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IMPROVING SYMPTOM TRAJECTORIES OF TIC DISORDERS AND CO-OCCURRING
DIAGNOSES: THE ROLE OF INTEGRATIVE INTENSIVE INTERVENTION

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To: Dean Michael Heithaus
College of Arts, Sciences, and Education

This dissertation, written by Chelsea F. Dale, and entitled Improving Symptom Trajectories of Tic Disorders and Co-Occurring Diagnoses: The Role of Integrative Intensive Intervention, having been approved with respect to style and intellectual content, is referred to you for judgment.

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DEDICATION

To my wonderful and loving family, friends, and mentors for their guidance, support, and advice.

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ABSTRACT OF THE DISSERTATION
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Tic Disorders (TDs) are neurodevelopmental disorders characterized by involuntary movements or sounds, which can often impact an individual's quality of life and functioning. Youth with TDs frequently also meet criteria for an additional co-occurring diagnosis (i.e., attention-deficit/hyperactivity disorder (ADHD), anxiety, obsessive-compulsive disorder). Comprehensive Behavioral Intervention for Tics (CBIT) is a well-established and first-line behavioral intervention for youth with TDs, however, despite recent investigation into alternative treatment formats, co-occurring symptoms have not yet been explicitly addressed within the context of CBIT. Intensive intervention formats are well established for co-occurring diagnoses of TDs; however, the literature on the utility, feasibility, and acceptability of intensive interventions for TDs and co-occurring diagnoses is limited. The portfolio of graduate research presented in this dissertation consists of three separate studies of increasing rigor evaluating Taming Tics Together (TTT), which was developed by Chelsea Dale, M.S., and Justin Parent, Ph.D. TTT is a novel, 5-day, videoconference-based intensive intervention to concurrently address TD and co-occurring diagnosis symptoms. The first study (Study 1) uses a case study design, which evaluates the TTT program in an individual format with an 11-year-old biracial

male diagnosed with Tourette's Disorder and ADHD. The second study (Study 2) builds upon family feedback and lessons learned in Study 1 and uses a group-based case series design with three male youth ($M_{\text{age}} = 9.67$, Range = 9 - 11) and their caregivers. The third study (Study 3) builds upon Study 1 and 2 using a randomized controlled trial design with 21 youth ($M_{\text{age}} = 12.55$, 50% male) and a caregiver (95% female) who were randomly assigned to an immediate treatment group vs. a 1-month waitlist. Primary outcomes across all three studies were TD-related, and secondary outcomes were related to co-occurring diagnostic symptom trajectories. Study 3 also incorporated exploratory outcomes related to caregiver-specific, youth quality of life, and interference outcomes. Satisfaction with treatment and attendance was high across all studies. Overall, results across all studies were promising, suggesting feasibility, acceptability, and preliminary support for TTT to simultaneously address TDs and co-occurring diagnoses. Strengths and limitations of each of the three studies, the clinical implications, and future directions are discussed.

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ABBREVIATIONS AND ACRONYMS

ADHD	Attention-Deficit/Hyperactivity Disorder
BPT	Behavioral Parent Training
CBIT	Comprehensive Behavioral Intervention for Tics
CBT	Cognitive Behavioral Therapy
CFW	Child and Family Well-being Clinic
GAD	Generalized Anxiety Disorder
I-TTT	Immediate Taming Tics Together Treatment Group
MDD	Major Depressive Disorder
OCD	Obsessive-Compulsive Disorder
ODD	Oppositional Defiant Disorder
SUDs	Subjective Units of Discomfort/Distress
TAA	Tourette Association of America
TD(s)	Tic Disorder(s)
TTT	Taming Tics Together
WL	Waitlist Group
YGTSS-TSS	Yale Global Tic Severity Scale – Total Tic Severity Score
YGTSS-I	Yale Global Tic Severity Scale – Impairment Score

I. INTRODUCTION TO PORTFOLIO

Youth with tic disorders (TDs) can experience significant impairment across domains of functioning. Youth diagnosed with a TD often also meet criteria for other diagnoses, such as attention-deficit/hyperactivity disorder (ADHD), disruptive behavior disorders, obsessive-compulsive disorder (OCD), as well as mood and anxiety disorders. Comprehensive Behavioral Intervention for Tics (CBIT) is a well-established intervention for youth with TDs; however, although alternative CBIT treatment modalities have begun to be investigated (i.e., self-guided, group-based, videoconference-based), co-occurring diagnosis symptoms are not explicitly addressed in within the standard CBIT treatment protocol. Additionally, numerous barriers (i.e., geographic barriers, cost, the time associated with travel and/or spent in the clinic weekly, etc.) to accessing a treatment provider who specializes in CBIT continue to persist, despite tremendous efforts put forth by the Tourette Association of America and other advocacy groups. Intensive outpatient programs (IOPs) are well-established for co-occurring diagnoses of TDs and provide a necessary alternative to weekly treatment sessions, however, the literature on integrated IOPs for TDs and co-occurring diagnoses is limited. Therefore, there is a significant need for alternative or non-traditional treatment modalities to increase access to evidence-based interventions that can simultaneously address both TDs and co-occurring diagnoses. Further, the Centers for Disease Control reported an increase in emergency room visits due to tics during the COVID-19 pandemic, which likely puts an additional strain on treatment providers and families seeking interventions for tics.

In response to the need for new treatment modalities and likely new increased need for specialized treatment providers as a result of the COVID-19 pandemic, Taming Tics Together (TTT) a novel, 5-day, videoconference-based intensive intervention to concurrently address TD and co-occurring diagnosis symptoms and incorporates both group- and individually-based intervention sessions, was developed by the doctoral candidate, Chelsea Dale, M.S., and Justin Parent, Ph.D. As such, the portfolio of graduate research presented in this dissertation consists of three studies that build upon the prior study with increasing rigor, and incorporate both family feedback and lessons learned from each previous study. The first study (Study 1) will present a case study using the initial TTT protocol with an 11-year-old biracial male presenting with Tourette's Disorder and ADHD and his caregivers. Study 1 is currently published online in the journal *Cognitive and Behavioral Practice*. Next, the second study (Study 2), builds upon caregiver feedback and lessons learned within Study 1 using a group-based case series design with three male youth ($M_{age} = 9.67$, Range = 9 - 11) and their caregivers. Study 2 incorporates both the individually-based and group-based session formats, allowing for broad transdiagnostic as well as person-centered skill acquisition for each child and their family. This study is currently under review for publication. Finally, the third study (Study 3) increases in evaluative rigor by assessing the TTT protocol with a randomized controlled trial design. Study 3 involves 21 youth (20 analyzed, $M_{childage} = 12.54$ years; 50% male; 40.0% Hispanic/Latinx) and their caregivers, who were randomly assigned to either the Immediate TTT group (I-TTT, n=14) or a 1-month Waitlist (WL, n=7). All three studies will discuss strengths and limitations, clinical implications, and future directions. Overall and taken together, this portfolio of studies provides preliminary

support for the feasibility, acceptability, and effectiveness of TTT as an additional treatment modality to address TDs and co-occurring diagnoses concurrently, as well as the possibility of aiding in increasing access to evidence-based care. The three studies are presented within this portfolio of graduate research in sequential order with increasing rigor in evaluative methodology.

II. STUDY 1: CASE STUDY

INTRODUCTION

Tic disorders (TDs) are neurodevelopmental disorders characterized by sudden and involuntary motor (i.e., arm jerking, shoulder shrugging) and phonic (i.e., throat clearing, animal sounds) tics. TDs broadly encompass a number of current DSM-5 diagnoses, such as Tourette's disorder, persistent or chronic motor or vocal tic disorder, and provisional tic disorder (American Psychiatric Association [APA], 2013). Importantly, TDs have a significant impact on youths' overall daily functioning. Specifically, they can have deleterious effects on youth's social, familial, and academic functioning (Kurlan et al., 2001; Roessner et al., 2011; Specht et al., 2011) and are demonstrated to impair their quality of life (Storch et al., 2007). Further, parents of children with TDs are likely to experience increased stress and caregiver burden (Cooper et al., 2003). As a result, TDs can impact the entire family's overall wellbeing.

A majority of children with a TD also meet criteria for at least one co-occurring disorder. In particular, over 60% of children with a TD also meet criteria for attention-deficit/hyperactivity disorder (ADHD) (Center for Disease Control and Prevention, 2009), a chronic neurodevelopmental disorder characterized by inappropriate levels of inattention, hyperactivity, and impulsivity (APA, 2013). Research suggests that co-occurring TDs and ADHD in childhood often lead to greater impairment than TDs alone (i.e., Lebowitz et al., 2012), including pronounced difficulties across familial and social domains (Sukhodolsky et al., 2003).

