatory power (Conners, Sitarenios, Parker, & Epstein, 1998), the Multidimensional Anxiety Scale for Children (MASC) (March, Parker, Sullivan, Stallings, & Conners, 1997), a self-report measure of physical anxiety, harm avoidance, social anxiety and separation anxiety that shows excellent internal consistency and adequate convergent and divergent validity (March et al., 1997), and the Children's Depression Inventory (CDI) (Kovacs, 1985), a self-report measure of depressive symptoms with strong internal consistency (Kovacs, 1985), acceptable test-retest reliability (Kovacs, 1992), and acceptable convergent validity (Saylor et al., 1984).

Procedure

Thirty participants were assessed at the University of Pennsylvania School of Medicine in Philadelphia, PA; 16 were assessed at the Institute of Living in Hartford, CT. Participants at the 2 sites did not differ in terms of age or total severity of TTM as measured by the PITS. There was, however, a significant sex difference between the 2 sites, with boys representing 40% of the Pennsylvania participants vs only 6.3% of the Hartford participants (Fisher's Exact Test=0.018). Children were accompanied by at least 1 parent, and written assent (children) and consent (parents) were obtained. Assessments were conducted in the clinic by a doctoral-level psychologist or postdoctoral fellow trained and supervised by the first or second author. Prior to the assessment, children and their parents were given a packet of questionnaires to complete at home. Assessments were completed in the context of recruitment for a CBT trial. Participants meeting eligibility requirements were offered free treatment within the context of a clinical trial for TTM.

Data analysis

Data were analyzed using SPSS 10.0. Missing items were infrequent and were replaced with the scale mean for each participant. Due to administrative error, some children did not complete some of the items on the PITS; these were not replaced. For the CPRS and MASC, scores were z-transformed using published means and standard deviations, and re-coded onto the original scale. These scores were then reported as above or below published clinical thresholds ($z \geqslant 1.5$), using weighted norms based on the male:female ratio of the sample. Examination of the relationship between TTM severity and co-morbid conditions was conducted using independent-samples t-tests.

Results

Description of hair-pulling behavior. As shown in Table 1, participants reported that their pulling began at a mean of age 9.1 (3.1) years. Thus, the average child had been pulling for 3.5 (2.8) years at the time of the assessment, and in most cases the symptoms had been present at least half the time since they began. However, pulling was not continuous for this time: in the majority of cases, the symptoms had reportedly gone away and come back, in nearly half the cases for 2 weeks or longer. Thus, hair-pulling in this sample appears to have followed a chronic, fluctuating course. The most prominent pulling site was the scalp, followed by eyelashes and eyebrows. Substantially fewer children reported pulling from any other parts of the body. The majority (72.1%) of children reported efforts (e.g. wearing hats or scarves, brushing their existing hair over bald patches, and wearing make-up) to conceal the effects of hair-pulling

Table 1. *Description of hair-pulling behavior from the Psychiatric Institute Trichotillomania Scale (PITS)*

Item		*n* (%) reporting	Mean (SD)
Age pulling began (*n*=44)			9.14 (3.07)
Pulling has gone away and come back (*n*=44)		22 (50.0)	
Pulling has been present more than half the time since it began (*n*=44)		26 (59.1)	
Pulling sites have shifted (*n*=43)		13 (30.2)	
Number of pulling sites (*n*=39)	No sites	0 (0.0)	
	1 Non-scalp site	3 (7.7)	
	1 Scalp site	21 (53.8)	
	2 Non-scalp sites	5 (12.8)	
	2 Sites including scalp	4 (10.3)	
	3 Sites	4 (10.3)	
	4 Sites	1 (2.2)	
	5 Or more sites	1 (2.2)	
Severity of alopecia (*n*=39)	0 (No loss)	0 (0.0)	3.64 (1.31)
	1 (Negligible loss [can't see loss even if site pointed out])	0 (0.0)	
	2 (Mild loss [seen only if area pointed out])	8 (20.5)	
	3 (Moderate loss [loss visible to observer upon inspection) e.g. thin spots on scalp])	13 (33.3)	
	4 (Loss of 50% of hair of brows or lashes or nearly bald spots on scalp or body part)	8 (20.5)	
	5 (Loss of 75% of hair of brows or lashes or medium-sized bald spot on scalp or body part)	5 (12.8)	
	6 (Loss of almost all hair of brows or lashes or large areas of baldness on scalp or other body part)	5 (12.82)	
	7 (Total loss of hair of brows or lashes or almost total loss of scalp hair or other body part)	0 (0.0)	
Duration of pulling episodes (*n*=39)	No time	1 (2.6)	
	Less than 5 minutes/day	3 (7.7)	
	5–15 minutes/day	8 (20.5)	
	16–30 minutes/day	3 (7.7)	
	31–60 minutes/day	7 (17.9)	
	1–2 hours/day	9 (23.1)	
	2–3 hours/day	6 (15.4)	
	More than 3 hours/day	2 (5.1)	
Resistance to pulling (0–7) (*n*=39)			4.26 (2.12)
Distress from pulling (0–7) (*n*=39)			3.79 (1.69)
Interference from pulling (0–7) (*n*=39)			1.56 (1.52)
Total score (0–42) (*n*=39)			19.92 (4.10)

