Randomized Trial of Parent Management Training in Children With Tic Disorders and Disruptive Behavior

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ABSTRACT

Oppositional, defiant, and disruptive behaviors are common in clinical samples of children with tic disorders. In this study, we sought to evaluate the short-term efficacy of a structured parent training program in children with tic disorders accompanied by disruptive behavior. Children with tic disorders and at least a moderate level of disruptive behavior were randomly assigned to a 10-session structured parent management training program or to continue treatment as usual. Twenty-four children (18 boys and 6 girls) between the ages of 6 and 12 years (mean 8.9 ± 2.0 years) were enrolled; 23 subjects completed the study. At baseline, subjects showed moderate to severe levels of oppositional and defiant behavior. Twenty subjects (83%) were on stable medication. The parent-rated Disruptive Behavior Rating Scale score decreased by 51% in the parent management training group compared with a decrease of 19% in the treatment as usual group (P < .05). On the Improvement scale of the Clinical Global Impression, a rater masked to treatment assignment classified 7 of 11 subjects who completed parent management training as much improved or very much improved compared with 2 of 12 subjects in the treatment as usual group (Fisher exact test, P < .05). These results suggest that parent management training is helpful for short-term improvement in disruptive behavior problems in children with tic disorders. Larger randomized clinical trials are needed. (*J Child Neurol* 2006;21:650–656; DOI 10.2310/7010.2006.00159).

Tourette syndrome is a neurologic disorder of childhood onset defined by an enduring pattern of motor and phonic tics. A diagnosis of chronic tic disorder is applied to children who exhibit motor or phonic tics, but not both, for at least a year. The tics of Tourette syndrome and chronic tic disorder vary from mild to

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severe across individuals and show a fluctuating course within individual patients. In most cases, tics decline in severity by late adolescence. Studies on the prevalence of Tourette syndrome in school-aged children show a range of 1 to 10 per 1000, although several studies indicate a prevalence in the narrower range from 3 to 8 per 1000. The prevalence of chronic tic disorder is less well established. Up to 80% of clinically referred children with Tourette syndrome also have co-occurring disruptive behavior problems, including explosive anger outburst, argumentativeness, and noncompliance. When present, disruptive behavior can substantially impair adaptive functioning.

The primary treatment for Tourette syndrome has been medication directed at tics, attention-deficit hyperactivity disorder (ADHD), or obsessive-compulsive disorder. However, few pharmacologic studies have evaluated the effect of medication on disruptive behavior. Notable exceptions include an openlabel study of paroxetine, which was inconclusive, and a small open-label study of olanzapine showing a modest reduction in aggressive behavior.

The relationship between tics and disruptive behavior is unclear and potentially puzzling to parents, teachers, and

clinicians. ¹⁰ Teachers might confuse impulsive calling out in the classroom with vocal tics. Given the involuntary nature of tics, some parents might wonder whether disruptive and explosive behaviors are also outside the child's control. Parents who struggle with this uncertainty might not provide clear limits for their child's behavior. Ironically, impulsive and explosive children, who might need more structure in the home and classroom, might receive inconsistent adult direction. The failure to achieve age-appropriate impulse control and emotional regulation might contribute to greater disability.

The notion that parent management training can improve disruptive and noncompliant behavior in these children is rooted in a considerable body of research. 11,12 This research indicates that disruptive behavior in children can be maintained by maladaptive parent-child interaction.¹³ For example, to avoid conflict with their children, parents can fail to limit unacceptable behavior. At other times, the application of parental authority can be overly harsh, which can promote anger and defiance in the child. Perhaps because of an accumulation of tension and hostility, parents can fail to acknowledge positive behavior in some situations. In parent management training, parents are taught principles of behavior modification, including positive reinforcement, setting clear expectations and limits, the use of mild punishment such as "time-outs," and how to manage point systems to promote selected behaviors. Thus, the goal of parent management training is to reduce the child's noncompliance and promote positive behavior. 12,14

Despite the common occurrence of disruptive behavior in children with Tourette syndrome, parent management training has not been evaluated in this population. In this pilot study, we sought to evaluate the short-term efficacy of a structured, 10-session parent management training program in children with tic disorders accompanied by moderate to severe disruptive behavior. We hypothesized that parent management training would be superior to treatment as usual in reducing disruptive behavior in children with tic disorders.