Numerous randomized controlled trials have suggested that Comprehensive Behavioral Intervention for Tics (CBIT) is effective in reducing tic symptom severity,

intensity, frequency, and duration among children (e.g., Piacentini et al., 2010; Himle et al., 2012). McGuire and colleagues' (2014) meta-analysis found that individuals with a TD who engaged in a behavioral intervention, such as CBIT, were more likely to benefit from treatment (i.e., reduction in tic symptoms), as compared to control interventions, and experienced comparable treatment effects to those found with alpha-adrenergic agonist (a-agonists) medication use. As such, the American Academy of Neurology (AAN; Pringsheim et al., 2019) recommends CBIT as the first line of treatment for individuals experiencing TDs, given its minimal risk (i.e., side effects) and equivalent efficacy when compared to alternative treatments (i.e., a-agonists, antipsychotics) (Pringsheim et al., 2019). These recommendations also include support for services provided via teleconference or video conference, should in-person services be unavailable or inaccessible.

Traditionally, CBIT was designed to be an eight to ten-week, one-hour, individual and in-person treatment, during which functional assessments, relaxation, habit reversal training, and relapse prevention are taught to and practiced with children and parents, with the goal of reducing children's tic symptom frequency, severity, duration, and intensity (Piacentini et al., 2010). However, despite substantial support for CBIT for improving TD symptoms, highly comorbid disorders, such as ADHD, are not explicitly addressed in the standard treatment protocol (Coffey, 2015). As a result of this important limitation, an alternative intervention modality that provides treatment strategies to address both TDs and equally impairing comorbidities is warranted.

An important next step in treatment development for TDs is to evaluate alternative treatment modalities that modify CBIT to be capable of treating co-occurring

ADHD. Indeed, non-traditional delivery formats, such as intensive interventions, as opposed to standard weekly sessions, are well-established for the treatment of related and comorbid disorders and have been shown to be an efficacious and cost-efficient modality for delivering multimodal interventions (Ehrenreich & Santucci, 2009; Fabiano, et al., 2014;). Intensive treatment delivery formats provide the opportunity for families to gain access to treatment for a limited period of time without the extended burden often associated with weekly treatment sessions. Recently, there has been emerging evidence to support intensive, group-based treatment, particularly for diagnoses that are less common or are known to require specialized interventions that are not widely accessible. For example, Cornacchio and colleagues (2019) found that an intensive, group-based format for children with selective mutism was associated with minimal perceived barriers from caregivers, high treatment satisfaction, and clinically significant reductions in symptoms eight weeks post-treatment. Additionally, Graziano and colleagues (2020) evaluated an intensive model for behavioral parent training (BPT), an evidence-based treatment for ADHD and disruptive behavior disorders (for reviews, see Evans et al., 2018; Fabiano et al., 2009; Pelham & Fabiano, 2008). Similarly, results from this study found that BPT delivered in an intensive format was equally efficacious compared to standard weekly BPT and that parents who experience high levels of stress were more likely to benefit from the intensive formats than standard BPT. Overall, intensive intervention modalities are effective across a number of childhood mental health illnesses, and this treatment delivery method has begun to be extended to CBIT to address TDs.

While initial support for an intensive form of CBIT has begun to emerge, the literature is limited. Flanbaum and colleagues (2011) adapted standard CBIT into a two-

week intensive format for a 25-year-old male exhibiting motor and vocal tics. Findings suggest reductions in the participant's subjective units of distress (SUDs), however, no post-assessment Yale Global Tic Severity Scale (YGTSS; Leckman, 1989), the gold-standard TD assessment, was conducted to assess changes in overall tic frequency, severity, intensity, duration, and impairment post-intervention. The authors also reported that the patient did not participate in all agreed-upon booster sessions following the end of treatment and, upon meeting with the patient 5-months following the end of treatment, the patient noted difficulty implementing CBIT strategies. Further, the authors note the importance of booster sessions in aiding the generalizability of skills into an at-home setting. Blount and colleagues (2018) conducted a four-day intensive program (N=5), which resulted in improvements following the end of treatment for 4 out of 5 primarily adult participants. To our knowledge, only three studies to date have evaluated intensive CBIT in an exclusively pediatric population. Blount and colleagues (2014) conducted a preliminary case series utilizing a four-day intensive approach, predominately focused on preadolescents (N=2), providing support for the utility of intensive CBIT for reducing tic symptoms rapidly. Kennedy and colleagues (2016) provided eight sessions over the course of two consecutive weeks to 10 children ($M_{age} = 13.7$) and found significant treatment-induced improvements in YGTSS-assessed total tic severity. In addition, Heijerman-Holtgreffe and colleagues (2020) evaluated the use of a group-based intensive exposure with response prevention (ERP) protocol for 14 children ($M_{age} = 11.69$ years) and found a 16% reduction in total tic scores. While improvements were noted, the reduction reported in this study is significantly lower when compared to studies utilizing CBIT. Taken together, preliminary evidence for the utilization of intensive CBIT for

children and adults is promising. However, none of the interventions described above included components targeting ADHD explicitly, despite the high incidence among individuals with TD.

One of the most well-studied psychosocial interventions for ADHD is the delivery of BPT to the parents of children with ADHD. Grounded in social learning theory principles (e.g., Patterson, 1982; Patterson & Fisher, 2002), BPT seeks to modify parenting behaviors that may inadvertently maintain disruptive behaviors. BPT programs largely share a core set of common principles, including positive parenting (e.g., praise, positive attending), effective commands and consequences, as well as the development of an incentive system to shape target behaviors through positive reinforcement. Taken together, these core set of principles have been shown to be successful in reducing maladaptive parenting practices and improving child externalizing behaviors (Chronis et al., 2004; Kaminski et al., 2008; Pelham & Fabiano, 2008). Given the chronic nature of ADHD and its associated impairments across domains of functioning, the active engagement of caregivers of children with ADHD is considered essential in the management of the disorder (Sibley et al., 2012). In doing so, caregivers can serve as agents of change for ADHD-related impairment.

Over 60% of children with a TD also meet criteria for ADHD and treatments for TDs, such as CBIT, and ADHD, such as BPT, have been shown to be effective. An integral next step is to evaluate a combined approach of CBIT and BPT provided in an intensive modality, as intensive CBIT may be utilized as a mechanism to target proximal (e.g., tics, disruptive behaviors) as well as distal (e.g., parental stress) outcomes. As such,

we used a case study design approach to evaluate the utility of an intensive and combined intervention for the treatment of a TD and co-occurring ADHD.

CASE PRESENTATION: JACOB

The following case study functions as a preliminary test of the acceptability, feasibility, and effectiveness of the treatment described below. Jacob's parents sought services outside of their geographic region, as there are no trained CBIT providers locally. Further, the family was restricted to a two-week window for the intervention. As such, the intervention described below was deemed to adequately suit the family's needs and time restrictions. A total of six intensive CBIT sessions (spanning across two weeks) were provided at a university-based outpatient clinic specializing in the treatment of TDs and ADHD.

Relevant Background and Context

At the time of intake, Jacob was an 11-year, 9-month-old biracial male who traveled with his biological parents to receive treatment for both motor and vocal tics. His parents reported that Jacob's first motor and vocal tics appeared when he was 8 and 9 years old, respectively. Presenting problems at the time of intake included frequent vocal tics, namely, cursing/swearing (coprolalia). Furthermore, Jacob's parents reported that his vocal tics had become more impairing and intense due to an accompanying increase in voice-level volume coupled with higher rates and more complex coprolalia (e.g., a string of cursing/swearing, as opposed to a single curse/swear word). Furthermore, his parents reported that both vocal and motor tics were a source of significant impairment across home and school settings, as well as with peers. Jacob was enrolled in a standard school setting, however, given the increased volume and frequency of his vocal tic, which

caused disruptions in the classroom, he often left school prior to standard dismissal time. Additionally, Jacob's parents were concerned about his self-esteem as he reported feelings of embarrassment and anger due to vocal tics within the school setting and in front of peers. As a result of the numerous domains of impairment at school, his parents reported switching his educational setting to home. Upon intake, Jacob was receiving a home school education. At home, in addition to vocal tics, Jacob presented with both simple and complex motor tics, varying in the degree of impairment.

Jacob also presented to treatment with a prior diagnosis of ADHD, combined presentation, which was given at the age of five. Before intake, Jacob had been prescribed Clonidine extended-release for his ADHD-related symptoms and TD. Despite subsequent adjustments in medication dosage over the course of a year, Jacob's parents reported that they chose to discontinue the medication due to a perceived lack of improvement in both ADHD and TD-related symptoms. At the time of Jacob's clinical intake, he was not taking medication.