from others. The proportion of children endorsing TTM criteria B and C (increasing sense of tension and gratification or relief from pulling) was also examined. Eighty-two percent reported feeling an increasing sense of tension prior to pulling, 84% reported gratification or relief when pulling, and 78% reported both. Fifteen percent denied both of these.

Due to administrative error, only 39 children responded to the PITS severity questions. Table 1 shows the item means or frequencies, depending on the nature of the scale. As shown in the Table, PITS total scores were consistent with reports of moderate TTM severity (although, as noted previously, the total score should be interpreted cautiously due to low internal consistency). Children reported pulling hair from multiple sites (a score of 3 implies 2 pulling sites). Alopecia severity ratings by the examiner showed that most children had at least moderate alopecia, with nearly half the sample exhibiting 50% or greater hair loss on the scalp, eyelashes, or eyelids. Most children reported at least 30–60 minutes per day pulling hair or thinking about pulling, and 20% reported a daily duration of more than 2 hours. Children reported some degree of control over pulling, and estimated that they could resist the urge to pull between 50% and 75% of the time. They reported worrying daily about hair-pulling, and distress was moderately severe. Self-ratings of avoidance of activities were not as high as the others (a rating of 2 implies "frequently avoids 1 or more minor activity, creating some inconvenience"). Male and female participants did not differ in total PITS scores (t (37)=1.44, p=0.159). Age was significantly correlated with total PITS score (r=0.435, p=0.006), with older children reporting greater TTM severity. Exploratory analysis of the individual PITS severity items suggested that this was due to older children reporting greater interference (r=0.408, p=0.010) and distress (r=0.353, p=0.027) due to TTM (correlations with the remaining PITS items were not significant).

Comorbid psychiatric disorders. Table 2 shows the frequency of comorbid diagnoses as assessed by the ADIS-C. Thirty-eight percent of the sample met criteria for at least 1 DSM-IV-TR diagnosis in addition to TTM. Anxiety disorders were the most common, diagnosed in 30% of the children.

Table 2. *Frequency of comorbid psychiatric disorders on the Anxiety Disorders Interview Schedule for DSM-IV, Child Version (ADIS-C) (n=46)*

Diagnosis	*n* (%) Meeting criteria
Obsessive-compulsive disorder	3 (6.5)
Separation anxiety disorder	0 (0.0)
Social phobia	4 (8.7)
Specific phobia	3 (6.5)
Panic disorder	1 (2.2)
Generalized anxiety disorder	6 (13.0)
Posttraumatic stress disorder	0 (0.0)
Any anxiety disorder	14 (30.4)
Tourette's disorder	0 (0.0)
Other tic disorder	1 (2.2)
Any tic disorder	1 (2.2)
Dysthymic disorder	0 (0.0)
Major depressive disorder	4 (8.7)
Any depressive disorder	4 (8.7)
Attention deficit hyperactivity disorder	4 (8.7)
Conduct disorder	0 (0.0)
Oppositional defiant disorder	3 (6.5)
Any externalizing disorder	5 (10.9)
Any comorbid disorder	18 (39.1)

Table 3. *Percentage of children scoring in the clinical range on the Conners Parent Rating Scale (CPRS; n=46)*

