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# Modified comprehensive behavioral intervention for Tics: treating children with Tic disorders, co-occurring ADHD, and psychosocial impairment

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BOSTON UNIVERSITY  
SCHOOL OF MEDICINE

Thesis

**MODIFIED COMPREHENSIVE BEHAVIORAL INTERVENTION FOR TICS:  
TREATING CHILDREN WITH TIC DISORDERS, CO-OCCURRING  
ADHD, AND PSYCHOSOCIAL IMPAIRMENT**

by

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B.A., University of California, Berkeley, 2017

Submitted in partial fulfillment of the  
requirements for the degree of  
Master of Science

2021

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To my mentor, Dr. Greenberg, thank you for allowing me to take part in this project and for acting as my teacher these past few months.

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AND PSYCHOSOCIAL IMPAIRMENT**

**CAROLINE ALBRIGHT**

**ABSTRACT**

**Objective:** To evaluate the feasibility and acceptability, and preliminary efficacy of Modified Comprehensive Behavioral Intervention for Tics (MCBIT) therapy for patients with persistent chronic tic disorders (CTD) and co-occurring attention deficit hyperactivity disorder (ADHD).

**Method:** Seventeen child and adolescent patients aged 10-17 with CTD and co-occurring ADHD were randomly assigned to the MCBIT group (n=9) or to a control group, where they received traditional Comprehensive Behavioral Intervention for Tics (CBIT) therapy (n=8). Both groups received ten fifty-five-minute weekly treatment sessions, and two fifty-five-minute biweekly relapse prevention sessions.

**Results:** Sixteen of the seventeen participants completed the study, and acceptability ratings in both treatment groups were high with no significant differences in expectation of improvement. The MCBIT and CBIT groups in combination showed significant improvement in measures of tic severity, ADHD symptom severity, and tic impairment and group differences were not statistically significant.

**Conclusion:** The results indicate that MCBIT treatment is feasible and acceptable for youth with CTD and ADHD, and the findings demonstrate preliminary support for the modified CBIT treatment's efficacy in reducing tic and ADHD symptoms and improving tic-related quality of life. Additional studies with more participants are warranted to further examine the role of a modular behavioral treatment approach in targeting commonly co-occurring disorders simultaneously and successfully.

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## LIST OF ABBREVIATIONS

ADHD	Attention-Deficit/Hyperactivity Disorder
ASAP	Adjunctive Services and Attrition Prevention
ASD	Autism Spectrum Disorder
CBIT	Comprehensive Behavioral Intervention for Tics
CBT	Cognitive Behavioral Therapy
CD	Conduct Disorder
CDI	Children’s Depression Inventory
CER	Client Expectancy Rating
CGI-I	Clinical Global Impression – Improvement
CGI-S	Clinical Global Impression – Severity
Concom	Concomitant Medication and Therapy Questionnaire
CSQ	The Caregiver Strain Questionnaire
CSQ-Satis	Client Satisfaction Questionnaire
CTD	Chronic Tic Disorders
CTIM-P	Child Tourette’s Syndrome Impairment Scale
CY-BOCS	Children’s Yale-Brown Obsessive-Compulsive Scale
DSM-V	Diagnostic and Statistical Manual of Mental Disorders
EOT	End of Treatment
ERQ-CA	Emotion Regulation Questionnaire for Children and Adolescents
HRT	Habit Reversal Therapy
ITT	Intention-to-Treat

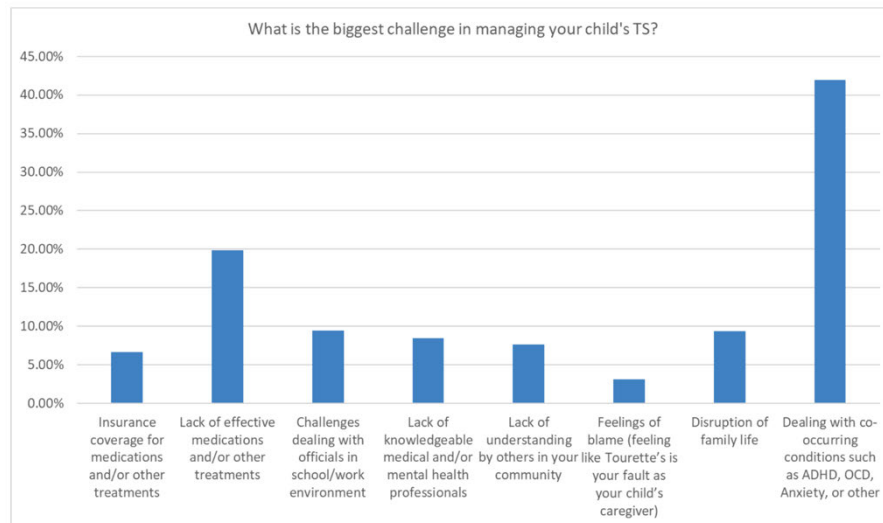
LWT .....	Living With Tics
MCBIT .....	Modified Comprehensive Behavioral Intervention for Tics
MGH .....	Massachusetts General Hospital
MINI-KID....	Mini International Neuropsychiatric Interview for Children and Adolescents
OCD .....	Obsessive-Compulsive Disorder
ODD .....	Oppositional Defiant Disorder
PedsQL .....	Pediatric Quality of Life Inventory-Child Version
PMVT.....	Persistent Motor or Vocal Tic Disorder
PTQ.....	Parent Tic Questionnaire
QoL .....	Quality of Life
RCT.....	Randomized Controlled Trial
TS.....	Tourette Syndrome
VAS.....	Vanderbilt Assessment Scale
WASI.....	Wechsler Abbreviated Scale of Intelligence
YGTSS .....	Yale Global Tic Severity Scale

## INTRODUCTION

### Background of Tic Disorders

Tic disorders, such as Tourette syndrome (TS) and persistent motor or vocal tic disorders (PMVT) (together henceforth CTD), are developmental neuropsychiatric disorders that consist of multiple motor or vocal tics that persist for at least one year. CTD affect about ~2% of the population (Scahill et al., 2014 and Scharf et al., 2015). Tourette Syndrome (TS) is a neuropsychiatric syndrome with childhood-onset characterized by the presence of motor and vocal tics. To meet DSM-5 criteria for TS, one must have at least two motor tics (e.g. blinking, shoulder shrugging) and at least one vocal tic (e.g. coughing, throat clearing) that occur for at least one year, and are not secondary to another medical condition or medication (American Psychiatric Association [APA], 2013). The criteria for persistent motor or vocal tic disorders (PMVT) are similar to that of TS, except that the individual has to have either motor *or* vocal tics (American Psychiatric Association [APA], 2013). Studies have shown up to 90% of individuals with TS have co-occurring conditions, with at least 50% having co-occurring ADHD (Hirschtritt et al., 2015 and Specht et al., 2011). Tic disorders are often associated with significant psychosocial consequences, including reduced self-esteem and reduced quality of life (Rowe et al., 2013 and Storch et al., 2007). Youth with TS and their parents also frequently report impairments in daily functioning, which is characterized by the inability to perform age-appropriate tasks in various domains including school, home and social. (Storch et al 2007). In youth with CTD, additional ADHD symptoms associate with increased social difficulties, decreased quality of life, and greater

psychopathological impairment (McGuire et al., 2013, Storch et al., 2007, El Malhany et al. and McGuire et al., 2015). In general, the presence of co-occurring disorders negatively impacts quality of life in those with CTD more than increased tic severity alone (Eddy et al., 2011 and Zhu et al., 2006). Storch et al. found that ~70% of the problems experienced by those with chronic tic disorders were non-tic related, typically secondary to ADHD or OCD (Storch et al., 2007). The 2018 Impact Survey from the Tourette Association of America reported that 36% of adults and 42% of children felt the greatest challenge of living with TS was having to manage the co-occurring conditions and both groups shared the second greatest challenge was the limited medication and treatment options available (“Impact Survey”, 2018). See Figure 1 for overview of parent-reported challenges associated with TS in youth.