Treatment Design and Assessment

Treatment Development. To address ADHD and TD symptoms simultaneously, the protocol by Woods and colleagues (2008), as well as standard BPT core constructs were adapted to meet the unique needs of families presenting with TDs and co-occurring diagnoses. First, overlapping strategies across both protocols were evaluated and selected based on their utility and appropriateness for children. For instance, the use of a reinforcement system through which specific behaviors and utilization of competing responses (CRs) can be rewarded was considered clinically useful for the simultaneous treatment of TDs and management of ADHD-related behaviors. Further, additional

strategies that incorporate differential attention, such as planned ignoring for reducing inappropriate behaviors, and positive reinforcement, such as labeled praise for increasing positive and/or desirable behaviors (e.g., practicing CRs for tics), were deemed most useful. Next, key elements from diagnosis-specific interventions, such as awareness training for detection of premonitory urges and strategies to evaluate and understand tics more thoroughly, and the use of effective behavioral management strategies (e.g., using short, clear, and direct commands), to address ADHD-related impairment were added to the treatment plan. Finally, the set of core skills selected were strategically adapted into an abbreviated format.

Assessment and Diagnosis. See Figure 1 for an overview of assessment and intervention sessions. Tic assessments were conducted at three time points: (1) psychoeducational evaluation, completed 8-months prior to treatment by providers not affiliated with the current study, (2) pre-treatment clinical intake assessment, completed by the first and third authors, and (3) a post-assessment, conducted four months following the end of treatment, conducted by the second author. The post-assessment was conducted at this time to align with the booster session, as well as evaluate long-term treatment outcomes. An immediate post-treatment assessment was not conducted given the family's time restraint.

Assessment one: Psychoeducational evaluation. Approximately eight months prior to the initial clinical assessment, Jacob received a full psychoeducational evaluation. The results of the psychoeducational evaluation included clinical diagnoses of Tourette Disorder and confirmation of ADHD, combined presentation. The Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) Global Tic Severity Scale Score from

the psychoeducational evaluation was 66/100, indicating significant elevations in tic severity and tic-related impairment.

Assessment two: Pre-treatment. Jacob's parents provided consent before intake procedures occurred, which included consent to use data collected throughout treatment for research purposes. A thorough assessment of symptoms before treatment included a semi-structured clinical interview with Jacob's mother and father and a separate interview with Jacob, to evaluate tic-related and ADHD symptoms. The Disruptive Behavior Rating Scale (DBD-RS; Pelham et al., 1992) was administered to Jacob's parents to assess for ADHD symptoms, whereby Jacob's parents endorsed 7/9 inattention (i.e., *is often distracted by extraneous stimuli*) and 6/9 hyperactive/impulsive (i.e., *often leaves seat in classroom or other situations in which remaining seated is expected*) symptoms. During the evaluation, Jacob required significant redirection and repetition of instructions and was observed fidgeting and squirming in his chair which was unrelated to his tics. Jacob and his parents completed the YGTSS, obtaining a Global Tic Severity Scale Score 66/100 for patient-and parent-reported assessments. Clinicians met to discuss parent and child severity ratings and used a multi-informant approach (i.e., evaluated parent and child reports) to determine a global severity score. Overall, parent and child reports were within ± 1 point on total tic severity scores; therefore, no significant differences were noted in either informants' YGTSS scores.

Assessment three: Post-treatment assessment. The YGTSS was re-administered four months following the end of treatment, at the same time as booster session two, in order to reduce the burden of additional assessment appointments. Further, the assessor

collected anecdotal evidence from Jacob's parents regarding his ADHD symptoms and their impairment (see *Treatment Outcomes* for the results of this assessment).

TREATMENT

Intensive Course of Treatment

A complete outline of session content, duration, frequency, and format (i.e., the entire family together or concurrent sessions) is provided in Table 1. Sessions were conducted across three unique phases. Phase one consisted of skill-building, phase two of practicing and reinforcing, and phase three of review and telehealth-based booster sessions. Within each phase, parent- and child-specific strategies, as well as combined strategies were employed. All sessions were conducted with at least one of two primary clinicians. At times, sessions were conducted concurrently, with one clinician involved in each session; however, each session concluded with both Jacob's parents and Jacob together to ensure that all parties involved were aware of and understood the content of each session.

Phase 1: Skill Building. Treatment began with the clinicians separately providing Jacob and his parents psychoeducation about tics (i.e., symptoms, genetics/heritability, information about the progression of tic disorders, treatment options, an overview of CBIT, and resources). Given that Jacob's parents received psychoeducation for ADHD prior to enrollment in CBIT, a brief review was conducted with Jacob's parents (i.e., how behavioral interventions can help manage behavioral problems and reduce impairment associated with ADHD). Jacob also created a hierarchical list of current tics, as well as learned awareness training and relaxation strategies (deep breathing and progressive muscle relaxation). Procedures for determining and utilizing CRs were also introduced to

Jacob. Parents were taught basic awareness training and CR development procedures and how to create and implement a reward system. In order to address ADHD symptoms, parents were provided with skills to utilize differential attention (e.g., ignoring inappropriate and praising appropriate behavior) and effective instruction (e.g., direct commands). Behavioral management strategies intended to address more serious behaviors such as non-compliance and aggression (e.g., time-out) were not taught, as Jacob readily followed directions with limited prompting and did not exhibit aggressive behavior toward others. With guidance from the clinicians, Jacob and his parents collaboratively developed a reward system in which Jacob actively participated by selecting prizes that he deemed rewarding and were subsequently used to reinforce appropriate behavior and skill use throughout the course of treatment. This final step was importantly established prior to phase 2 (practicing and reinforcing) in order to ensure that Jacob was familiar with the procedures and was adequately reinforced by the selected prizes. During phase 2, sessions lasted 1.5 hours and were conducted simultaneously but separately with Jacob and a clinician, as well as Jacob's parents and a separate clinician.

Phase 2: Practicing and Reinforcing. During this phase, the clinician met with Jacob alone and practiced awareness training and created a CR for two current and impairing tics (one motor and one vocal tic). A CR was deemed appropriate if it met the following criteria: (1) the CR behavior was physically incompatible with the tic; (2) the CR could be maintained for at least 1 minute or until the urge to tic subsided, whichever came first; and, (3) the CR was less noticeable than the tic itself. For Jacob's motor tic, a CR was created to impede the first tic behavior from occurring. For the vocal tic, Jacob

was instructed to engage in controlled breathing (e.g., slowly breathing in and out) through the nose while keeping his lips closed, as this CR impeded Jacob from engaging in the vocal tic. Once the CRs were created, sessions consisted of repeated practice using the CRs, as well as implementing and modifying in-session and at-home behavioral charts and reward systems. Specifically, a behavioral chart for the two-week intensive intervention was created to increase the frequency and duration of out-of-session practice. At the end of treatment, an at-home behavioral chart was created to help address behaviors that occur in a more naturalistic setting. As practice with CRs continued, necessary modifications were made as needed in order to ensure that the most effective CR was utilized. During this phase, session 4 took place 2 days following session 3 to allow for extended practice time. In order to meet Jacob and his family's unique needs, additional time was spent on awareness training, which utilized immediate rewards in conjunction with practice. For example, a minimum of 45 minutes in sessions 1 and 2 was spent on awareness training and CR practice. Each time Jacob was engaged in awareness training (i.e., identifying an urge, or utilizing a CR), he earned points towards a larger reward. Further, sessions 2-6 also utilized this strategy to help Jacob engage in awareness training and CR practice. Outside of session practice (homework) was emphasized throughout treatment and Jacob was successful in completing a large majority of practice between sessions with some support from his mother. Awareness and/or CR practice was suggested between each session for a minimum of one hour with parent and child together. Once the behavioral chart was created, Jacob's parents implemented this chart on a daily basis as part of their out-of-session homework. Finally, practice using relaxation strategies (i.e., deep breathing, progressive muscle relaxation)

was incorporated into out-of-session practice once these strategies were introduced in session. Throughout out-of-session practice, Jacob's parents were encouraged to implement parent-based strategies (i.e., praise for practice, ignoring negative behaviors) as needed.

To address Jacob's ADHD-related impairment, CBIT sessions were modified. For example, CBIT sessions are traditionally held in 1-hour weekly sessions where both the provider and patient sit in chairs and only stand up when it is required of them (e.g., to practice a CR). In contrast, Jacob and his family met with clinicians for a total of 6 days, for approximately 1 to 1.5 hours each day (see Table 1). As previously noted, Jacob experienced high levels of hyperactivity, making it particularly difficult for him to sit for more than a few minutes at a time. In response to his hyperactivity impairment, session formats allowed for movement in an assigned area, while simultaneously practicing identifying urges and utilizing CRs. This modification was coupled with positive reinforcement, frequent breaks, and fun activities which were contingent upon Jacob's participation in the therapeutic session component. In addition, Jacob was easily distracted by extraneous stimuli. To address his distractibility and overall engagement, an intensive token-reward system was used to increase target behaviors, namely recognizing the urge to tic and the use of CRs. Points were awarded immediately after Jacob exhibited either behavior as prior studies have shown that immediate feedback is necessary to obtain clinically meaningful improvement for children with ADHD (Abramowitz et al., 1992; Fabiano & Pelham, 2003). At the end of each day, Jacob was able to exchange the points earned in session for a tangible reward. Of note, ADHD-related impairment was addressed in session to facilitate the delivery and practice of the CBIT treatment

components. The goal of using immediate reinforcement (e.g., awarding points) was to help increase Jacob's awareness of premonitory urges and/or tics, as well as reinforce the use of CRs.