METHODS

Design

A research assistant who was not involved in the study intervention or in the outcome assessment randomly assigned subjects to a structured 10-week parent management training program in addition to treatment as usual or to continue with treatment as usual only. Prior to randomization, both the parent management training group and the treatment as usual group received a structured psychoeducational session about tic disorders developed by the investigators for this project. This 90-minute session, which was delivered by the same therapist in all cases (D.F.), reviewed the contemporary view of tic disorders and the common co-occurrence of ADHD, obsessive-compulsive behavior, and disruptive behavior. The session did not include any information about parental management of disruptive behavior. Families of subjects randomized to treatment as usual were offered parent management training after the randomized trial.

Setting and Subjects

Subjects between 6 and 12 years of age were recruited from the Tic Disorders Clinic at the Yale Child Study Center. To be eligible for the study, children had to be healthy and had to display at least a moderate level of disruptive behavior, as evidenced by a score of 3.5 on

the parent-rated Home Situations Questionnaire¹⁴ and a score of at least moderate (≥ 4) on the severity item of the Clinical Global Impression scale. Concomitant medication was allowed if stable for at least 6 weeks and if there were no planned changes for 3 months. Similarly, concurrent child psychotherapy was allowed to continue, but parents were asked not to initiate any new psychotherapy for their child during the 3-month intervention period. Children who lived more than 2 hours from the medical center, who had an IQ below 85, or who had an untreated condition that warranted standard treatment (eg, tics, ADHD, obsessive-compulsive disorder, depression, bipolar disorder, or psychosis) were excluded. The study was approved by the Yale Institutional Review Board, and parents gave written informed consent prior to study entry. Families were paid \$20 for each assessment or treatment visit.

Procedures

Recruitment

All subjects were seen for a standard clinical evaluation in the Tic Disorder Clinic at the Yale Child Study Center. This 2-hour assessment, involving the child and primary caregivers, was conducted by a multidisciplinary team with expertise in Tourette syndrome and related conditions. In addition to the face-to-face interview, the evaluation session was augmented by parent ratings of tic and obsessive-compulsive symptom severity and family, medical, and developmental histories. Children who appeared to be eligible for the study were referred to the research team, and interested families were scheduled for screening.

Baseline Assessment

The screening procedure took place over two visits, beginning with informed consent. Assessments included a review of the medical history; the Schedule for Affective Disorders and Schizophrenia for School-Age Children, ¹⁵ the Kaufman Brief Intelligence Test (K-BIT), ¹⁶ the Child Behavior Checklist, ¹⁷ and other parent ratings; child self reports; and clinician ratings (see the Measures section). Data were also collected on family composition, race, parental occupation, and education, as well as the child's current and prior medications, concurrent psychotherapy, and school interventions. These baseline assessments were conducted by a child psychiatric nurse practitioner (D.H.C. or A.L.R.) who was not part of the intervention program and was masked to treatment assignment. After a review of all available information, two clinicians independently assigned *Diagnostic and Statistical Manual of Mental Disorders-IV* (DSM-IV)^{18,19} diagnoses; disagreements were resolved by consensus.²⁰

Measures

The Schedule for Affective Disorders and Schizophrenia for School-Age Children is a semistructured diagnostic interview for children that has been revised in accordance with the *DSM-IV*. The Schedule for Affective Disorders and Schizophrenia for School-Age Children was administered at the screening visit to rule out the presence of another psychiatric diagnosis requiring treatment.

The Child Behavior Checklist is a 118-item parent-rated instrument designed to assess behavioral problems and competencies in children between the ages of 4 and 16 years. Problem scales include eight narrow-band syndrome scales (Withdrawn, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Behavior, Aggressive Behavior), three broadband scales (Internalizing Problems, Externalizing Problems, Total Problems), and three competency scales (Social, School, and Community Activities). Based on normative data, scores for a given child can be compared with

the population mean of 50 ± 10 . Higher scores on Problem scales indicate greater psychopathology; lower scores on Competency scales indicate lower adaptive functioning. Thus, scores of 1.5 SD above (Problem scales) or below (Competency scales) are regarded as clinically significant.

The Kaufman Brief Intelligence Test (K-BIT) was used to obtain an estimate of intelligence. This brief test shows high correlation with more detailed intelligent tests.