**Figure 1. Biggest Challenges in Managing TS in Children.** Figure taken from “Impact Survey” (2018).

## **Background of CBIT Therapy**

Comprehensive behavioral intervention for tics (CBIT) is a behavioral therapy specifically designed for the treatment of tics. The CBIT model was first published in 2010, and is now considered a first-line treatment for those with tic disorders (Piacentini et al., 2013 and Scahill et al., 2013). CBIT blends components of habit reversal therapy (HRT), including awareness training, competing response development and relapse prevention, with relaxation techniques and function-based intervention (where contextual factors that support or maintain tic expression are assessed and addressed). Of note, CBIT can be challenging for those ten and under as they have difficulty detecting the “premonitory urges” that are key to the awareness training component of CBIT (McGuire et al., 2014 and Woods et al., 2005). That said, more recent studies have called this supposition into question and have successfully enrolled children as young as nine into CBIT therapy trials with no significant difference in outcome for those under ten (Ricketts et al., 2018). While very effective in tic reduction, CBIT tends to be “tic-specific” (Woods et al., 2011, p.863). In a study comparing CBIT to psycho-education/supportive therapy, CBIT did not result in differential changes to co-occurring psychiatric symptoms and/or psychosocial functioning (Woods et al., 2011). A cognitive behavioral therapy (CBT)-based program, called “Living with Tics” (LWT) specifically targeted the psychosocial and functional impairment associated with CTD (Storch et al 2012) by providing coping skills and promoting resiliency. In a small randomized controlled trial (RCT), they found that the LWT treatment was acceptable, and associated with significantly improved quality of life, in addition to reduced Yale Global Tic

Severity Scale (YGTSS) tic-related impairment (McGuire et al., 2014, McGuire et al., 2015 and Storch et al., 2012). Regarding tic reduction, The LWT group did not differ from the waitlist group.

While CBIT is generally effective in reducing tics, there is some evidence that the presence of co-occurring ADHD negatively moderates the effect of the behavioral treatment in those with CTD, and overall (McGuire et al., 2014 and Halldorsdottir et al., 2015). Hypotheses for this finding include symptoms of ADHD impeding one's ability to engage in the therapy session, and/or the ADHD symptoms themselves (e.g. inattention, impulsivity) negatively impacting tic suppression (McGuire et al., 2014). That said, there has recently been more mixed data as a recent analysis concluded that "the presence of ADHD did not moderate treatment effect in participants under 18 years old" in either the CBIT or psychoeducation and supportive therapy groups (Sukhodolsky et al., 2017). Further research will be helpful in elucidating why ADHD symptoms appear to moderate change in some studies but not others.

Regardless, evidence shows that behavioral treatments can indeed be effective for improving ADHD symptoms (Antshel et al., 2014, Fabiano et al., 2009, Gould et al 2018., Safren et al., 2005, and Sprich et al., 2012, Sprich et al 2016), particularly when there are residual symptoms after pharmacological treatment. Approximately 5% of the school-age population is affected by ADHD which is characterized by a combination of hyperactive, impulsive, and inattentive symptoms that lead to impairment in at least two settings (e.g. school, family, social) (Kofler et al., 2019). Approximately 40-75% of those with ADHD experience emotional lability such as low frustration intolerance, impatience,

quickness to anger, and excitability. Additionally, executive dysfunction is common in the ADHD population, with 90% of those with ADHD experiencing some form of executive dysfunction such as disinhibition, difficulties with working memory, and reduced organization and planning ability (Kofler et al., 2019). Stimulant medication is the gold standard treatment in ADHD, and in school-aged youth, is recommended as the first line of treatment. There is also evidence that behavioral treatment can help improve ADHD-associated symptoms, particularly when used in conjunction with stimulant medication (“A 14-month randomized clinical trial of treatment strategies for ADHD”, 1999).

Regarding behavioral therapy treatment approaches, there are precedents for modifying and combining treatment protocols to better target specific populations. In 2015, Scribberas et al modified a CBT protocol for anxiety to account for co-occurring ADHD symptoms (Scriberras et al 2015), and Jarrett and Ollendick (2012) successfully combined treatment protocols to improve outcomes in youth with co-occurring ADHD and anxiety. Ricketts et al (2015) demonstrated that modifying the CBIT protocol to six twenty minutes sessions was feasible and acceptable and resulted in improved tic symptoms.

Given the high rates of comorbid psychiatric diagnoses in youth, there has been a recent push towards tailoring behavioral treatments more towards the individual patient and their particular symptom profile rather than a specific diagnosis. This has been achieved through the development of modular treatment protocols, where the clinician selects the module(s) most appropriate for the patient (McGuire et al., 2014; Thomas et

al., 2020; Wilhelm et al., 2011; Wood et al., 2014). Jeppesen et al. developed a transdiagnostic, modular CBT approach that demonstrated increased reduction in anxiety and depressive symptoms compared to typical CBT, and resulted in reduced impact of symptoms as reported by parents (Jeppesen et al., 2020). A modular approach to behavioral therapy has also demonstrated preliminary effectiveness in those with tic disorders and co-occurring oppositional defiant disorder (ODD). It is estimated that 25-65% of those with tic disorders have co-occurring ODD (Espil, 2020). Espil et al developed a process in treating co-occurring tics disorders and ODD that allowed therapists to match treatment modules to individual client's profiles, thus providing a customized approach to therapy (Espil, 2020).

Another novel approach to behavioral treatment delivery is the Unified Protocol for Transdiagnostic Treatment of Emotional Disorders. This protocol, which combined elements from the treatment of multiple disorders was shown to be at least as effective as traditional single-disorder protocols used to treat specific anxiety disorders (Barlow et al., 2017). Training clinicians to provide single protocol therapy that simultaneously targeted common co-occurring disorders was demonstrated to be at least as efficacious as single diagnosis protocols, and was noted to be more efficient and cost-effective. Additionally, the transdiagnostic approach demonstrated less attrition compared to the single diagnosis protocol group (Barlow et al., 2017).

Currently, there is no standardized behavioral treatment for CTD that accounts for ADHD symptoms and/or addresses the impact that ADHD symptoms have on tic-related behavioral treatment or quality of life. Given the high rates of comorbidities in tic

disorders, including ADHD, the ability to treat tic and non-tic related symptoms and impairments concurrently would be paramount.

### **Goals of the Present Study**

The purpose of this study was to address the gap in treatment availability, and develop a model within the effective CBIT-framework that directly addresses ADHD symptoms. We hypothesized that modifying the current CBIT protocol would be feasible and acceptable to participants, and would demonstrate efficacy in the treatment of tics, ADHD, and quality of life measures in those with CTD and co-occurring ADHD.

### **Specific Aims**

- 1) The primary aim of this study was to determine the treatment feasibility and acceptability of a modified CBIT (MCBIT) protocol for children and adolescents with CTD and ADHD.
- 2) The secondary aim was to evaluate preliminary changes in tic, ADHD and quality of life symptoms using the Yale Global Tic Severity Scale (YGTSS), NICHQ Vanderbilt Assessment Scales (VAS), Pediatric Quality of Life Inventory-Child Version (PedsQL) scale, and Clinical Global Impression (CGI) scales as rated by a blinded clinician.