Concurrently, Jacob's parents met with a clinician to discuss BPT strategies to address Jacob's ADHD symptoms. Specifically, over the course of six sessions, Jacob's parents were introduced to the following skills/strategies: (1) differential attention (praise and ignoring), (2) effective instructions, and (3) rewards and behavioral charts. During session reviews, at which time both Jacob's parents and Jacob were present in the same room, Jacob's parents were able to briefly practice utilizing the skills taught throughout sessions and receive live coaching during sessions (e.g., ignoring negative behaviors). While it is possible that ADHD symptoms (e.g., hyperactivity, distractibility) were reduced during the week-long intensive program, ADHD is a chronic disorder for which continued behavioral treatment is necessary to produce sustained behavioral improvements. As such, the BPT that was concurrently provided to Jacob's parents was intended to address ADHD-related impairment during and post-treatment, whereas in-session strategies used with Jacob were employed primarily to sufficiently manage his behavior for the purpose of providing CBIT treatment. Further, BPT skills review and brief live coaching were incorporated as a core component of booster sessions to ensure Jacob's parents were able to implement strategies effectively once they returned home.

Phase 3: Review and Telehealth-based Booster Sessions. The final in-person sessions consisted of relapse prevention, with particular emphasis placed on creating CRs for new tics that may appear in the future. Further, CRs for both tics and relaxation strategies were practiced. Lastly, the clinicians worked collaboratively with Jacob's

mother to establish a behavioral chart and reward system to ensure continued practice at home, such that Jacob would earn incentives when using CRs and relaxation techniques as well as when meeting behavioral expectations (e.g., staying on task during school assignments). Rewards were tiered into small, medium, and large rewards based on size, cost, and Jacob's indication of the degree of desirability, and included additional screen time (i.e., extra television, tablet, or videogame usage), time with friends, (i.e., playdates or sleepovers), toys and small desserts. Given the family's time restrictions on the last day, a post-treatment assessment was not conducted and, instead, the final session focused entirely on ensuring the family was familiar and comfortable with skills learned throughout the two weeks.

Telehealth-based booster sessions took place approximately three, four, and six months following the end of treatment and were individualized to meet the family's needs at each time point. The telehealth format was utilized given geographic barriers of the family, however, the use of telehealth as a treatment delivery platform has been shown to be efficacious (i.e., Comer et al., 2017). The first telehealth-based booster session (three months post-treatment) consisted of a review of progress since their last session and addressing any questions or difficulties they were experiencing with using the strategies learned throughout treatment. Specifically, Jacob's parents requested a review of each CR, as well as the steps to identifying premonitory urges and creating CRs. At the second telehealth-based (four months post-treatment) booster session skills associated with both disorders were discussed. Specifically, BPT skills (e.g., positive reinforcement, differential attention) were reviewed to ensure continued use of these strategies for ADHD-related symptoms. In addition, previous CRs were reviewed and modified if

necessary. Parents were receptive to the intervention strategies and were able to aid their child in the utilization of the skills at home. At this time, the YGTSS was conducted to evaluate long-term tic symptom severity. At the third telehealth-based booster session (six months post-treatment), a review of CRs, as well as a review of household rules, differential attention, and overall school progress. At this time, Jacob's parents reported an improvement in Jacob's academic performance and indicated no concerns about school.

TREATMENT OUTCOMES

The YGTSS global total score 4-months following the end of treatment was 40/100, indicating a 26-point decrease from the psychoeducational evaluation and pre-treatment assessments (see Figure 2). Additionally, Jacob's total tic severity score decreased from 26 to 20, and his impairment score decreased by 50% (from 40 to 20) from pre-treatment assessments to post-treatment assessment. While Jacob was prescribed Clonidine extended-release to address TD and ADHD symptoms between the psychoeducational evaluation and his initial behavioral intervention intake, he was not taking any active medications during the behavioral intervention at the center of this case study. However, pre-treatment YGTSS scores indicated no change from the psychoeducational evaluation to the initial intake assessment, despite the use of medications for over one year prior to the initial intake assessment, but was not prescribed or being taken at the time of intake. This would suggest that the pharmacological treatment had no impact on parent-reported scores on the YGTSS prior to the start of CBIT treatment and was consistent with parent report on the reason for its discontinuation. In contrast, there was approximately a 39% reduction in the total tic

severity score from pre- to post-treatment assessments following the combined CBIT + BPT intensive intervention, suggesting that improvements in YGTSS scores were a reflection of the psychosocial treatment provided. Jacob's six-point raw score reduction on the total tic severity scale indicates a significant treatment response (Storch et al., 2011). Importantly, score reductions noted in the current case study are consistent with findings reported on the initial CBIT randomized controlled trial, which indicated a 7.6-point decrease on the total tic severity scale, and a 51% decrease on the impairment scale (Piacentini et al., 2010). Further, these findings support the combination of CBIT and BPT – two well-established psychosocial interventions – as non-pharmacological alternatives for the treatment of co-occurring TD and ADHD (Barbarese et al., 2020). Programs designed to address tics or ADHD should consider utilizing a combined treatment approach that assesses for and provides individuals with the skills to reduce symptoms of either disorder.

During the booster sessions, Jacob's parents indicated significant improvements in tic severity, duration, and frequency. During booster session one, Jacob's parents indicated brief instances of "flare ups" occurred; however, these periods lasted approximately one day (as compared to pre-treatment, when such periods of time would have lasted up to three weeks). While his parents reported that their use of strategies had decreased following the end of treatment, they felt confident in their ability to reinstate any needed skills should tic symptoms reemerge significantly. During the final booster session, parents reported improvements in Jacob's behavior, particularly with his ability to remain on task during at-home school assignments with additional support from parents, as well as his use of CRs

when needed. Jacob was also receptive to attending booster sessions and was able to collaboratively create and modify CRs with minimal support from the clinician.

Additionally, during the final booster session, the clinician received feedback from Jacob's parents regarding the format, structure, and content of the two-week intensive intervention they had received. Jacob's parents reported overwhelming support for the simultaneous CBIT and BPT approach, indicating that learning skills related specifically to TDs and ADHD within the same treatment format, and with the same treatment providers, was invaluable. His parents also reported that the use of CBIT strategies were helpful and necessary core skills; however, the additional parenting skills, such as differential attention and praise were vital, as they provided strategies to promote positive interactions with Jacob, as well as improve his behavior. Overall, Jacob and his parents attended all sessions as indicated, and reported clinically meaningful improvements in TD and ADHD-symptom severity.

DISCUSSION

The current case study provides preliminary support for combining CBIT and BPT and suggests that simultaneous implementation of strategies may be useful in treating TDs and co-occurring diagnoses, such as ADHD. Jacob exhibited significant improvements in tic symptomatology, as well as reductions in parent-reported impairment associated with ADHD. The findings in the current study are consistent with reductions in tic symptom severity following standard CBIT strategies (Dutta & Cavanna, 2013). Earlier case studies suggest that children with disruptive behaviors may not be suitable for intensive CBIT (Blount et al., 2014), however, these studies were limited to the standard CBIT protocol, which does not include strategies to manage such behaviors.

By incorporating behavioral management strategies, such as a behavioral chart to target the most impairing behaviors and the utilization of reinforcement strategies, for example, a point system – through which Jacob earned points immediately after exhibiting appropriate behaviors – we were able to successfully address co-occurring ADHD symptoms and ultimately increase Jacob’s active participation throughout sessions.

The evidence from the current study is promising and begins to fill an important gap in the TD treatment literature. Jacob’s parent’s qualitative feedback suggested overall acceptability of the intervention, as well as areas of improvement for treatment to further engage future families. Specifically, his parents indicated that more time in session throughout the day, such as morning and afternoon sessions within the same day, would be beneficial to ensure proficiency with using each skill. Thus, this extended time within the same day may provide clinicians with the opportunity to incorporate additional strategies to address comorbid diagnoses, such as Cognitive Behavioral Therapy skills, or expand upon BPT skills learned within treatment, and also aid families in identifying associated triggers and/or stressors that may be limited to certain hours of the day. It was noted by Jacob’s parents that a group-based format would have also been useful for children and parents to provide and gain social support and share their unique experiences with TDs and common comorbidities.