Primary Outcome Measures

The Disruptive Behavior Rating Scale 14 is an eight-item parent-rated scale keyed to the DSM-IV criteria for oppositional defiant disorder. Examples of relevant items on this scale include loses temper, argues with adults, actively defies adult requests, is touchy or easily annoyed, and is angry and resentful. The scale asks the parent to rate each item using a 4-point response format, where 0 = never or rarely, 1 = sometimes, 2 = often, and 3 = very often. Scores of 12 and higher are considered clinically significant. This scale was selected because it is sensitive to change with treatment and has been used in other clinical studies. 21

The Clinical Global Impression-Severity scale ²² is a clinician-rated scale scored from 1 (normal, not at all ill) to 7 (among the most extremely ill patients). It was rated at screening and again at baseline to ensure eligibility. The Clinical Global Impression-Improvement scale, which reflects the clinician's assessment of overall progress compared with baseline, is a 7-point scale rated from very much improved (score of 1) through no change (score of 4) to very much worse (score of 7). The Clinical Global Impression-Improvement scale was rated within 2 weeks of post-treatment by a clinician who was masked to treatment assignment. Ratings of much improved or very much improved defined a positive response. All other scores were classified as a negative response.

To rate the Clinical Global Impression-Improvement scale, the independent evaluators (D.H.C. and A.L.R.) used all available information, including parent ratings and parent-nominated target symptoms. At baseline, parents were asked to describe the child's two most pressing problems. These parent-nominated target symptoms were further documented according to their frequency (episodes per day or per week), intensity (duration and appearance of the behavior), and impact (degree of disruption at home and school). For example, the target symptom interview might reveal explosive outbursts that occur three to five times per day. These outbursts can last from 10 to 30 minutes and be accompanied by yelling, screaming, slamming doors, and threats of harm, with occasional physical aggression or property destruction. The target symptoms were reviewed again at end point.

Secondary Outcome Measures

The Home Situations Questionnaire¹⁴ is a 20-item questionnaire used to assess noncompliance across a range of everyday situations. Parents are asked to answer yes or no for each item. Those items marked yes are then rated on a severity scale ranging from 1 (mild) to 9 (severe). Thus, the measure yields two scores: the number of yes items and the mean severity score (the raw severity score divided by the number of items endorsed as a problem). The scale has been shown to discriminate between children with and without behavioral problems. ^{24,25} Based on population data, a mean severity score of 3.5 or higher on the Home Situations Questionnaire indicates significant noncompliance. The Home Situations Questionnaire is sensitive to change, ²⁶ but it has not been used as extensively as the Disruptive Behavior Rating Scale for this purpose. In the current study, the Home Situations Questionnaire was used to define eligibility and as a secondary outcome measure.

The Yale Global Tic Severity Scale²⁷ is a clinician-rated, semi-structured interview for evaluating the current severity of tic symptoms. Motor and phonic tics are separately rated according to number, frequency, intensity, complexity, and interference on a 6-point ordinal scale (0 = absent; 1 through 5 for severity), yielding three scores: Total Motor (0–25), Total Phonic (0–25), and Total Tic (0–50).

The Children's Yale-Brown Obsessive Compulsive Scale (CYBOCS)²⁸ is a semistructured clinician-rated scale designed to rate the current severity of obsessive-compulsive disorder in children and adolescents. Once the current obsessions and compulsions are identified, Time Spent, Interference, Distress, Resistance, and Control are rated separately for obsessions and compulsions. Each item scored on a 5-point scale from 0 (least symptomatic) to 4 (most symptomatic), with possible scores ranging from 0 to 20 for obsessions, 0 to 20 for compulsions, and 0 to 40 for the combined total score.

The Parenting Stress Index-Short Form (PSI-SF)²⁹ is a 36-item questionnaire used to measure the impact of chronic tic disorder on the family. Because this scale has been shown to be sensitive to short-and long-term treatment effects in children with disruptive behavior disorders, ³⁰ it was repeated at end point.