CPRS Subscale	*n* in clinical range	% in clinical range
Social problems	14	30.4
ADHD Index	14	30.4
DSM-IV ADHD Inattentive	14	30.4
Global Index – Total	12	26.1
DSM-IV ADHD Total	12	26.1
Cognitive problems	11	23.9
Global Index – Emotional Lability	10	21.7
DSM-IV ADHD Hyperactive-Impulsive	9	19.6
Oppositional	8	17.4
Hyperactivity	8	17.4
Psychosomatic	8	17.4
Global Index – Restless/Impulsive	8	17.4
Anxious-shy	4	8.7
Perfectionism	3	6.5

Of these, generalized anxiety disorder was the most prevalent (13%). Only 3 (6.5%) children met criteria for obsessive-compulsive disorder (OCD). Eleven percent of the sample met criteria for an externalizing disorder, primarily attention deficit-hyperactivity disorder (ADHD). Nine percent met criteria for major depressive disorder. Tic disorders were uncommon. Male and female participants did not differ in terms of the prevalence of comorbid anxiety, mood, tic, externalizing, or other disorders (all $ps > 0.05$).

Table 3 shows the number of children exceeding clinical cut-offs on the CPRS subscales. Significant elevations were seen in the percentage of children rated as having oppositional behavior, cognitive problems, hyperactivity, social problems and psychosomatic concerns. In addition, all of the CPRS ADHD scales were significantly elevated, with approximately 30% of children scoring in the clinical range for the overall ADHD index. Of note, more children were described as meeting DSM-IV-TR criteria for inattentive type than hyperactive/impulsive type, although this difference was not statistically significant ($p > 0.05$). Children with co-morbid anxiety ($t=0.13$), mood ($t=0.89$), and externalizing disorders ($t=0.04$) did not exhibit greater TTM severity than did children without these disorders (all p's > 0.05).

School functioning. On the CMDQ, parents also described children's school functioning. Fifteen percent of the children had been identified as having a learning disability, and 24% were currently enrolled in a special education program at school. Nine percent had repeated a grade, and none had skipped a grade. Eighty percent of children were described as having at least 1 specific problem in school, and 46% were identified as having 3 or more problems. Specific problems endorsed included: problems with attention and memory (57%), problems with at least 1 basic academic skill area (52%), problems completing schoolwork (35%), problems following school rules (28%), and test anxiety (20%).

Family history of mental disorder. Forty-four parents provided information about family history of psychiatric disorders on the CMDQ. Family history of mental disorder was common, with 29 (65.9%) reporting at least 1 known mental disorder in a first degree relative (parent or sibling). As shown in Table 4, the frequency of familial depression was quite high (42%), in both mothers (30%) and fathers (23%). Substance abuse, suicidality, bipolar disorder, obsessive-compulsive disorder, attention deficit disorder, and anxiety disorders were also common (9–21%) among family members of children with TTM. Family history of tic disorder and schizophrenia were uncommon. Male and female participants did not differ in terms of the rate of any family psychiatric disorder (all $ps > 0.05$). PITS total scores did not differ significantly between participants with and without a family history of any psychiatric disorder (all $ps > 0.05$).

Table 4. *Family history of psychiatric disorders, as reported by parents on the Conners-March Developmental Questionnaire (n=44)*

	n (%) Reporting			
	Mother or mother's family	Father or father's family	Siblings	Any relative
Depression	13 (29.5)	10 (22.7)	4 (9.1)	19 (42.2)
Bipolar disorder	3 (6.8)	2 (4.5)	0 (0)	5 (11.4)
Obsessive-compulsive disorder	1 (2.3)	4 (9.1)	1 (2.3)	5 (11.4)
Tic disorder	0 (0.0)	1 (2.3)	0 (0.0)	1 (2.3)
Attention deficit disorder	3 (6.8)	1 (2.3)	1 (2.3)	4 (9.1)
Anxiety disorder	5 (11.4)	1 (2.3)	0 (0.0)	5 (11.4)
Schizophrenia	1 (2.3)	0 (0.0)	0 (0.0)	1 (2.3)
Substance abuse	5 (11.4)	6 (13.6)	2 (4.5)	9 (20.5)
Suicidality	3 (6.8)	3 (6.8)	2 (4.5)	7 (15.9)
Other psychiatric disorder	4 (9.1)	3 (6.8)	3 (6.8)	9 (20.5)

Discussion

The present results suggest that, as in adult samples, pediatric TTM is a chronic and fluctuating condition that follows a relapsing and remitting course. Expanding upon the results of previous small-sample studies (Hanna, 1997; Reeve, 1999; Tay et al., 2004), most children reported pulling from 2 or more sites, most frequently the scalp, with thin or bald patches clearly visible in most children.