Given that access to trained providers in both CBIT and BPT is limited, the current study’s format provides the opportunity for families to access treatment providers in an abbreviated manner, while also allowing for longer-term care through booster sessions conducted via telehealth (as needed). This study was conducted over the course of two weeks, however, the total number of intervention sessions, including the initial

evaluation, was conducted over the course of seven days. Therefore, this intervention is likely amenable to further alterations to the treatment schedule to allow for accommodations associated with each family's differing schedules, such as occurring over one week of school vacation or the summer. While it is possible that there may be reductions in between-session CR practice, intensive formats could be more conducive to maximizing in-session practice as compared to a weekly format and may ensure that additional practice using CRs is accomplished during treatment. Overall, an intensive, combined intervention approach may be an acceptable alternative to traditional intervention modalities, as it may reduce several barriers to treatment.

Telehealth-based booster sessions also offer additional time spent practicing and modifying strategies that may be integral to maintaining gains seen throughout treatment and generalize these gains to the patient's naturalistic home environment. Booster sessions allow for individualized review and practice of skills that may reinforce their use after intensive treatment termination. Importantly, Flancbaum and colleagues (2011) noted that, while improvements were initially seen post-treatment, due, in part, to lack of participation in booster sessions, treatment gains were not maintained once the patient returned home. Therefore, these booster sessions likely provide additional and necessary support to families in order to ensure that skills learned throughout treatment are accurately and effectively implemented at home. Future studies should evaluate the utility of internet-based booster sessions and their effectiveness in maintaining treatment gains.

Preliminary evidence from the current case study suggests that combined CBIT and BPT can be a useful treatment modality for children experiencing TDs and co-occurring ADHD. The current treatment design provides an alternative to attending

sessions with separate providers who specialize in TDs or ADHD and may facilitate increased access to care for families without providers within their community. Families and children experiencing significant distress and/or impairment from tics and who exhibit symptoms of co-occurring diagnoses are likely good candidates for the current treatment and may benefit from an intensive format in which presenting diagnoses can be targeted simultaneously.

This study is not without limitations. First, the results are based on a single patient and, therefore, the generalizability of the current findings is not known at this time. Second, specific patient and parent treatment satisfaction measures were not utilized; instead, this information was collected via anecdotal evidence from parents. Future studies would benefit from utilizing standardized treatment satisfaction questionnaires to evaluate the family's overall perspectives of the treatment protocol. However, to avoid clinician bias, post-treatment assessments of tic symptoms were collected by a third-party clinician who was not involved in Jacob's treatment. Third, while immediate post-treatment assessments were not able to be collected, evaluations conducted four months following the end of treatment indicated long-term improvements, which may reflect a more important metric of the longevity of improvements and a possible underestimate of immediate post-treatment reductions in TD symptoms. Nonetheless, future studies should evaluate immediate post-treatment tic symptoms in addition to long-term changes. Fourth, future studies should evaluate the effectiveness and acceptability of combining CBIT and BPT in a larger sample to determine the generalizability of the current study's findings. Fifth, it is important to note that intensive outpatient treatment is sometimes not fully covered by all insurance types (i.e., private vs. government, i.e., Cook et al., 2014;

James et al., 2014), therefore, intensive formats may not be amenable to all families. Policy and insurance companies should consider increasing coverage for intensive programs as a means to more efficiently treat comorbid disorders. Sixth, the current case presentation utilized two clinicians to provide treatment (i.e., one to meet with the child and one to meet with the parents) in order to provide treatment simultaneously and reduce the amount of time the family was required to spend in the clinic. The clinicians who participated in the treatment had knowledge and experience in both CBIT and BPT, therefore, the family did not attend separate sessions in order to receive specialized treatment from either provider. However, the use of two clinicians may not be feasible across all treatment providers and settings. Nonetheless, this treatment can easily be modified to be provided by one clinician to include parent-only sessions and child-only sessions at different time points throughout the day or may be modified to be conducted as one joint parent-child session. Seventh, post-treatment assessment of ADHD outcomes was limited to anecdotal parent feedback. As such, future studies would benefit from conducting standard ADHD post-treatment assessments to elucidate treatment-induced changes in ADHD symptoms. Finally, the utilization of an experimental design with an active treatment and control group would aid in elucidating clinically significant improvements from those associated with the natural passage of time (e.g., the child's symptoms may have improved, given the waxing and waning nature of tics).

Notwithstanding its limitations, the current study provides preliminary evidence for an abbreviated and combined intervention to address tic- and ADHD-related symptoms concurrently. Importantly, an intensive intervention format allows for families to exclusively focus on the acquisition and use of strategies without as many

interferences from non-treatment-related responsibilities. Additionally, a group-based version of this format would allow for treatment providers to offer services to a larger number of patients, particularly given the shortage of trained CBIT therapists nationally, while simultaneously reducing the geographic barrier often associated with finding a provider. Future studies are warranted to determine the efficacy of this treatment format, as well as the possibility of utilizing patient feedback provided in the current case study to evaluate a group-based combined intervention. Specifically, given that individual parent and child, as well as combined sessions, were conducted to address TD and ADHD symptoms simultaneously, group-based intensive interventions should consider adopting this multimodal approach to provide general TD management strategies within a group setting, as well as individual patient-specific sessions to address their unique impairment due to co-occurring diagnoses strategies. This format likely alleviates many of the limitations indicated in the current case study (i.e., group-based social support, increased number of sessions per day, reduction in the total number of days spent in treatment) to improve overall outcomes for children experiencing TDs and ADHD and may also be more cost-effective for treatment providers.

TDs are prevalent throughout the United States and can lead to significant impairment in children's school and social functioning, as well as impact their family's overall well-being. Youth with TDs are also likely to experience a comorbidity, with ADHD as the most prevalent co-occurring diagnosis. Despite significant effort to train providers in CBIT nationally, access to providers still remains geographically limited. Currently, CBIT and BPT are both first-line non-pharmacological interventions to address TDs and ADHD and may drastically reduce impairment associated with these

diagnoses. Therefore, the findings from the current study provide preliminary evidence for a cost- and time-efficient treatment alternative for families seeking behavioral interventions for TDs and ADHD that can simultaneously address common co-occurring diagnoses symptomatology.