The Children's Inventory of Anger³¹ is a 40-item child self-report rated from 1 (no anger) to 4 (extreme anger). Children are asked to evaluate their response to potentially provoking events (eg, "Someone cuts in front of you in a lunch line"). Although the Children's Inventory of Anger has not been used in studies of parent management training, it has demonstrated sensitivity to change in psychosocial interventions with children.³²

Interventions

Parent Management Training

The parent management training manual used in this study was developed by Barkley. ¹⁴ This is a structured curriculum consisting of 10 sessions intended to improve parental competence in dealing with child behavior problems, increase parental understanding about the origins of noncompliant and defiant behavior, improve the child's compliance with parental instructions, and decrease family conflict. The core skills include providing positive reinforcement for appropriate behavior, communicating directions effectively, and being consistent with consequences for disruptive behaviors. Parents learn techniques such as positive attending, selective ignoring, token economies, and time-out. Parenting skills are taught through modeling, role-play, and corrective feedback provided by the therapist. Assignments to implement new parenting skills at home were given after each session. Parent management training was conducted by one of two experienced clinicians (D.F. and V.H.).

Treatment as Usual

Subjects in both treatment conditions continued to see their treating clinicians according to usual practice. Treatment as usual at the Yale Child Study Center includes parent education (concerning the natural history of Tourette syndrome and related disorders), clinical monitoring of specific symptoms, medication management, and school consultations, if needed. These interventions were provided independent of the study, according to the needs of the child and family and at the discretion of the treating clinician. Subjects in both conditions were also allowed to continue to receive ongoing clinical services elsewhere in the community, including child individual psychotherapy and school-based mental health services. Parents were asked not to initiate new

treatments or to stop existing treatments during the study period. Nonetheless, changes in clinical services were monitored throughout the study.

Treatment Fidelity

Treatment fidelity was addressed in three ways. First, although both study therapists had previous experience with Barkley's manual, ¹⁴ the parent management training program was applied in four pilot cases to determine if any modifications would be required in this clinical population. The only modification was the development of the Tourette syndrome psychoeducation module. As noted above, this educational session was given to both groups prior to randomization. Second, the accuracy of parent management training implementation was monitored during regular discussions at the research team meetings. Third, all parent management training sessions for two subjects (one for each therapist) were observed by an independent rater using a session-by-session checklist. This review showed greater than 90% adherence to the manual.

Analytic Strategy

Two-way repeated measures analysis of variance (ANOVA) was used for continuous measures (eg, Disruptive Behavior Rating Scale, Home Situations Questionnaire, Yale Global Tic Severity Scale). Using two treatment groups and two time points, the ANOVA tests for a significant interaction between treatment group and time. To compare the results of this trial with those of previous studies, we also calculated effect size for the primary outcome measure (determined by the change in the Disruptive Behavior Rating Scale from baseline to end point in parent management training minus the change in treatment as usual divided by the standard deviation for the whole sample at end point). Because only two data points (baseline and end point) were available for this study, data from one subject who withdrew from the parent management training condition were not included in the analyses involving repeated measures. The proportion of subjects in each group showing a positive response was evaluated by chi-square (Fisher exact test when cell sizes

were less than 5). Statistical significance for all analyses was set at alpha = 0.05 for a two-tailed test.

RESULTS

One hundred seventy-two children between the ages of 6 and 12 years were seen in the Tic Disorder Clinic over a 4-year study period; 48 subjects appeared to be potentially eligible, and 29 families agreed to participate. Of these, five subjects did not meet the inclusion criteria or refused to participate. Thus, 24 subjects (18 boys and 6 girls; mean age 8.9 ± 2.0 years) participated in the trial. Eighteen subjects met the criteria for Tourette syndrome and six met the criteria for chronic tic disorder. Other DSM-IV diagnoses included ADHD (n = 10), obsessive-compulsive disorder (n = 4), and oppositional defiant disorder (n = 20). Eight children also had obsessive-compulsive symptoms. There were no significant differences in demographic or clinical characteristics across the two groups at baseline, including mean scores on the Disruptive Behavior Rating Scale; frequency of ADHD, oppositional defiant disorder, or obsessivecompulsive disorder; or frequency of medication use (Table 1).

Table 1 shows that children in this study had high levels of disruptive behavior, as evidenced by the Disruptive Behavior Rating Scale score of 16.1 ± 3.6 and the Child Behavior Checklist Externalizing score of 68.2 ± 9.4 . The level of tic severity was mild, as evidenced by the Yale Global Tic Severity Scale total tic score of 18.2 ± 8.7 . Noting that more than 80% of the children were on medication, tic severity might have been mitigated by medication.