With regard to the impact of TTM on quality of life, children reported spending 30–60 minutes per day pulling hair, and experienced significant distress about their symptoms. However, it was surprising that avoidance of activities, while present, was rated by children as mild, particularly given the high frequency of self-reported avoidance of recreational and social activities in adult samples (Diefenbach, Tolin, Hannan et al., 2005). Adults in the Diefenbach et al. study were asked about their avoidance of a number of specific activities, whereas the present study relied on the PITS that asks a more general, open-ended question about avoidance of activities. Although one possibility is that actual avoidance of activities is low among children and adolescents with TTM, another possibility is that such avoidance is disclosed only in response to detailed questioning. It is also possible that parents would have reported greater avoidance among their children than would the children themselves (DiBartolo, Albano, Barlow, & Heimberg, 1998); unfortunately, such data were not collected systematically.

Twenty-two percent of participants denied either a sense of rising tension before pulling or a sense of relief after pulling; 15% denied both of these DSM-IV-TR criteria. Thus, participants in the present sample were somewhat more likely to meet full criteria for TTM than were those in previous, smaller studies (Hanna, 1997; King, Scahill et al., 1995; Reeve et al., 1992). Nevertheless, these results are consistent with the notion that the tension-relief cycle is not a necessary criterion for clinically significant hair-pulling in children and adolescents.

The majority of children were described as having problems in school functioning, particularly in basic skill areas as well as attention and memory. In the absence of psychometric data on the CMDQ, these data must be regarded as tentative. Parent reports of school functioning may over-estimate children's school problems, particularly in a treatment-seeking sample. However, the rather high number of children enrolled in special education programs at school is consistent with legitimate problems with academic functioning. The present data also do not necessarily indicate a specific relationship between problems in school functioning and TTM. Rather, any such difficulties could be due to comorbid symptoms such as ADHD. Although only 9% of the sample was diagnosed with ADHD during a structured interview, on rating scales parents described inattentive ADHD *symptoms* in 30% of cases. It is possible, however, that parents seeking treatment for their children (as was the case in

the present study) over-report certain symptoms compared with parents sampled in epidemiologic research.

Over one-third of the children in this sample met diagnostic criteria for at least 1 comorbid axis I disorder. This figure is somewhat lower than observations in smaller samples of children and adolescents (King, Scahill et al., 1995; Reeve et al., 1992), who reported comorbidity rates of 60–70%, as well as those reported in samples of adults (Christenson et al., 1992; Schlosser et al., 1994; Swedo & Leonard, 1992), where comorbidity rates of 45–86% have been reported. This discrepancy may be due to the fact that, for research purposes, patients who presented with comorbid conditions that, in the evaluator's opinion, were of greater severity than was the TTM, were excluded. The profile of comorbid diagnoses is consistent with that reported by King, Scahill et al. (1995), who also found a preponderance of anxiety and externalizing disorders in that sample.

TTM has been designated as part of an "obsessive-compulsive spectrum" (Hollander et al. 1996), although some researchers have suggested that TTM functionally resembles impulse control disorders (Franklin et al., 2006). In the present sample, 11% of children had a family member with OCD, weighted more heavily on the father's side. Only 7% of these children themselves met criteria for OCD. Tic disorders, also considered part of this spectrum, were rather rare in both family members and in the children themselves. In contrast, familial rates of anxiety (other than OCD) and depressive disorders were high in both children with TTM and their family members; high rates of social anxiety were particularly noteworthy and consistent with the likely social isolation experienced by some children with visible alopecia. It is noted, however, that many of the children in this sample remained socially active despite their symptoms. Only a few children were severely socially isolated. There was a rather high rate of substance use disorders and ADHD in parents of children with TTM, which may be consistent with an impulsive, rather than compulsive, model of TTM (Franklin et al., 2006). As described above, the frequency of externalizing disorders (primarily ADHD) was 10%, although the rate was 30% according to parent report. Inconsistent with the impulsivity model, however, the rate of inattentive type ADHD appeared marginally higher than the rate of hyperactive/impulsive type. Oppositional problems were also reported by a high number of parents.