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Figure 1. Diagram of Events

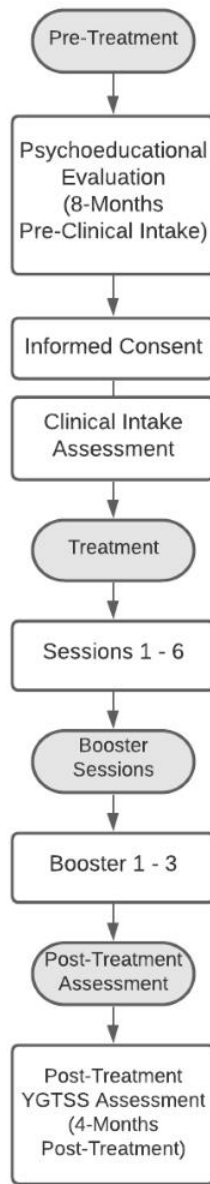


Figure 2. YGTSS Global Tic Severity Scale Score by Timepoint

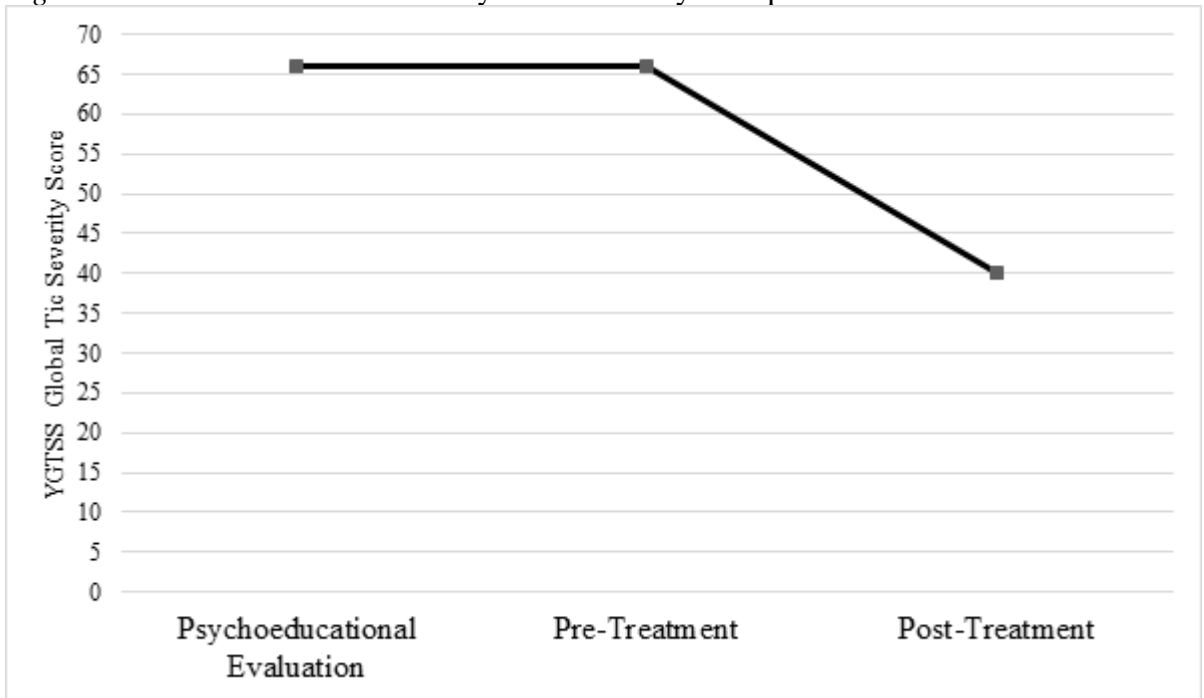


Table 1. Session Outline and Description of Session Content

Phase	Session Number	Combined Session Content	Individual Session Content – Parent	Individual Session Content – Child
1	1 ^a	Psychoeducation, awareness training, introduction to reinforcement	Parent orientation to CBIT	Identification of target tics, awareness training practice, relaxation
1	2 ^a	Brief check-in	Praise and rewards	Awareness training practice, development of competing response for Tic 1, relaxation
2	3 ^a	Review of competing response for Tic 1, modification of competing response, behavioral chart creation, and tangible rewards	Session conducted jointly	
2	4 ^b	Brief check-in, review of competing response for tic 1, modification of behavioral chart and tangible rewards, relaxation	Differential attention, effective instructions	Competing response practice for tic 1, competing response creation for tic 2
2	5 ^a	Competing response practice, behavioral chart for at-home use, tangible rewards	Session conducted jointly	
3	6 ^a	Relapse prevention, competing response practice, relaxation, home-based rewards, and behavioral chart	Session conducted jointly	
3	Boosters ^c	Check-in and problem solving; review of awareness training and creation of competing responses	Session conducted jointly	

^a duration of session =1.5 hours; ^b duration of session =1 hour; ^c duration of session = 30 minutes per session

III. STUDY 2: CASE SERIES

INTRODUCTION

Tic disorders (TDs) are neurodevelopmental disorders characterized by rapid, sudden, involuntary, repetitive, and non-rhythmic phonic (i.e., coughing, whistling) and motor (i.e., shoulder shrugging, head nodding) tics, and are estimated to affect at least 300,000 children in the United States alone (Cavanna et al., 2017). Tics typically present at approximately age six, with the peak severity of tic symptoms occurring at approximately age nine (Wolicki et al., 2019). Importantly, TDs can negatively impact youths' overall functioning across several domains (Conelea et al., 2011), including school or educational experiences (Tourette Association of America [TAA] Impact Survey Working Group, 2018) and quality of life (Storch et al., 2007). Notably, as a result of their TD, many youth with TDs experience some form of negative consequence, such as bullying, or being asked to leave class at school or extracurricular activities (TAA, 2018). Additionally, it is estimated that over 80% of youth diagnosed with a TD also meet criteria for an additional co-occurring diagnosis (Hirschtritt et al., 2015; Wolicki et al., 2019). Specifically, it has been suggested that between 32-62% of youth with a TD meet criteria for attention-deficit/hyperactivity disorder (ADHD), approximately 50% meet criteria for an anxiety disorder, and over 33% exhibit clinical elevations in obsessive-compulsive symptoms (Hirschtritt et al., 2015). Notably, families report that they often experience difficulties managing their child's co-occurring diagnosis symptoms in addition to the TD symptoms, and report that sometimes the burden of managing the co-occurring disorder is the most significant stressor they experience (TAA Impact Working Group, 2018).

Comprehensive Behavioral Intervention for Tics (CBIT) is a well-established behavioral intervention for the management of TD among youth. CBIT is typically conducted across roughly eight to ten sessions over approximately 10 weeks and has been shown to be an effective intervention modality, with similar effect sizes when compared to antipsychotic medications, and without the associated side effects of these medications (Piacentini et al., 2010). As a result, CBIT has been deemed the first-line intervention for youth with TDs both nationally (American Academy of Child & Adolescent Psychiatry, Murphy et al., 2013; American Academy of Neurology; Pringsheim et al., 2019) and internationally (Canadian Guidelines, Steeves et al., 2012; European Guidelines, Verdellen et al., 2011). Nonetheless, families often experience several significant barriers to accessing specialty treatment, specifically CBIT. A lack of access to appropriate services is often compounded by a perceived burden associated with traveling for weekly services (Woods et al., 2010) and presumed high costs of services (TAA Impact Working Group, 2018; Woods et al., 2010). One example is a recent study by Wolicki and colleagues (2020), which showed that although 77.4% of their sample ($N=115$ children) was able to access interventions for TDs, only 17.1% of the subsample indicated receiving CBIT or habit reversal training as part of the intervention. Further, a study conducted by the Tourette Association of America (TAA Impact Working Group, 2018) found that only 36% of their sample ($N= 623$ parent-report on their child with Tourette's Disorder) had ever received CBIT, whereas approximately 45% indicated being unfamiliar with the treatment. In addition, several respondents indicated not having insurance coverage for CBIT nor access to a provider in their geographic region. Lastly, an important limitation to CBIT is that it does not directly address co-occurring diagnoses

(e.g., ADHD, OCD) that typically accompany TDs and are often reported by families to be as impairing as TD, if not more. Given the multitude of limitations posed to families that receive or attempt to receive CBIT, such as barriers to accessing efficacious treatment, cost of services, and a reported need to address co-occurring diagnosis symptoms in treatment, there is an important need for innovative strategies to increase treatment accessibility.

Attempts at mitigating barriers to accessing treatment have emphasized the use of telehealth to deliver CBIT services. Telehealth or videoconference formats have become increasingly popular and provide an alternative to in-person services (Connolly et al., 2020), particularly for hard-to-reach families (Comer & Myers, 2015; Whaibeh et al., 2020). Given that mental health services often do not require in-person interactions to be completed (i.e., rely primarily on the ability to observe and communicate verbally), telehealth platforms present a feasible approach to increasing access to evidence-based psychological services (Comer & Barlow, 2014). Recent studies evaluating telehealth CBIT have found promising support for its utility. For example, Himle and colleagues (2012) evaluated the utility of CBIT delivered over videoconference compared to standard, in-person CBIT with 20 youth, and found both formats to be effective in reducing tic symptoms and to be acceptable to caregivers and their children. Similarly, Ricketts and colleagues (2016) evaluated telehealth CBIT delivery in 20 youth and found similar reductions in tic severity as those found in a large-scale randomized controlled trial of CBIT (Piacentini et al., 2010). Taken together, internet-based platforms may pose a unique opportunity to facilitate families' access to treatment providers specialized in CBIT, without sacrificing the quality of care and effectiveness of treatment. Nonetheless,

both studies utilized a one-to-one weekly treatment delivery format, which may reduce the number of patients a clinician can treat or may fill their caseload. Therefore, a possible modality to increase accessibility and acceptability may be to use an intensive telehealth group-based format to deliver CBIT.

Previous research has illustrated the benefits of conducting intensive outpatient programs (IOPs) when it comes to increasing access to specialty evidence-based services (e.g., Pelham & Fabiano, 2008). Notably, evidence on intensive, group-based treatment to address common TD comorbidities suggests that IOPs are efficacious and result in symptom reduction and functional improvements (e.g., Cook et al., 2012; Cornacchio et al., 2019; Pelham & Fabiano, 2008). For example, a one-week intensive outpatient adaptation of Parent-Child Interaction Therapy (PCIT; Funderburk & Eyberg, 2011), a well-established and evidence-based intervention to address child externalizing behavior problems, was demonstrated to significantly reduce child externalizing problems and parenting stress (Graziano et al., 2020). Importantly, these gains were maintained at a follow-up assessment four months following treatment termination (Graziano et al., 2020). Further, Santucci and Ehrenreich-May (2013) evaluated a week-long intervention for youth with separation anxiety disorder using a randomized controlled trial, which resulted in a reduction in symptom severity and impairment in children and noted high levels of satisfaction within the immediate treatment group compared to a waitlist control group.

There is also emerging evidence to support the utility of an intensive individual- (i.e., Blount et al., 2018; Flancbaum et al., 2011) and group-based (i.e., Heijerman-Holtgreffe et al., 2020; Kennedy et al., 2016) CBIT formats for the treatment of TDs.