The mean Disruptive Behavior Rating Scale score decreased from 16.7 ± 3.91 at baseline to 8.2 ± 4.75 at end point in the parent management training group. This 51% improvement was significantly better than the 19% decline in the treatment as usual control group (from 15.5 ± 3.37 at baseline to 12.5 ± 6.64 at end point; effect size = 0.96, two-way repeated measures ANOVA

Table 1. Demographic and Clinical Characteristics by Treatment Group at Baseline

Characteristic	<i>PMT</i> (n = <i>12)</i> <i>Mean (SD)</i>	TAU (n = 12) Mean (SD)	t- <i>Test</i>	Р
Age (yr)	8.9 (1.58)	8.9 (2.37)	$t_{22} = 0.09$.93
Family Hollingshead	37.1 (12.19)	33.4 (11.48)	$t_{22} = 0.76$.46
K-BIT	112.3 (11.72)	105.3 (8.77)	$t_{20} = 1.59$.13
YGTSS total tic score	18.0 (6.77)	18.4 (10.62)	$t_{22} = -0.12$.91
CYBOCS total score	7.4 (9.21)	7.7 (8.58)	$t_{20} = -0.10$.93
CBCL				
Internalizing symptoms	izing symptoms 63.2 (14.31)		$t_{22} = 0.45$.66
Externalizing symptoms	69.4 (8.71)	67.0 (10.07)	$t_{22} = 0.63$.54
Total competency score	36.0 (6.58)	39.5 (8.13)	$t_{22} = 1.15$.27
	n <i>(%)</i>	n <i>(%)</i>	χ² (Fisher Exact Test) P	
Boys	8 (67)	10 (83)	.35	
Caucasian	12 (100)	11 (92)	.31	
Special education	4 (33)	4 (33)	1.00	
Single-parent family	2 (17)	2 (17)	1.00	
Medication	11 (92)	9 (75)	.27	
α_2 -Agonist	7 (58)	6 (50)	.68	
SSRI	4 (33)	2 (17)	.35	
Antipsychotic	2 (17)	2 (17)	1.00	
Atomoxetine	moxetine 1 (8)		.31	
Concomitant psychotherapy	7 (58)	2 (17)	.09	

ADHD = attention-deficit hyperactivity disorder; CBCL = Child Behavior Checklist; CYBOCS = Children's Yale-Brown Obsessive Compulsive Scale; K-BIT = Kaufman Brief Intelligence Test; PMT = parent management training; SSRI = selective serotonin reuptake inhibitor; TAU = treatment as usual; YGTSS = Yale Global Tic Severity Scale.

Measure	<i>PMT</i> (n = 12)		TAU (n = 12)			
	Baseline Mean (SD)	End Point Mean (SD)	Baseline Mean (SD)	End Point Mean (SD)	F Test	Р
DBRS	16.5 (4.03)	8.2 (4.75)	15.6 (3.37)	12.5 (6.64)	F(1,21) = 4.66	< .05
HSQ	5.9 (0.89)	3.5 (1.71)	5.5 (0.78)	4.04 (1.53)	F(1,21) = 1.69	.21
PSI-SF	104.4 (23.18)	94.5 (12.47)	97.2 (18.44)	89.8 (24.89)	F(1,20) = .09	.77
YGTSS	17.2 (6.57)	15.4 (7.28)	18.4 (10.62)	14.9 (9.59)	F(1,21) = .48	.49
CHiA	100.0 (19.87)	94.5 (24.89)	88.7 (19.50)	85.7 (29.44)	F(1,18) = .09	.77

Table 2. Baseline and End-Point Scores for the Parent Management Training and Treatment as Usual Groups

CHiA = Child Inventory of Anger; DBRS = Disruptive Behaviors Rating Scale; HSQ = Home Situations Questionnaire; PMT = parent management training; PSI-SF = Parenting Stress Index-Short Form; TAU = treatment as usual; YGTSS = Yale Global Tic Severity Scale total tic score.

 $[F(1,21)=4.66,\ P<.05])$. On the Improvement item of the Clinical Global Impression scale, the independent evaluator rated 7 of 11 subjects (64%) in the parent management training condition as much improved or very much improved compared with 2 of 12 (17%) in the treatment as usual condition (Fisher exact test, P<.05). As shown in Table 2, there were no differences between parent management training and treatment as usual on the secondary outcome measures.