## **METHODS**

### **Participants**

Participants included in the study were children and adolescents aged 10-17 years with diagnoses of CTD and co-occurring ADHD. Seventeen youths were enrolled and randomized to either the modified CBIT condition (MCBIT) or control group (CBIT). Inclusion criteria were: ages 10-17 years (inclusive), a primary diagnosis of TS or PMVT with a secondary diagnosis of ADHD using the DSM-5 criteria, a YGTSS of >13 (or >9 if PMVT), total impairment score of >19 on the YGTSS, English speaking, and ability to communicate meaningfully with the investigators and provide written assent.

Exclusion criteria were additional co-occurring psychiatric diagnoses including alcohol or substance abuse within the past three months, psychosis, current mania, organic mental disorder, developmental delay, IQ <80 on the Wechsler Abbreviated Scale of Intelligence (WASI), or any cognitive or communication impairment that would preclude the participant from effectively engaging in CBT. Additional exclusion criteria included active suicidality within the past six months, current severe illness warranting immediate psychopharmacological evaluation or intervention, any clinical features requiring a higher level of care than outpatient treatment as determined by the evaluator, other concurrent psychotherapy (decisions made on a case by case basis depending on what the additional therapy was targeting), and four or more previous CBT or CBIT treatment sessions within the last five years. Receiving psychotropic medication was not cause for exclusion, but participants were required to maintain a stable dose for four

weeks prior to the baseline assessment, and were required to maintain this dosage throughout the duration of the study.

Over the course of the study, certain inclusion and exclusion criteria were amended in order to increase recruitment and limit attrition including: broadening the age range from 11-17 to 10-17, allowing the follow-up assessments to be conducted via telephone, allowing participants to continue in the study if there was a gap in treatment greater than two weeks, requiring patients to be on a steady dose of medication for four weeks rather than six, allowing those with a diagnosis of autism spectrum disorder (ASD) to enroll so long as they were able to communicate effectively, and relaxing the criteria of having a current clinically active diagnosis of ADHD to having a documented formal history of ADHD with ongoing persistent symptoms (as per DSM-V criteria).

Demographics of the participants and their parents as well as psychiatric comorbidities and medication usage are presented in Table 1. Participants were randomized based on sex and presence/absence of prescribed psychotropic medication. Participants ranged in age from 10 to 17 years and the majority of participants were male (82.3%). Most of the participants identified as Caucasian (71%). A majority of participants' parents (88%) had an annual income that exceeded \$100,000. All participants were previously diagnosed with a co-occurring psychiatric disorder other than Tourette Syndrome and ADHD. The most common co-occurring disorders included obsessive-compulsive disorder (OCD; 59%), anxiety disorder (47%), and oppositional defiant disorder (ODD; 35%).

Over half of participants (53%) were currently taking a psychotropic medication with the most commonly taken being alpha agonists (29%), antidepressants (29%), and stimulants (24%).

**Table 1. Characteristics of Study Population**

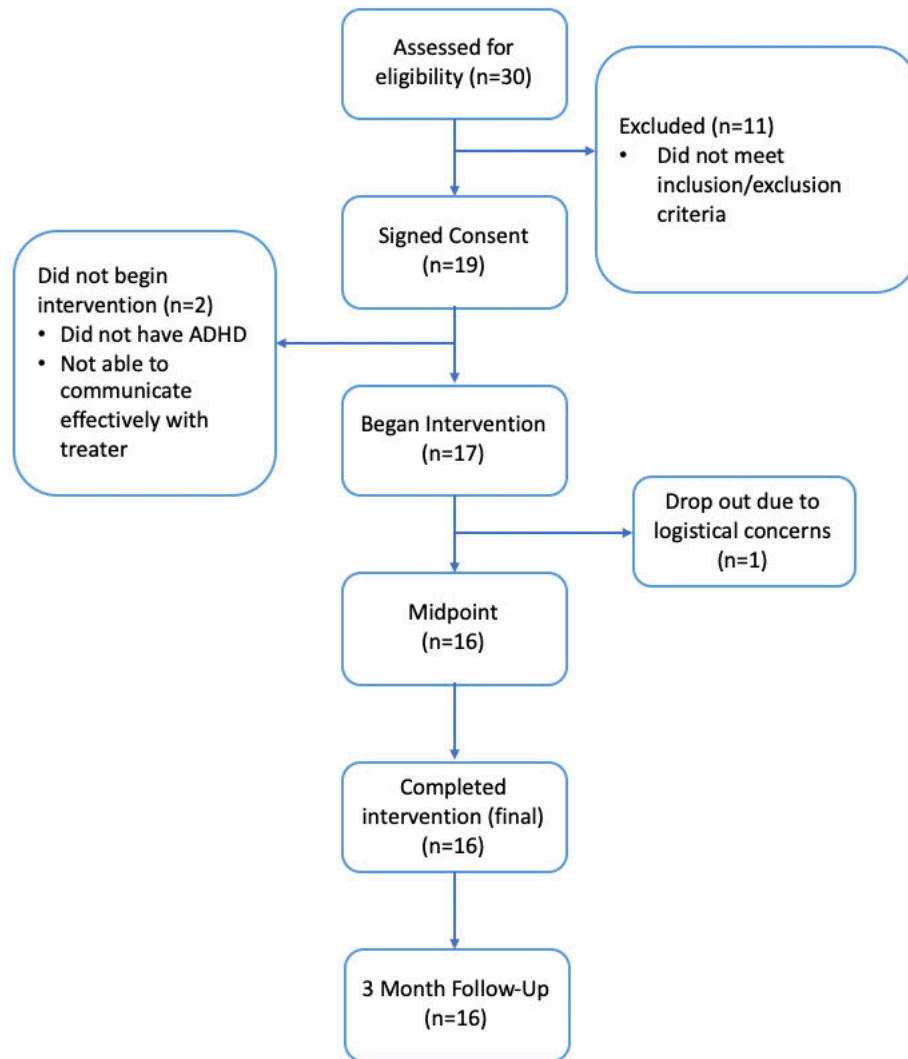
Variable	MCBIT (n=9)		CBIT (control) (n=8)		Total (n=17)	
	mean/N	(SD)/%	mean/n	(SD)/%	mean/N	(SD)/%
<b>Demographics (Child)</b>						
Age, mean (SD)	13.2	2.4	13.3	1.8		
Male Gender, N (%)	7	78%	7	88%	14	82%
Race, N (%)						
White	7	78%	5	63%	12	71%
Asian	0	0%	1	13%	1	6%
More than one race	1	11%	2	25%	3	18%
Other	1	11%	0	0%	1	6%
<b>Demographics (Parents)</b>						
Income, N (%)	1	11%	0	0%	1	6%
\$25,000-49,000	0	0%	1	13%	1	6%
\$50,000-74,999	5	56%	3	38%	8	47%
\$100,000-200,000	3	33%	4	50%	7	41%
>\$200,000						
<b>Psychiatric Comorbidities (Child)</b>						
DSM-V Axis I Diagnoses, N (%)						
ADHD, Combined	6	67%	4	50%	10	59%
ADHD, Inattentive Type	3	33%	2	25%	5	29%
ADHD, Hyperactive/Impulsivity Type	0	0%	2	25%	2	12%
ADHD, Unspecified	0	0%	0	0%	0	0%
Anxiety Disorder (generalized, social, separation, mixed)	3	33%	5	63%	8	47%
Major Depressive Disorder/Mood Disorder (inc. dysthymia)	2	22%	0	0%	2	12%
Obsessive-compulsive disorder	4	44%	6	75%	10	59%
Oppositional Defiant Disorder	2	22%	4	50%	6	35%
Any Axis 1 disorder (outside of TS/ADHD)	9	100%	8	100%	17	100%
<b>Current Psychotropic Medication Use (Child)</b>						
Any, N (%)	5	56%	4	50%	9	53%
Alpha agonist, N (%)	2	22%	3	38%	5	29%
Antidepressant, N (%)	2	22%	3	38%	5	29%
Antipsychotic, N (%)	0	0%	3	38%	3	18%
Mood stabilizer or anticonvulsant, N (%)	1	11%	2	25%	3	18%
Stimulant, N (%)	3	33%	1	13%	4	24%