Study 2: open trial of cognitive behavioral therapy

Method

Participants. Of the 46 children and adolescents who provided descriptive information, the first 22 (15 at the Institute of Living and 7 at the University of Pennsylvania) were enrolled in an open trial of CBT (the remainder were enrolled in a randomized controlled trial, the results of which have not yet been published). Mean age was 12.6 (SD=3.0) years. Seventeen (77.3%) of the children were female, and 19 (86.4%) were white. Three (13.6%) met DSM-IV-TR criteria for a comorbid anxiety disorder. None met criteria for major depressive disorder.

Materials. The primary TTM outcome measures were the NIMH-TSS and NIMH-TIS. Global clinician-rated severity of illness was assessed using the NIMH Clinical Global Impression (CGI) (Guy, 1976), a clinician-rated assessment of global illness, including severity of symptoms (CGI-S) and global improvement (CGI-I) ratings. Test-retest reliability for both subscales is good (Dahlke, Lohaus, & Gutzmann, 1992). Validity of the CGI is demonstrated by strong correlations with clinician-rated anxiety and depression symptoms (Leon et al., 1993). The CGI has been used as a treatment outcome measure for children receiving treatment for generalized anxiety disorder (Rynn, Siqueland, & Rickels, 2001), depression (Emslie et al., 1997), and OCD (Diler & Avci, 2000; Franklin et al., 1998), as well as in studies of TTM with adults (Diefenbach et al., 2006; Ninan et al., 2000). Secondary analyses were conducted using the CDI and MASC, in order to examine whether the intervention affected levels of depression and anxiety, respectively. At the post-treatment assessment, children and parents completed an exit questionnaire that included a Satisfaction Rating. Parents responded to the question, "Overall, my level of satisfaction

with quality of the treatment services that I have received is:'' Ratings were from 0 (very dissatisfied) to 7 (very satisfied). Children responded to the question, ''How happy are you with the treatment that you received?'' Ratings were from 0 (very unhappy) to 7 (very happy).

Procedure. An independent evaluator (IE) who was not involved with the child's treatment conducted all clinical assessments at pre-treatment, post-treatment, and 1-, 3-, and 6-month follow-up. IEs were 3 PhD-level clinicians with experience with TTM assessment. The same IE interviewed a given child at each assessment point. The IEs explained the risks and benefits of participation, obtained written informed consent from parents and written assent from children, and administered all measures prior to study entry. After the initial assessment, eligible participants were then referred to individual CBT. Study clinicians included 7 licensed psychologists with experience treating TTM, 3 advanced pre-doctoral psychology interns, and 3 postdoctoral fellows supervised by the first and second authors. All treatment sessions were audio-taped for supervisory purposes, and rated according to a treatment fidelity checklist developed by the first author. Fidelity ratings were used for supervisory purposes, and systematic fidelity scores are not available. Cross-site supervision was conducted on a weekly basis via conference call to ensure consistency of treatment between the 2 sites.

Treatment description. Treatment was based on a preliminary version of a CBT manual developed by the first and second authors that has subsequently been published (Franklin & Tolin, in press). Because of concerns about relapse following active treatment (Lerner et al., 1998), the present CBT program was divided into 2 phases. The first phase (*active treatment*) lasted 8 weeks and consisted of weekly individual CBT sessions with emphasis on increasing awareness of pulling, teaching new strategies for preventing pulling, and assigning homework for practicing strategies. In session, 1, the therapist established rapport, gathered information about the child's hair-pulling behavior, and provided psychoeducation. In session 2, the therapist introduced the strategies of competing response training (engaging in a behavior that is incompatible with pulling, progressively earlier in the pulling sequence) (Azrin & Nunn, 1973) and stimulus control (altering ones' environment to reduce the likelihood of pulling) (Carroll & Yates, 1981). These strategies were continued and refined through sessions 3–4. In session 5, the therapist instructed the child in progressive muscle relaxation (Bernstein & Borkovec, 1973). In session 6, cognitive restructuring (Beck, 1995) was introduced and the child was encouraged to identify and challenge maladaptive beliefs about stressful situations. Cognitive restructuring was continued into session 7, at which time the child received additional instructions for guided self-dialogue (Kendall, 1994). In session 8, the therapist discussed relapse prevention strategies (McKay, 1997; McKay et al., 1996), such as distinguishing minor slips from full-blown relapses. Covert modeling was used, with children imagining and describing themselves responding well to stressful situations without resorting to pulling. The second phase (*relapse prevention*) consisted of 4 bi-weekly sessions, interspersed with brief (15–20 minute) telephone contacts with the therapist. In this phase, the emphasis was on reminding the child to employ previously-learned strategies and to remain aware of pulling behaviors. During these sessions, the therapist reviewed the child's use of strategies, and helped trouble-shoot any problems that arose between visits.