DISCUSSION

Disruptive behavior is common in school-aged children with tic disorders and is associated with substantial impairment. ^{4,6} Despite these observations, treatment and clinical research in this population have focused primarily on tics. This study examined the effects of parent management training, an empirically supported psychosocial treatment for disruptive behavior disorders, in school-aged children with a chronic tic disorder. Parent management training was associated with a 51% decline in parent-rated oppositional and disruptive behavior compared with a 19% decline in the control group (effect size = 0.96). In addition, clinicians who were masked to treatment assignment rated 64% of the children in parent management training as much improved or very much improved compared with 17% of children in the control group.

The effect size of 0.96 observed in this study is comparable to the large effects reported in studies of parent management training with children with defiant and oppositional behavior who do not have a tic disorder. For example, randomized studies of parent management training in children 3 to 8 years of age^{33,34} and children 7 to 13 years old³⁰ reported effect sizes of 1.0 or greater on parent-rated outcome measures. Furthermore, a meta-analysis of 26 randomized controlled studies of parent management training revealed an overall effect size of 0.86.³⁵

Disruptive behavior in children with Tourette syndrome can also be associated with explosive outbursts. Parents and clinicians often wonder whether tics and explosive outbursts have the same underlying cause. If so, the explosive behavior can be viewed as involuntary in the same way that tics are involuntary. However, the link between the neurobiology of tics and disruptive and explosive behavior is unclear. The positive results in this study suggest that disruptive behavior in children with tic disorders is indeed similar to that observed in children without tics. Further study is needed in children and adolescents with Tourette syndrome for whom the primary complaint is explosive behavior.

Findings from clinical samples indicate that disruptive behavior is more likely to be present in children with Tourette syndrome plus ADHD than Tourette syndrome alone. The small sample size in the current study did not permit a separate evaluation of parent management training in children with ADHD. By design, children with untreated ADHD were excluded from the study. Indeed, the 10 children with ADHD in the current study were on stable medication, suggesting that their ADHD symptoms were well managed. Our data indicate, however, that parent management training exerted additive effects on disruptive and noncompliant behavior to ongoing pharmacotherapy. Large-scale studies of parent management training in children with tic disorders are needed to evaluate the additive impact of parent management training on medication treatment and the effectiveness of parent management training in clinically important subgroups such as those with ADHD.

There was no difference between groups on the Home Situations Questionnaire, a measure of child noncompliance with parental requests and household rules. However, the 40% reduction in the Home Situations Questionnaire score from baseline to post-treatment within the parent management training group is consistent with the 32% reduction on this measure in children with ADHD. ²⁶ There were modest improvements in parenting stress and self-reported anger by children in the parent management training group. Perhaps because of the small sample size, these improvements were not significant. Future studies with larger samples could examine these outcomes. There was no change in tic severity, suggesting that parent management training is specific for disruptive behavior and might not be directly relevant for the treatment of tics.

LIMITATIONS

The major limitation of this study is the small sample size. However, it provides initial evidence that parent management training for children with Tourette syndrome and disruptive behavior yields results similar to those reported in children with disruptive behavior who do not have tic disorders. The sample included mostly middle-class Caucasian children from intact families, and the results might not be generalizable to children with disruptive behavior who are of lower socioeconomic status. Another limitation is that only baseline and immediate post-treatment data were collected in this study. Therefore, the long-term benefits of parent management training in children with Tourette syndrome and disruptive behavior are unclear. Although we used an existing manual and only two therapists, we did not rigorously monitor treatment integrity. Questions about

generalizability, longer-term benefit, and treatment integrity await answers from large-scale, multisite studies in this population.

CONCLUSIONS

This study provides preliminary evidence that parent management training can be effective for the disruptive and explosive behavior often observed in clinical samples of children with Tourette syndrome. These results support the view that the disruptive and explosive behavior in children with Tourette syndrome is similar to that observed in other children and stand in contrast to the view that this behavioral pattern is qualitatively different in Tourette syndrome. Thus, the integration of parent management training in the treatment of children with Tourette syndrome accompanied by disruptive behavior is appropriate, although more study is warranted.

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