## Procedure

Study procedures and all recruitment materials were approved and monitored by the Partners Institutional Review Board for human subjects research. Participants were

recruited from April 2016 to March 2019 and were informed about the study by clinicians in the OCD and Related Disorders Clinic, the Child Psychiatry Outpatient Clinic, the Neurology Tic Disorders Clinic, and the Pediatric Neuropsychiatry and Immunology Clinic at Massachusetts General Hospital (MGH). Participants were also informed about the study through local symposia on TS and related disorders and via fliers posted at MGH and local community restaurants, coffee shops, etc.

Eligibility criteria were assessed via a phone-based interview. Those that were deemed appropriate to enroll were asked to attend a pre-treatment assessment session in which the study procedures were explained and assent and informed consent were obtained (by a study representative or Dr. Greenberg). If Dr. Greenberg found the potential participant to be eligible following the baseline assessment, they were brought back in for their first treatment session where they were enrolled and immediately randomized to the MCBIT or control group. Subjects were not told which group they had been placed in; however, given the difference in materials presented in treatment sessions, some participants were likely able to determine their group. The treater was not blinded to which group subjects had been placed, but the independent evaluator who provided the assessments was blinded. Figure 2 describes the participant flow through the study.



**Figure 2: Patient CONSORT Diagram**

## **Assessment and Measures**

Assessments were conducted at baseline, the mid-point of treatment (after the 6<sup>th</sup> session), the end of treatment (EOT) (after the 12<sup>th</sup> session), and at the 3-month follow-up mark. Additionally, participants and the treatment therapist completed weekly scales (see Table 2). Baseline only measurements included: Mini International Neuropsychiatric Interview for children and Adolescents (MINI-KID), the Wechsler Abbreviated Scale of Intelligence (WASI) and the Client Expectancy Rating (CER). Ongoing measurements included the Yale global Tic Severity Scale (YGTSS), NICHQ Vanderbilt Assessment Scale (VAS), Clinical Global Impression-Improvement (CGI-Improvement), Clinical Global Impression-Severity (CGI-Severity), Pediatric Quality of Life Inventory-Child Version (PedsQL), Likert Scales/Open-Ended Questions (regarding the satisfaction/effectiveness of each session), and the Concomitant Medication and Therapy Questionnaire (Concom). The Client Satisfaction Questionnaire (CSQ – Satis) was given at the mid-point and end of treatment. Other scales collected included the Children's Depression Inventory (CDI), Caregiver Strain Questionnaire (CSQ), Child Tourette's Syndrome Impairment Scale (CTIM-P), Children's Yale-Brown Obsessive-Compulsive Scale (CY-BOCS), Emotion Regulation Questionnaire for Children and Adolescents (ERQ-CA), and the Parent Tic Questionnaire (PTQ). The baseline assessment scales were collected by study staff of at least a Bachelor's level education prior to subject randomization. Mid-point and post-treatment assessments were collected by Master's level or highly trained Bachelor's level study staff. See Appendix A for summary of individual assessment scales.

**Table 2. Assessment Measure Administration Schedule**

Measures	Rater	Self	Screen	Baseline	Weekly	Mid-Point	Final Session	3-month f/up
YGTSS	X		X	X		X	X	X
VAS - ADHD	X		X					
VAS - Abbrev		X		X		X	X	X
ADHD - SR		X		X		X	X	X
MINI-KID	X			X				
CTIM		X		X		X	X	X
PedsQL		X		X		X	X	X
PTQ		X		X		X	X	X
CGI-I	X	X			X	X	X	X
CGI-S	X			X	X	X	X	X
CSQ-8		X		X		X	X	X
CER		X		X		X		
WASI	X			X				
Likert		X			X	X	X	
CY-BOCS	X			X		X	X	X
CSQ-Satis.		X				X	X	X
ERQ-CA		X		X		X	X	X
CONCOM	X			X	X	X	X	X
CDI		X		X		X	X	X

**Primary Outcome Measures**

The primary aim of the study was to determine the treatment feasibility and acceptability of the developed MCBIT protocol. The primary measures used to assess this aim were the Client Satisfaction Questionnaire (CSQ-Satis) and the Client Expectancy Rating (CER). The CSQ-Satis is an eight question, four-point Likert self-report scale that measures satisfaction with treatment. The CER is also a four-point self-report questionnaire that assesses patient’s judgements about the credibility of the treatment

rationale, expectancy of change, and treatment acceptability. Additionally, patient retention and attrition were evaluated to assess treatment feasibility and acceptability.

### **Secondary Outcome Measures**

The secondary aim of this study was to pilot test the effectiveness of the MCBIT treatment protocol in youth with tic disorders and co-occurring ADHD. The secondary measures used to assess this aim were the Yale Global Tic Severity Scale (YGTSS), the NICHQ Vanderbilt Assessment Scale (VAS), the Pediatric Quality of Life Inventory-Child Version (PedsQL), and the CGI-I clinician-rated scale. The YGTSS is a clinician-rated semi-structured interview that measures tic symptom severity over the previous. The YGTSS has two separate components including a Total Tic Score (range: 0-50), and a Total Impairment Score (range: 0-50) with higher ratings corresponding to greater tic severity and impairment. The VAS is a parent rated scale that includes the DSM-IV symptom list for ADHD and other externalizing behaviors, including oppositional defiant disorder and conduct disorder (CD). VAS is measured on a 4-point scale which ranges from 0 (not at all) to 3 (very much). PedsQL is a twenty-three-item child-rated scale that measures a youth's quality of life. Each item is rated on a five-point scale, with higher scores indicating better quality of life. The CGI-Improvement is a seven-point Likert scale indicating any changes in overall mental health status and ranges from very much worse (0) to very much improved (6)

## **Treatment**

The MCBIT treatment was based on the original CBIT protocol (Piacentini et al., 2010), CBT for ADHD in adolescents protocol (Sprich et al., 2016), some elements of "Living with Tics" (Storch et al., 2012 and McGuire et al., 2014), and common cognitive behavioral therapy (CBT) techniques (e.g. thinking errors) broadly aimed at improving quality of life. Ten fifty-five-minute weekly sessions were conducted, followed by two relapse prevention sessions spaced two weeks apart. Individuals in both the CBIT and MCBIT group were provided with a tracking binder for CBIT-related hand-outs. Both groups completed weekly tic hierarchies, were assigned homework at the end of each session, and participated in a behavioral reward program. Both groups were taught the three core components of CBIT, including functional behavior assessment and intervention, habit reversal therapy, and relaxation training. In almost all cases, at least one parent participated in the sessions throughout the entirety of the treatment. Parents would step out of the room upon request from the participant for any particularly sensitive topics.