Data analysis. Data were analyzed using SPSS 10.0. The intent-to-treat sample was analyzed with last observation carried forward. For CGI-I ratings, children who discontinued before a rating could be made were assumed not to have improved.

Results. From the 22 children enrolled in the open trial, 8 (36.4%; all female) discontinued the study (3 during active treatment, 5 during relapse prevention), leaving 14 treatment completers. Two (14.3%) children who completed treatment were lost during the follow up phase. Children who dropped out did not differ from treatment completers in terms of age, CGI-S, NIMH-TSS, or NIMH-TIS. There was a significant difference between participants who did and did not complete the study on the MASC (t (20)=2.48, $p<0.05$), with participants who discontinued the study showing lower pre-treatment levels of anxiety. None of the participants who discontinued the

Table 5. *Mean (SD) scores on outcome measures before, during, and after treatment*

	NIMH-TSS		NIMH-TIS		CGI-S		CDI		MASC	
Week 0 (pre-treatment)	8.43	(4.33)[a]	4.32	(2.44)[a]	4.09	(1.48)[a]	7.27	(6.25)[a]	42.05	(18.23)[a]
Week 4 (midpoint of active treatment)	3.77	(3.94)[b]	3.23	(2.54)[b]	3.32	(1.36)[b]	4.41	(4.94)[b]	33.91	(18.05)[b]
Week 8 (end active treatment)	4.59	(4.38)[b]	2.73	(1.93)[b]	3.27	(1.28)[b]	3.82	(4.93)[b]	32.50	(20.94)[b]
Week 16 (end relapse prevention)	4.27	(3.98)[b]	2.18	(1.92)[c]	2.68	(1.43)[c]	3.77	(5.30)[b]	32.59	(23.19)[b]
Week 20 (1 month follow-up)	3.95	(4.01)[b]	2.36	(2.08)[c]	2.73	(1.35)[c]	4.23	(5.61)[b]	32.32	(21.99)[b]
Week 28 (3 month follow-up)	3.77	(4.12)[b]	2.23	(2.14)[c]	2.77	(1.38)[c]	3.91	(5.46)[b]	34.18	(21.50)[c]
Week 40 (6 month follow-up)	3.91	(4.42)[b]	2.59	(2.34)[d]	3.05	(1.43)[c]	4.23	(5.76)[b]	33.73	(21.27)[c]

Intent-to-treat analyses with last observation carried forward. Within each column, time-points with different superscript letters are significantly different from one another ($p<0.05$). NIMH-TSS=NIMH-Trichotillomania Severity Scale; NIMH-TIS=NIMH-Trichotillomania Impairment Scale; CGI-S=Clinical Global Impression-Severity; CDI=Child Depression Inventory; MASC=Multidimensional Anxiety Scale for Children.

study were diagnosed with a comorbid anxiety disorder.

Results at each time-point for the NIMH-TSS, NIMH-TIS, and CGI-S are depicted in Table 5. For the NIMH-TSS, a repeated-measures analysis of variance (ANOVA) showed a significant main effect of time, $F(6, 126)=13.35$, $p<0.001$, partial $\eta^2=0.389$. Follow-up paired-samples *t*-tests indicated that NIMH-TSS scores decreased significantly from pre-treatment to week 4, and did not decrease further after that point. At each time-point, NIMH-TSS scores remained significantly lower than at pre-treatment. For the NIMH-TIS, a repeated-measures ANOVA showed a significant main effect of time, $F(6, 126)=11.45$, $p<0.001$, partial $\eta^2=0.353$. Follow-up paired-samples *t*-tests indicated that NIMH-TIS scores decreased significantly from pre-treatment to week 4, and again from week 8 to week 16. There was a significant increase in scores between week 28 and week 40. At each time-point, however, NIMH-TSS scores remained significantly lower than at pre-treatment.