However, all individually based intensive intervention formats to date either (1) were conducted with an adult population or (2) required prior treatment of comorbidities before enrollment and/or did not explicitly address co-occurring diagnosis symptoms in treatment. Importantly, a recent study by Bhikram and colleagues (2021), found that only 32% of providers across four geographic regions (Canada, United States, United Kingdom, and Europe) offered CBIT in a group-based format. Of note, most respondents indicated practicing in urban areas, potentially limiting access for families living in rural areas or who are unable to travel.

A common thread among several of the aforementioned IOPs includes the incorporation of interventions for both caregivers and children. Specifically, caregivers are taught their own skills (i.e., parenting strategies), as opposed to being incorporated into treatment through brief updates on session content at the end of a session. Including caregivers as an integral component of treatment, may, in turn, address the limitation of many manualized, evidence-based programs not predominantly and/or robustly intervening on both children and caregivers to address more complex clinical presentations. In turn, intensive, group-based treatments present a window of opportunity for the fusion of caregiver and child services, which may prove beneficial when it comes to increasing access to evidence-based services conveniently for families. Notably, some evidence suggests that intensive interventions for youth, particularly for those whose mental health trajectories are complex and multi-faceted, can be a cost-effective alternative to traditional outpatient (Hare & Graziano, 2020) and inpatient clinical services (e.g., Boege et al., 2020). Therefore, the adaptation of evidence-based caregiver and child interventions to a comprehensive intensive outpatient format may aid in

addressing traditional barriers to treatment, including financial constraints, access to specialized treatment, low family engagement, and clinical case complexity (e.g., Guo et al., 2020).

The literature on the treatment of TDs has grown tremendously in the previous decade, however, significant gaps persist. While preliminary support exists for the use of intensive treatment formats to independently address TDs and other mental health conditions, formats that comprehensively address co-existing conditions are lacking. This is concerning given the impact that co-occurring diagnoses have on impairment in quality of life and functioning for youth with tics. To address this limitation, studies have aimed to address problems associated with co-occurring diagnoses (i.e., Scahill et al., 2006; Sukhodolsky et al., 2009), but have primarily been conducted using a standard weekly session format, with limited to no emphasis on CBIT-related strategies. Additionally, there is evidence to suggest that addressing psychosocial impairment related to tics can also improve the quality of life for youth with TDs (McGuire et al., 2014). Nonetheless, these protocols typically entail a one-time weekly session treatment format. Although there is evidence to support the use of evidence-based interventions to address TDs and co-occurring diagnoses separately, to date, there is only a single case study that has examined an intervention to address co-occurring diagnosis- and TD-related symptoms simultaneously in an intensive format (Dale et al., 2021, see Study 1). This study combined core components from CBIT and Behavioral Parent Training (BPT) to simultaneously address tic and co-occurring ADHD symptoms. The results of this case study indicated a decrease in tic symptoms that were maintained four months post-treatment and parent-reported improvements in overall quality of life. While this study

takes an important initial step towards simultaneously addressing tic and co-occurring diagnoses and was foundational in the development of a novel treatment intervention, described below, no studies to date have evaluated the combined, group-based intensive modality and used telehealth to address co-occurring and tic-related symptomatology simultaneously and rapidly.

The aim of the current paper is twofold. First, we aim to present a clinical overview of the structure and content of Taming Tics Together, a novel 5-day intensive group-based intervention conducted via telehealth using lessons learned from a previous intensive CBIT intervention (Dale et al., 2021, see Study 1). Specifically, we have modified the treatment format from an exclusively individually-based to a group-based format, however, we have retained the opportunity for youth to meet individually with a therapist, allowing for the time and space in treatment to address tic and co-occurring diagnoses in a person-centered manner. To provide an overview of the Taming Tics Together protocol, we will highlight the initial intake assessment procedures, staff required to facilitate this program, an overview of the five-day, telehealth-based intensive intervention format, and booster sessions. Second, using a case series design, we present outcome measures for tics and co-occurring diagnosis symptoms at baseline, during treatment, post-treatment, and at the 1-month follow-up. In addition, our study builds upon the prior case study by incorporating family feedback regarding improvements to treatment and lessons learned within the case study. Finally, this overview of Taming Tics Together and case series example offers preliminary support for the feasibility, utility, acceptability, and effectiveness of this treatment format.

METHODOLOGY

Treatment Development and Rationale

The protocol for Taming Tics Together (TTT) was developed by the first and last authors. TTT builds upon established evidence-based interventions for TDs (e.g., Woods et al., 2008) and common co-occurring disruptive behaviors and anxiety disorders (e.g., BPT, Forehand et al., 2012; Group-based CBT, Silverman et al., 1999). All treatments utilized were evidence-based and adapted to an intensive intervention format for TDs and co-occurring conditions (Dale et al., 2021, see Study 1).

Specifically, TTT offers caregiver-only and child-only group-based sessions, as well as individually based child-only sessions to provide one-to-one intervention strategies targeting each child's and family's unique presenting symptoms. All child group-based strategies involve tic-specific skills (i.e., awareness and habit-reversal training), transdiagnostic skills (i.e., relaxation techniques, problem-solving, emotion regulation), and peer support content related to living with tics. All caregiver group-based strategies also involve tic-specific skills but include additional strategies to manage co-occurring behavior (i.e., praise, behavioral and reinforcement systems), and caregiver support content to help children manage their tics and co-occurring diagnoses (i.e., talking to your child's school about each diagnosis; see Figures 1 - 4 for selected manual excerpts of the TTT manual).

TTT Team Members

TTT requires a 1:1 child-to-staff ratio for individual sessions, one to two group leaders (caregiver and child group leaders can be the same clinicians, referred to as the "group leader"), and at least one supervisor who can join telehealth sessions as needed.

An additional group leader is beneficial in the telehealth format to address technology-related issues (i.e., family difficulties logging into session, difficulties related to audio or video) and may aid in the management of materials and tallying points while the other group leader facilitates discussion. However, in a non-training clinic where additional clinicians may not be easily accessible or available, two clinicians would likely be sufficient to run groups and meet with patients individually, thus, eliminating the need for additional staff. Depending on the size of the group, it may be beneficial to have one additional “floater” clinician as a backup who may join sessions as needed, however, group leaders may also fill this role if staffing capacity is limited.

Children are paired individually with one clinician (henceforth referred to as the “individual clinician”) with whom they will meet during their initial intake assessment, on a daily basis during the program, and for booster sessions. The group leaders are typically masters- or pre-doctoral-level clinicians with prior CBIT and co-occurring diagnosis treatment (i.e., CBT, BPT) experience. The program is always overseen by a licensed clinical psychologist, who provides daily supervision to all staff members. Staff members are currently recruited within the affiliated university’s clinical science child- and adolescent-focused doctoral program and typically include advanced graduate student clinicians. Before participation as a staff member, students are required to attend a training, typically conducted over one to two days, which includes an overview of TDs and co-occurring diagnoses, an extensive review and opportunity to practice utilizing TTT skills, and a review of assessment and intervention procedure videotapes. All team members are required to extensively review the manual prior to participation in the program.

Measures

Tic Symptoms and History. The Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) is a semi-structured clinical interview used to assess past and current tic symptoms and is currently the gold standard for assessing tics in children (Martino et al., 2017). The YGTSS evaluates the frequency, number, complexity, intensity, and interference of motor and vocal tics, using a 6-point scale (range 0-5). Further, an impairment score (range 0-50) is also collected. In the current case series example, the Total Tic Severity Score (YGTSS-TSS), Impairment Score (YGTSS-I), and Global Tic Severity Scale Score are presented.

Subjective Units of Discomfort (SUDs). As outlined in the treatment manual from Woods and colleagues (2008), a SUDs rating was used to evaluate the amount of discomfort/distress associated with each current tic. Youth rate the level of discomfort/distress each tic causes on a scale from 0 (not present or not causing discomfort/distress) to 10 (creating significant discomfort/distress).

Co-occurring Diagnosis Symptoms.

Internalizing Symptomatology. The Revised Children's Anxiety and Depression Scale (RCADS; Chorpita et al., 2000) is a 47-item child- and parent-report evaluating internalizing symptomatology. Subscales include separation anxiety disorder, social phobia, generalized anxiety disorder, panic disorder, major depressive disorder, and obsessive compulsive disorder. Items are scored on a 4-point Likert scale (0 = Never, 3 = Always). The RCADS has shown good reliability when used in a clinical sample (Chorpita, Moffitt, & Gray, 2005).