Participants in the MCBIT group received additional hand-outs including session outlines and summaries, visual aids during the sessions, and homework reminders. In addition, MCBIT subjects had access to fidget toys in the treatment room and were given one minute "brain breaks" between session activities. The treatment plan for both groups was divided into three phases: Evaluation/Psychoeducation, Basic Intervention, and Relapse Prevention for a total of twelve sessions. Both groups received a binder with

blank session-related handouts at the end of the treatment, and were given the opportunity to provide feedback about the treatment process.

Summary of MCBIT treatment sessions:

Evaluation/Psychoeducation (Session 1): The first session primarily focused on assessing the subject's tics and ADHD symptoms as well as the impact of tics and ADHD on their life. The therapist created a tic hierarchy with the patient to determine which tics were most bothersome. Psychoeducation about tics/Tourette Syndrome and ADHD, as well as CBIT and CBT models were provided to participants in an age-appropriate manner. The idea of function-based interventions, as well as planning and organization skills, were introduced

Basic Interventions (Session 2-10): These sessions focused on teaching CBIT principles, CBT for ADHD principles, and other general CBT techniques aimed at improving quality of life. Each session included review of homework from the previous session, review of current symptoms, and at least some time focusing on CBIT principles. Two sessions were dedicated solely to CBIT, and included additional elements of parent guidance (Ricketts et al., 2015). Three sessions focused on common ADHD symptoms (such as organization, planning, and distractibility) and treatment techniques. Four sessions focused on other common challenges typically affecting youth with co-occurring tic disorders and ADHD, such as emotion dysregulation, self-esteem and anxiety/thinking errors. Hand-outs describing topics covered during each session were provided to the participant/parent(s) at the conclusion of the session.

Relapse Prevention (Session 11-12): The final two sessions were held at two-week intervals beginning two weeks after the final intervention session, and focused on relapse prevention. During these sessions, the treater focused on transitioning the role of ‘therapist’ to the participant. Concerns about ending treatment and what to expect in the future were addressed. All skills learned over the course of the treatment were reviewed, and rated regarding degree of helpfulness. Strategies for continuing to practice the skills learned during the treatment were reviewed.

### **ASAP Sessions**

The protocol was amended to include the addition of extra Adjunctive Services and Attrition Prevention (ASAP) sessions to address crises and/or unusual needs or circumstances that arose. The goal of these sessions was to increase subject retention and each subject was allowed up to three ASAP sessions. Any subject that required more than the three allotted ASAP sessions was no longer considered eligible and terminated from the study. Two subjects utilized ASAP sessions.

### **Adverse Events**

Adverse events were monitored throughout the course of the study. The provider assessed for tic worsening, new health complaints, or behavioral changes during each treatment visit. Any replies indicating a possible adverse event prompted further inquiry and steps were taken to document and address it. Three subjects experienced adverse events. One subject reported tic and ADHD symptoms that were “very much worse” at

therapy session five, while one subject's parent reported a drop in mood and requested an ASAP session. Both subjects were monitored and assessed at their subsequent sessions and determined to not be at risk of harm and were able to continue in the study. The third subject reported feeling ill and rescheduled an assessment for a later time.

### **Statistical Analysis**

All statistical analyses were conducted using SASS software. In all tests conducted, values of  $P < .05$  were considered to be statistically significant. All outcome measures were modeled as repeated measures mixed models with treatment-group and time as predictors. Repeated measures were modeled using a Toeplitz covariance structure with heterogenous variances. Specific contrasts were used to test hypotheses about overall treatment effects from baseline to EOT, treatment differences over time by EOT, and treatment effects from baseline to EOT within each treatment group. All analyses were done according to the Intention-to-Treat (ITT) principle including all participants assessed at baseline ( $n=17$ ).

## **RESULTS**

### **Attrition**

Following randomization, the retention rate was strong, with sixteen of the seventeen participants completing the study. The participant who elected to discontinue did so secondary to logistical difficulties. All other participants were able to attend all treatment and follow-up sessions as scheduled. Of note, final assessments were changed from in-person to via phone for a handful of the participants (n=4) in order to ease burden of traveling to Boston for a non-treatment appointment.

### **Primary Outcome: Feasibility and Acceptability**

Participants in both the MCBIT and CBIT groups expressed high levels of satisfaction with the treatment protocol. There was no significant difference in the Client Satisfaction Questionnaire (CSQ-Satis) ratings between groups, and both groups trended towards improved satisfaction over time ( $p=0.18$ ) with the MCBIT group improving from 27.0/30 to at midpoint to 28.75/30 at end of treatment, and the CBIT group improving from 27.75/30 at midpoint to 29.29/30 at end of treatment. The majority of participants expected to improve, expressed satisfaction with the treatment, and were optimistic about their outcomes. There was no significant difference in the Client Expectancy Rating (CER) score between groups, and both groups' expectation of improvement (on a scale of 0-100% improvement) increased from baseline to midpoint of treatment.

## Secondary Outcomes: Treatment Effectiveness

Summary statistics including means (M), standard deviations (SD) and within-group effect sizes for all secondary measures for both groups are reported in Table 3.

**Table 3. Summary Statistics of Secondary Measures**

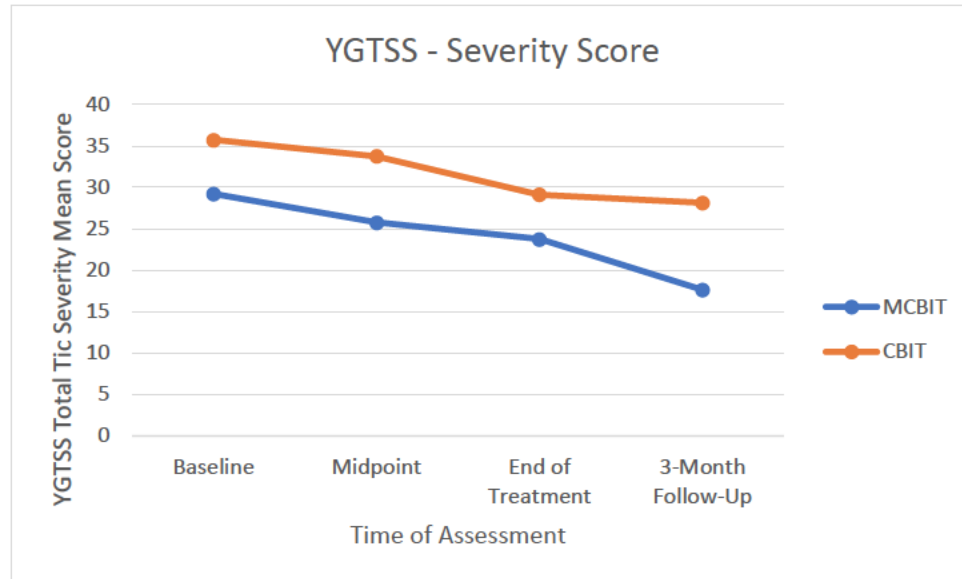
Variable	Group	Baseline		End of Treatment		Follow-up		Effect Size Baseline to Follow-Up
		M	SD	M	SD	M	SD	
YGTSS Total Severity	MCB T	29.22	10.24	23.75	10.17	17.63	8.9	-1.13
	CB T	35.75	8.17	29.13	5.84	28.14	4.74	-0.93
YGTSS Total impairment	MCB T	28.89	7.82	13.75	11.88	12.5	12.82	-2.1
	CB T	31.25	9.91	22.5	10.35	21.43	10.69	-0.99
Vanderbilt (ADHD total score)	MCB T	28.67	8.73	22.63	7.33	20.43	7.83	-0.94
	CB T	24.5	8.93	17.38	9.77	20	10.02	-0.5
PEDQL (Child-Rated)	MCB T	73.78	5.62	73.23	10.29	82.1	11.85	1.48
	CB T	68.48	8.13	66.08	12.42	69.67	11.19	0.15

### YGTSS Total Tic Severity

YGTSS tic severity scores demonstrated statistically significant improvement by end of treatment (EOT) when looking at the CBIT (control) and MCBIT groups in combination (pre: M=32.3, SD=9.64; post: M=26.4, SD=8.48;  $p=0.005$ ,  $d_{ws}=-0.61$ ).