On the CGI-S, a repeated-measures ANOVA showed a significant main effect of time, $F(6, 126)=10.65$, $p<0.001$, partial $\eta^2=0.336$. Follow-up paired-samples *t*-tests indicated that CGI-S scores decreased significantly from pre-treatment to week 4, and again from week 8 to week 16. There was, however, a marginally significant ($p=0.06$) increase in CGI-S scores from week 28 to week 40. At each time-point, CGI-S scores remained significantly lower than at pre-treatment. The percentage of children who were considered ''treatment responders'' on the CGI-I was also examined. For the intent-to-treat analyses, patients who did not have additional assessments after pre-treatment (and thus were never assigned a CGI-I rating) were conservatively assumed to have made no change. At weeks 4, 8 and 16 of treatment, the percentages of children who received a CGI-I rating of ''much improved'' or ''very much improved'' were 45.4%, 54.5% and 77.3%, respectively. During the follow-up assessments at weeks 20, 28 and 40, the percentages were 72.7%, 68.2% and 63.6%, respectively. Of the 12 patients classified as ''responders'' at the end of active treatment (week 8), 3 (25.0%) lost that designation during the follow-up phase. Of the 17 patients classified as ''responders'' at the end of the relapse prevention phase (week 16), 4 (23.5%) lost this designation during follow-up.

For a more stringent index of treatment response, participants were classified as ''excellent responders'' if their CGI-S scores were mild or better, and their CGI-I scores were ''very much improved.'' Using this criterion, only 2 (9.1%) were classified as ''excellent responders'' at the end of active treatment (week 8). However, 7 (31.8%) were classified as ''excellent responders'' at the end of the relapse prevention phase, and this percentage remained the same at each

follow-up assessment. None of the children classified as "excellent responders" at week 8 or 16 lost that designation during follow-up.

For the CDI, a repeated-measures ANOVA showed a significant main effect of time, $F(6, 126)=7.03$, $p<0.001$, partial $\eta^2=0.251$. Follow-up paired-samples *t*-tests indicated that CDI scores decreased from pre-treatment to week 4, and did not decrease significantly at subsequent assessments. At each time-point, CDI scores remained significantly lower than at pre-treatment. For the MASC total score, a repeated-measures ANOVA showed a significant main effect of time, $F(6, 126)=7.17$, $p<0.001$, partial $\eta^2=0.254$. Follow-up paired-samples *t*-tests indicated that MASC scores decreased from pre-treatment to week 4, and did not decrease significantly at subsequent assessments. There was a significant increase in MASC scores from week 20 to week 28. At each time-point, MASC scores remained significantly lower than at pre-treatment.

Ratings on the 0–7 Satisfaction Rating were analyzed for children and parents at post-treatment. Children ($M=6.25$, $SD=1.39$) and parents ($M=6.75$, $SD=0.46$) both gave high ratings of satisfaction with treatment.

Discussion

The results of the present study, although preliminary, suggest that a brief course of CBT is promising for children and adolescents with TTM. As described previously, 3 controlled trials attest to the efficacy of CBT for adults with TTM (Azrin et al., 1980; Ninan et al., 2000; van Minnen et al., 2003); however, examinations of CBT in pediatric samples have been limited thus far to case reports. In the present sample, participants demonstrated significant reductions in clinician-rated hair-pulling severity. In addition, at the end of the treatment program, 77% of patients were classified as "treatment responders" on the CGI-I, and 32% were classified as "excellent responders" (i.e. they were rated as very much improved, and their overall severity was mild or better). At the end of the 6-month follow-up period, the percentages were 63% and 32%, respectively. Self-ratings of depression and anxiety also appeared to decrease over the course of treatment.