There was significant improvement in tic severity in the control group ( $p=0.016$ ,  $d_{ws}=-0.81$ ) and a trend towards significant improvement in the MCBIT group ( $p=0.085$ ,  $d_{ws}=-0.53$ ), though the treatments did not significantly differ from one another over time ( $p=0.572$ ,  $d_{IGPP}=.28$ ). These changes in tic reduction were maintained and strengthened over time by follow-up in both groups, particularly in the MCBIT group (within-group effect sizes: control  $d_{ws}=-0.93$ , MCBIT  $d_{ws}=-1.13$ ), though the group differences were still not statistically significant at follow-up ( $p=0.476$ ,  $d_{IGPP}=-0.20$ ). Figure 3 shows the

change in YGTSS severity as measured at baseline, midpoint, EOT, and three-month follow-up.

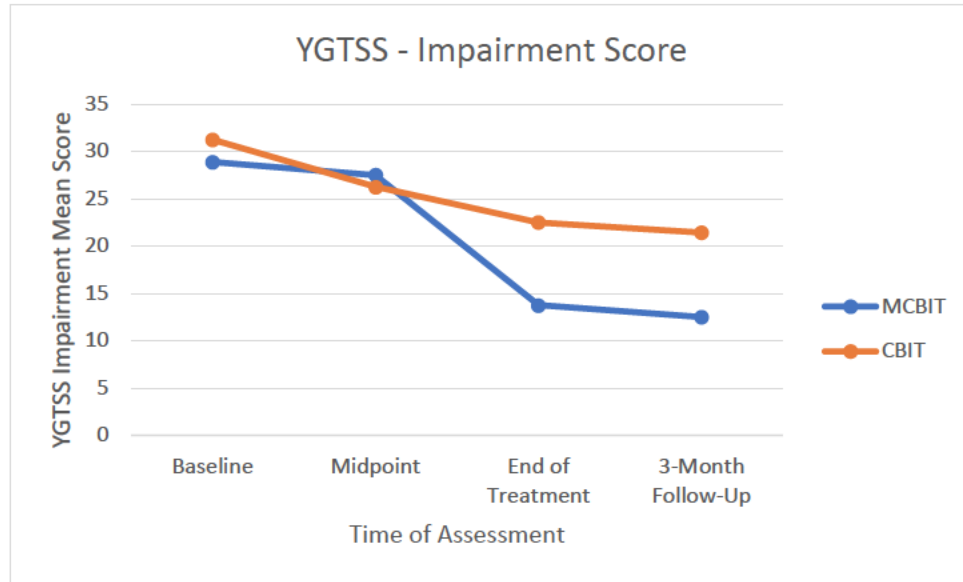


**Figure 3: YGTSS-S Ratings Over Time**

### YGTSS Impairment

An overall improvement in tic-related impairment as measured by the YGTSS impairment scale was demonstrated when looking at the CBIT and MCBIT groups in combination ( $p < 0.0001$ ,  $d_{ws} = -1.37$ ) by EOT. There was a large improvement in tic impairment severity in the control group ( $p = 0.017$ ,  $d_{ws} = -0.88$ ) and a greater improvement in the MCBIT group ( $p < 0.0001$ ,  $d_{ws} = -1.94$ ), though the treatments did not significantly differ from one another over time through EOT ( $p = 0.148$ ,  $d_{IGPP} = -1.05$ ). At follow-up, the groups continued to show large effects overall ( $p < 0.0001$ ,  $d_{ws} = -1.54$ ), though were not statistically different from one another ( $p = 0.086$ ,  $d_{IGPP} = -1.11$ ). The within group

effect-sizes from baseline to follow-up were  $d_{ws}=-0.99$  For CBIT and  $d_{ws}=-2.10$  for MCBIT. Figure 4 shows the decrease in YGTSS Impairment as measured at baseline, midpoint, EOT, and three-month follow-up.

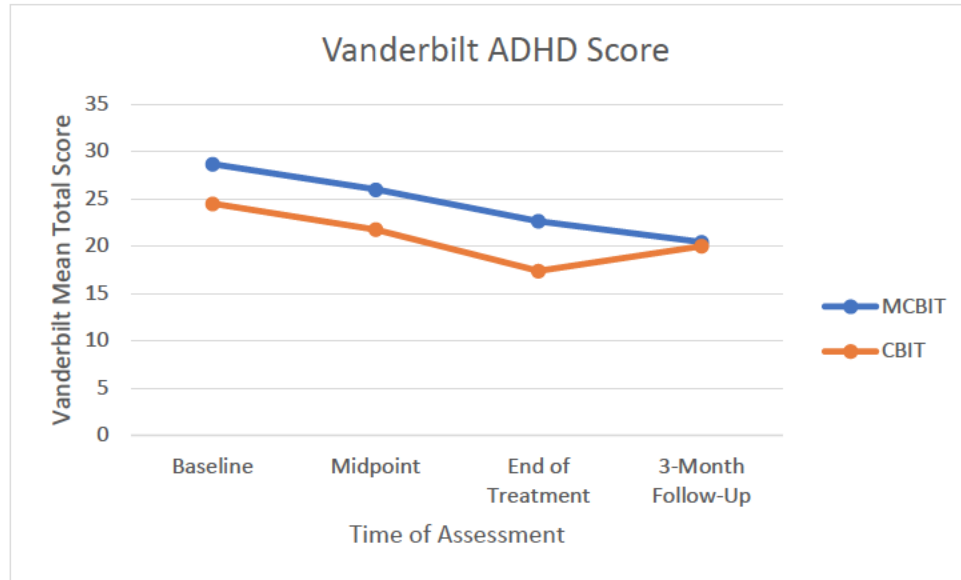


**Figure 4: YGTSS-I Ratings Over Time**

### Vanderbilt Assessment Scale

Similar to the YGTSS measures, there was an overall improvement in ADHD symptom severity in the CBIT and MCBIT groups in combination (pre:  $M=26.7$ ,  $SD=8.8$ ; post:  $M=20.0$ ,  $SD=8.8$ ;  $p=0.002$ ,  $d_{ws}=-0.76$ ) through EOT. There was a significant improvement in ADHD symptom severity in both the control group ( $p=0.017$ ,  $d_{ws}=-0.80$ ) and the MCBIT group ( $p=0.034$ ,  $d_{ws}=-0.69$ ) through EOT, though the treatments did not significantly differ from one another ( $p=0.807$ ,  $d_{IGPP}=0.11$ ). These changes were maintained throughout follow-up in both groups (within-group effect sizes: control  $d_{ws}=-$

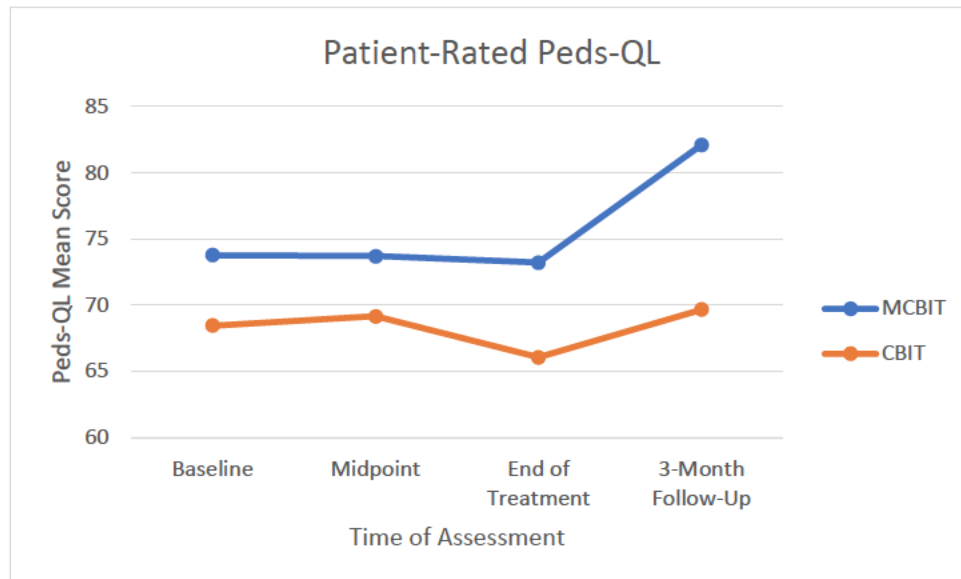
0.50, MCBIT  $d_{ws}=-0.94$ ), and continued to not significantly differ from one another ( $p=0.447$ ,  $d_{IGPP}= -0.44$ ). Figure 5 shows the decrease in VAS as measured at baseline, midpoint, EOT, and three-month follow-up.



**Figure 5: VAS Ratings Over Time**

### **PedsQL-Child Rated**

There was no significant change in child-rated quality of life measures for either the CBIT ( $p=0.536$ ;  $d_{ws}=-0.30$ ) or MCBIT group ( $p=0.633$ ;  $d_{ws}=-0.10$ ), including through EOT and follow-up (CBIT:  $p=0.969$ ,  $d_{ws}=0.15$ ; mCBIT:  $p=0.183$ ,  $d_{ws}=1.48$ ). Of note, there appeared to be a large, albeit not statistically significant improvement at follow-up in the MCBIT group. Figure 6 shows the overall trend in PedsQL as measured at baseline, midpoint, and EOT, as well as the sharp increase at three-month follow-up.



**Figure 6: PedsQL Ratings Over Time**

### **CGI-I Clinician-Rated**

Clinician rated CGI-I was evaluated for quality of life (QoL) measures as well as for overall improvement. From baseline to EOT, 63% of the MCBIT group demonstrated “very much improvement” or “much improvement” for both overall and QoL measures. From EOT to three-month follow-up, 71% of the MCBIT group reported “very much improvement” or “much improvement” for both measures. For the CBIT group 88% of participants reported “very much improvement” or “much improvement” in QoL while 75% of participants reported improvement overall from baseline to EOT. From EOT to three-month follow-up 71% of the CBIT group reported improvement for both QoL and overall measures. One child reported worse severity, and was thus evaluated for continuation in the treatment, and was determined to be acceptable to continue.

## DISCUSSION

In this study, we evaluated the acceptability and feasibility of a modified CBIT treatment that accounts for co-occurring ADHD symptoms and worse quality of life in youth with CTD and co-occurring ADHD, as well as present new preliminary data on the efficacy of this modified CBIT treatment.

The results of this study indicate that MCBIT was both feasible and acceptable. There was an unusually high retention rate (94%), and the attrition rate of 6% was lower than other typically reported rates in small scale ( $n < 50$ ) behavioral therapy studies, such as 12.5 % (Jarrett and Ollendick, 2012), 36% (Ricketts et al., 2015), and 22% (Sprich et al., 2016). The strong retention rate indicates that MCBIT treatment was acceptable to both participants and parents alike, and is a feasible treatment model for youths with CTD and co-occurring ADHD. The CSQ-Satis and CER mean scores improved from baseline to follow-up for both the MCBIT and CBIT groups, with no significant difference between groups. This demonstrates that the MCBIT treatment was as equally as well tolerated as traditional CBIT, and that both treatment groups had good expectations around treatment success. We believe that in the future, incorporating a remote component could improve feasibility and acceptability even further as reasons for drop-out, and increased time between sessions were all secondary to logistical difficulties.

Of note, inclusion criteria were expanded during the study to increase enrollment and limit attrition. In particular, the inclusion criteria were amended to allow for participants with a formal clinical history of ADHD but current subclinical symptoms to

enroll. This is because it is common for individuals who have been treated with stimulants to have ongoing residual ADHD symptoms that require intervention to limit associated functional impairment. Allowing individuals with subclinical cases of ADHD to participate in the study more realistically represents the general population of those with CTD and co-occurring ADHD, and allows the results to be more generalizable.

Regarding adverse events, these were broadly thought to be due to factors outside of the treatment protocol, including needing to reschedule due to illness, and temporary worsening in mood and ADHD symptom severity that did not preclude the subjects from continuing in the study. Additionally, given that the participant that reported worse symptoms and had difficulty with the treatment was in primary school, it may be helpful to limit participants to those at least in middle school given the content of the ADHD material and strategies.

The present study offers preliminary evidence that MCBIT therapy improves tic severity, reduces tic-associated impairment, improves ADHD severity, and may improve quality of life. At EOT, there was statistically significant improvement in total tic severity for the MCBIT and CBIT groups taken in combination. Although the groups did not significantly differ from one another, these improvements were maintained and seemed to strengthen past EOT to three-month follow-up, particularly for the MCBIT group. These results indicate that MCBIT is likely at least as effective as CBIT in tic symptom improvement, which is important given that less time is spent focusing on tics directly. The results also point towards perhaps longer lasting improvement following treatment.

There was an overall improvement in tic disorder-related impairment in both groups. The MCBIT group trended towards greater improvement from baseline to EOT, though again the groups did not significantly differ from one another. This reduction in impairment held steady between EOT and three-month follow-up for both groups, indicating that treatment effects for MCBIT persist past EOT.

There was overall improvement in ADHD symptom severity in the CBIT group in addition to in the MCBIT group. Similar to both YGTSS measures, the improvement in ADHD severity was seen for the CBIT and MCBIT groups in combination, and the groups did not statistically differ from one another. These changes were maintained throughout follow-up in both groups; again, indicating that treatment effects persist past EOT. It is interesting to note that the CBIT group demonstrated improvement in ADHD symptoms despite not learning specific skills tailored towards addressing them. One possibility is that the decrease in tic severity enabled participants to experience less tic-related distraction and thereby maintain better attention. As there is a trend towards ongoing ADHD improvement following EOT in the MCBIT, a hypothesis is that they continue to benefit from the specific ADHD treatment skills in addition over time. A larger follow-up study would be needed to determine any significant difference in ADHD symptom outcomes for MCBIT versus traditional CBIT therapy, and could also help clarify the degree to which ADHD is indeed a moderating factor in tic treatment response.

Despite there being no significant change in child-rated quality of life measures (PedsQL) for either the CBIT or MCBIT groups from baseline to EOT, there was a sharp

increase between EOT and follow-up in both groups, with a greater trend in the MCBIT group. This may indicate that quality of life improvements take additional time to develop, and may be better captured during a longer follow-up period.