Of particular concern was the likelihood of relapse following successful treatment, as has been documented in a previous open trial for adults (Lerner et al., 1998). In that study, two-thirds of the treatment responders appeared to relapse during a long follow-up period. As many as half of adult patients responding to group treatment appear to lose their treatment gains during follow-up (Diefenbach et al., 2006; Mouton & Stanley, 1996). To address this concern, a relapse prevention phase was added to the present study, in which patients met with the therapist on a bi-weekly schedule for 8 more weeks after the active treatment, during which time no new interventions were introduced but emphasis was placed instead on reinforcing the use of already-acquired skills. Although overall scores remained well below pre-treatment levels and the majority of patients were still classified as responders at the end of the follow-up period, there was some suggestion of return of symptoms between the 3-month and 6-month follow-up assessments, and approximately one-quarter of patients classified as "treatment responders" lost this designation over the 6-month follow-up phase. It is perhaps particularly noteworthy that none of the patients who attained "excellent responder" status at week 16 lost this designation during the follow-up phase, suggesting that the strength of initial treatment response may be an important predictor of maintenance of gains after treatment withdrawal. This finding is convergent with observations about the maintenance of treatment gains over time in other disorders (e.g. OCD; Simpson et al., 2004).

The drop-out rate of 36% is somewhat higher than that seen in previous studies of CBT for adult TTM (Diefenbach et al., 2006; Ninan et al., 2000; van Minnen et al., 2003), although it is identical to that in the Lerner et al. (1998) study. Unfortunately, detailed information about reasons for drop-out is not available. It was surprising that patients with lower levels of baseline anxiety, and without a co-morbid anxiety diagnosis, appeared more likely to drop out. One possible explanation is that these children are less bothered by their condition (even though the objective severity of the condition is the same) and therefore are less likely to follow through with treatment. The high drop-out rate is a limitation of the present study, and additional interventions to reduce attrition may need to be explored. It is also

possible that the drop-out rate reflects a limitation of the treatment itself, although the satisfaction ratings suggest that the treatment was highly acceptable to the patients and parents who remained in the study. To the extent that larger studies continue to demonstrate a high attrition rate, additional strategies such as motivational enhancement (Miller & Rollnick, 2002) might be considered, as has been done successfully for adults with OCD (Maltby & Tolin, 2005).

A further limitation is the fact that IEs were not blind to treatment condition or to time-point. One option is to use different IEs for each child at each time-point, thus keeping IEs blind as to whether the child was beginning, in the middle of, or ending treatment. However, in the context of this initial open trial, it was felt that the need for the IEs to establish continued rapport with the children and their families, as well as to assess degree of clinical improvement by comparing findings to baseline, outweighed this concern. A randomized controlled trial, using IEs blind to treatment condition, would help reduce the potential for interviewer bias.

General discussion

The baseline psychopathology data from study 1 highlight the chronic and serious nature of TTM in treatment-seeking pediatric samples. In addition to the visible alopecia and substantial distress associated with hair-pulling itself, a large percentage of children with TTM were described as having problems with school functioning and comorbid psychopathology. Results from the open trial of CBT, although preliminary, are certainly encouraging, with the majority of children showing a favorable treatment response. The addition of extended lower-frequency sessions emphasizing relapse prevention appears to help, as most children were able to maintain their treatment gains after treatment was withdrawn. Although no data are available regarding changes in school functioning, the significant decreases in levels of depression and anxiety suggest that effective treatment for TTM may also have salutary effects on comorbid symptoms.

The empirical literature on TTM is rather sparse, even more so for pediatric TTM. The next major question regarding CBT is whether this treatment is more effective than wait list, placebo, or alternative treatments. A second important question is whether the various elements of CBT (e.g. self-monitoring, competing response training, stimulus control, relaxation, cognitive restructuring) are equally useful. Observations during this trial suggest that children perceive self-monitoring, competing response training, and stimulus control as more helpful than they do relaxation training and cognitive restructuring (Tolin, Franklin, & Diefenbach, 2002). However, dismantling studies are needed to investigate this possibility more closely. It has been suggested that certain elements of CBT might be considered "core techniques" and applied to most or all TTM patients, whereas other elements might be considered "optional techniques" that may be employed on an as-needed basis (Franklin et al., 2006). It is also possible that parent and family interventions, which were not included in the current study, may be helpful to enhance the treatment of CBT for pediatric TTM. Additional research is needed to clarify which specific interventions are efficacious for different children.

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