Taken together, these results suggest that a modified model of CBIT offers an acceptable and feasible, and based on preliminary results, helpful alternative treatment to CBIT in youth with co-occurring CTD and ADHD. By incorporating specific modules that focus on ADHD symptoms such as limiting distractibility, improving organizational skills, and managing overwhelming tasks, a modified CBIT approach enables participants to address ADHD symptoms and tics concurrently so that both improve in tandem, and the improvement can be sustained (and potentially even increase) over time.

Given the prevalence of co-occurring psychiatric conditions in those with CTD, it has become increasingly important to address the unique presentations of each child and family with personalized, adaptable treatment protocols. Recently, there has been a shift towards utilizing modular cognitive-behavioral therapy models, where depending on the specific needs and profile of the patient, individual treatment sessions can be swapped in or out. We utilized that approach in our MCBIT Session 10 where we only covered mood dysregulation/distress tolerance if there were specific concerns. If there were concerns about mood dysregulation or that are able to address multiple co-occurring psychiatric conditions and are tailored to the specific needs of the patients such as ASD and anxiety (Wood et al., 2014), body dysmorphic disorder (Wilhelm et al., 2011), and avoidant/restrictive food intake disorder (Thomas et al., 2020). Effective interventions exist for CTD and ADHD, but there is no established treatment to address both

concurrently. Modular approaches to behavioral therapy may offer greater flexibility and provide a method to individualize therapy to promote better treatment outcomes (Espil, 2020).

### **Strengths and Limitations**

A particular strength of the current study is that patients with a large range of co-occurring psychiatric diagnoses were included and that those participating were concurrently taking a variety of psychotropic medication. Given this, we believe the results of this study may be generalized to additional clinical settings where TS and PMVT are treated.

The greatest limitation of the current study was the sample size. Given the small number of participants, the power of the study was somewhat limited. A larger sample would be needed in order to detect statistically significant differences in the outcomes of the MCBIT group in comparison to the CBIT group. An additional issue that arose was the logistical demand that weekly fifty-five-minute sessions placed on some participants. Although the retention rate was strong, there was a single participant lost to follow-up after the parent expressed difficulty with driving into Boston to attend weekly meetings. Of note, the participant lost to follow-up was the sole African-American participant as well as the only subject whose parents' annual income fell in the lowest income bracket of \$25,000 - \$49,000 which may restrict generalizability. Allowing for virtual visits in addition to the telephone encounters offered may help to alleviate this logistical burden and increase access to treatment for lower-income patients.

Additional research is warranted to further investigate the efficacy of a MCBIT model of therapy for youths with tic disorders and co-occurring ADHD. Future research should aim to compare the efficacy of the MCBIT versus CBIT approach as well as the long-term maintenance of treatment gains made.

This study presents new data on the feasibility and acceptability, as well as preliminary data on the efficacy of a modified treatment model of CBIT therapy. Per our knowledge, this is the first study to modify the standardized CBIT to target youth with tics and co-occurring ADHD and impaired quality of life.

### **Conclusions**

The results of this study demonstrate preliminary evidence that a modified model of CBIT therapy is both feasible and acceptable for youth with co-occurring CTD and ADHD. In addition, the results provide preliminary support that MCBIT therapy improves tic severity for patients with co-occurring ADHD, and that these improvements are maintained at three-month follow-up.

## APPENDIX A

**Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989):** The YGTSS is a clinician-rated scale that measures changes in tic symptom severity over time. The YGTSS provides a Tic Score, as well as an Impairment Score ranging between 0-50, with higher scores corresponding to greater tic severity and impairment.

**NICHQ Vanderbilt Assessment Scale (VAS; NICHQ, American Academy of Pediatrics, McNeil, 2002):** The VAS is a parent (and teacher) rated scale that includes the DSM-IV symptom lists for ADHD and other externalizing behaviors, including oppositional defiant disorder (ODD) and conduct disorder (CD). VAS is rated on a four-point scale between 0 (not at all) to 3 (very much).

**Clinical Global Impression-Improvement (CGI-Improvement; Guy, 1976):** The CGI-Improvement is a seven-point Likert scale between 0-6 corresponding to very much worse to very much improved, respectively. CGI may be rated by a clinician or patient.

**Clinical Global Impression-Severity (CGI-Severity; Guy, 1976):** Akin to CGI-I, the CGI-Severity is also seven-point Likert rating indicating illness severity. Values range from 0-6 indicating no illness to extremely severe illness as rated by a clinician or patient.

**The Caregiver Strain Questionnaire (CSQ; Brannan et al., 1997):** The Caregiver Strain Questionnaire measures how significantly a child's chronic condition has negatively affected his or her family. Parents rate items on a zero to five scale. The questionnaire includes ten items that measure objective strain, and eleven items that assess subjective strain.

**Child Tourette's Syndrome Impairment Scale (CTIM-P; Storch et al., 2007):** The CTIM-P is parent-rated scale that includes thirty seven items related to school, home, and social activities that tics may negatively impact.

**Children's Depression Inventory (CDI): The CDI (Kovacs, 1992):** is a brief self-report (27-item) questionnaire used to assess depressive symptoms in children and adolescents age 7-17 years old. It has been found to have high reliability and concurrent validity.

**Children's Yale-Brown Obsessive-Compulsive Scale (CY-BOCS; Scahill et al., 1997):** The CY-BOCS is used to measure obsessive-compulsive symptom severity over time with total severity scores ranging between 0 and 40.

**Client Expectancy Rating (CER; Borkovec and Nau, 1972):** The Client Expectancy Rating assesses patients' judgments about the credibility and acceptability of the

treatment, and their expectation of improvement as measured on is a four-item self-report scale.

**Emotion Regulation Questionnaire for Children and Adolescents (ERQ-CA; Gullone and Taffe, 2012):** The ERQ-CA is a 10-item, valid, age-appropriate, internally consistent, self-administered measurement which measures the use of two specific emotion regulation strategies in children/adolescents aged 10-18.

**Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999):** The WASI is a brief, measure of intelligence in multiple settings. It is nationally standardized, linked to the Wechsler Intelligence Scale for Children – Fourth Edition (WISC-IV), and in its two-subtest form (about 15min) yields full scale IQ scores.

**Parent Tic Questionnaire (PTQ; Chang et al., 2009):** The Parent Tic Questionnaire is rated on a 1-4 severity scale and lists twenty-eight motor and vocal tics that are marked as present or absent over the last week. These are summed together to generate a total score between 0 and 112.

**Pediatric Quality of Life Inventory-Child Version (PedsQL; Varni et al., 2003):** The PedsQL contains twenty-three items that assesses a child's quality of life. Items are rated on a five-point scale, with higher scores indicating improved quality of life.

**Client Satisfaction Questionnaire (CSQ – Satis):** The Client Satisfaction Questionnaire is an 8 question, 4-point Likert self-report scale that measures satisfaction with treatment.

**Concomitant Medication and Therapy Questionnaire (Concom):** The Concomitant Medication and Therapy Questionnaire tracks whether there are any concurrent therapy or medication changes over the course of the treatment. If yes, it is followed up by a Medication and/or Therapy log depending on the change.

**Likert Scales/Multiple Choice Questions:** After each session, we will provide subjects with brief Likert scales/multiple choice questions, which they can complete at the end of each session assessing the satisfaction/effectiveness of each session.

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## CURRICULUM VITAE