Externalizing Symptomatology. Parents completed the Disruptive Behavior Disorder Rating Scale (DBD-RS; Pelham et al., 1992), a 42-item questionnaire assessing symptoms of conduct disorder (CD; 16 items, e.g., “has been physically cruel to people”), oppositional defiant disorder (ODD; 8 items, e.g., “often argues with adults”), ADHD-inattention (9 items, e.g., “is often easily distracted by extraneous stimuli”) and ADHD-hyperactivity/impulsivity (9 items, e.g., “often talks excessively”). Items are rated on a 4-point Likert scale, ranging from 0 (not at all) to 3 (very much).

Upon completion of screening self- or parent-report assessment, a child and/or caregiver who endorses elevations in a certain domain would be administered an additional assessment to evaluate symptoms further. For example, should a family endorse elevations on the initial screening measures for obsessive-compulsive spectrum symptoms would then receive the clinician-administered Children’s Yale-Brown Obsessive Compulsive Scale (CY-BOCS; Scahill et al., 1997), whereas a child and/or caregiver who endorses separation anxiety will partake in the clinician-administered Kiddie Schedule for Affective Disorders and Schizophrenia for School-age Children (K-SADS; Kaufman et al., 2000).

Global Functioning. Each child’s overall global functioning was measured via the Children’s Global Assessment Scale (CGAS; Shaffer et al., 1983). The CGAS is a clinician-rated measure of youth’s global functioning and is commonly utilized. Clinicians rate the child’s overall functioning on a scale ranging from 0 to 100, with higher scores indicating less impairment and greater functioning. The CGAS was rated by an evaluator at each timepoint (baseline, immediately post-treatment, and 1-month following the end of treatment).

Feasibility and Acceptability.

To evaluate overall treatment acceptability and satisfaction, parents will complete the Client Satisfaction Questionnaire (CSQ; Larsen et al., 1979). The CSQ was utilized to evaluate overall satisfaction with the program. The CSQ is a brief, 8-item parent-report measure. Items are rated on a 4-point Likert scale, with scores ranging from 8 to 32. Higher scores on the CSQ indicate greater satisfaction with treatment. The CSQ was administered at the 1-month follow-up timepoint. Additionally, parents also provide qualitative feedback (i.e., *What would you describe as your most favorite component(s) of treatment?*) on the overall treatment. Finally, attendance was taken at all sessions.

Intake Assessment Procedures

Prior to enrollment in treatment, children are assessed to determine fit for the program. Assessments are conducted entirely via remote technologies (i.e., videoconference, phone). Following the informed consent process, caregivers and youth ages 10 and older complete a series of questionnaires (via a secure online platform) regarding tic and possible co-occurring symptoms, emotion regulation, sleep, parenting/caregiver relationship and practices, adverse childhood experiences, among other clinically relevant assessments.

Caregivers and youth meet separately with a clinician to complete the YGTSS as well as diagnosis-specific diagnostic assessments, however, modifications can be made to complete the YGTSS collaboratively with the caregiver and child together if needed or if deemed more clinically appropriate. Evaluations are conducted to determine if the child meets criteria for a TD and has, at minimum, elevations in co-occurring symptomatology. Families are excluded from participating in the program if the assessment determines that

(1) the primary or co-primary diagnosis is not a TD and/or (2) the child has a formal diagnosis of an autism spectrum disorder.

Treatment Overview

The program occurs across five consecutive days and consists of three main components: (1) child-only group, (2) caregiver-only group, and (3) one-to-one or individual sessions. The program is broken down into three unique formats to allow for peer- and caregiver-based support with discussion in each group, and a person-centered approach during individual sessions to address each child's unique needs. TTT child-only groups are held in the morning, whereas parent groups are held in the evening, to allow for more flexibility in scheduling individual sessions throughout the remainder of the day during the summertime. Families and their individual clinicians coordinate their schedules. TTT is open to children between the ages of seven and 17, grouped by age (e.g., children ages seven to nine, children ages 10 to 12, children ages 13 to 15, etc.). With adequate staff and patient volume, multiple simultaneous groups can be run throughout the day. Groups are limited to no more than 10 children per group to ensure that each child is able to receive individual attention, and caregiver sessions are a comfortable size to encourage participation and discussion. Given its current telehealth/videoconference-based format, no physical space is needed, however, it is recommended that all participants, including staff, find a space within their home/office that is quiet and away from others for confidentiality and privacy purposes.

Child-Only Group. The child-only group is designed to provide an opportunity for children to meet same-aged peers who also experience tics and co-occurring symptoms. This component of treatment allows children to freely discuss living with tics

and helps address the psychosocial challenges associated with TDs, while also gaining an introduction to tic-related content they will practice in their one-to-one sessions. Further, children learn and practice transdiagnostic content (i.e., problem-solving, cognitive-behavioral triangle), engage in peer-support activities (i.e., Peer Support Activity, see Figure 4), and participate in activities that require physical participation (i.e., Scavenger Hunt), to increase group participation, provide opportunities to receive praise and rewards, and engage in an active “break” from screen time. Throughout the sessions, children can earn points towards a reward at the end of the week (typically an electronic gift card). Behaviors included in reward charts often entail using competing responses, actively participating in activities, and following activity instructions. Behaviors that earn rewards are discussed at the beginning of each group session, and children are informed of the number of points they have earned throughout the session. Points are tallied each day and are modeled as an example of a behavioral chart and reward system during caregiver-only groups.

Caregiver-Only Group. Caregivers participate throughout TTT in daily evening sessions from Monday through Friday. During this time, the group leader (typically the same clinician who leads the child-only group) provides an overview of tic-specific and transdiagnostic skills (see Table 1 for the full schedule of training topics) through didactic and discussion within the group. Caregivers, typically parents, are encouraged to invite additional family members or adults who play a significant role in child-rearing (i.e., grandparents, aunts/uncles, adult siblings). During these daily sessions, parents are introduced to tic-specific skills (i.e., psychoeducation, awareness training, competing response creation), transdiagnostic skills (i.e., behavioral and reward charts, differential

attention, talking to your child's school about areas of concern related to diagnoses) and are provided with strategies to increase their well-being (i.e., strategies to manage parental stress). Specifically, we discuss, in detail, differential attention, structure (i.e., limit setting, household rules, and routines), rewards, and consequences. Strategies are adapted to meet the presenting problems of the group (i.e., emphasis on rewarding brave approach behavior for youth with anxiety, emphasis on using differential attention to ignore disruptive behavior and/or avoidance behavior). Caregivers are encouraged to share their experiences and/or offer support to other families.

One-to-One Sessions. One-to-one sessions are offered for approximately 90 to 120 minutes per day, every day of the five-day program. Families and their individual clinician schedule the sessions based on their availability (i.e., one two-hour block, two one-hour blocks, etc.), considering the best time of day for each child to attend sessions. For example, children who experience more tics in the afternoon may benefit from having their sessions conducted at that time, whereas children who experience tics across the day may choose to participate at a time that is most convenient for their family. During each session, the individual clinician tailors the session content to meet the child's unique needs, however, all sessions include an emphasis on tic-related strategies (see below for case examples). The reward system established during the child-only group can be extended to individual sessions as needed in order to provide children with the opportunity to continue earning points towards a reward at the end of the week. Additional skills discussed in one-to-one sessions also include exposure and response prevention, organizational skills, cognitive restructuring, present-moment awareness, emotion identification, and additional relaxation strategies.

Booster Sessions. Given that strategies to address tics and comorbidities often require additional time to fully implement and test learned skills, optional 30-minute booster sessions (typically offered two and three weeks following the end of treatment) are also provided. Families who choose to participate in booster sessions can, for example, discuss any specific questions they have regarding their child’s progress, or review and/or problem-solve/brainstorm strategies. Most children have made significant improvements in tic symptoms following the end of treatment; therefore, several families may choose not to participate. However, some families find the option to “check-in” as needed as a valuable resource to ensure the maintenance of gains achieved during treatment, or, in cases when children are still experiencing difficulties, families can discuss additional services as needed.

Telehealth Considerations. Given the differences in conducting treatment groups via telehealth compared to in-person, several additional steps were taken to ensure groups ran as smoothly as possible. First, prior to intake, families completed the standard clinic informed consent, as well as a telehealth consent form. Both documents were reviewed via phone before completing any study measures, as well as before partaking in the videoconference-based initial intake appointment. Families were notified of the unique limitations of confidentiality associated with the telehealth format, as well as the steps that were taken to reduce risks to confidentiality. For example, families were asked to locate a specific area in their home where the caregiver(s) and child could participate in their sessions. When possible, families were asked to find a separate room or space where they would be able to participate uninterrupted. When a separate space was not feasible, families were asked to use headphones and make sure they were facing away from others

in their home (i.e., sitting with their back against a wall to prevent others from seeing their screen). Further, at least one caregiver was required to be at home with the child during all group and individual sessions. Caregivers also provided a phone number where they could be reached during these sessions if needed. Finally, to mitigate technology difficulties, families were provided with additional technology-related support as needed (i.e., difficulties logging onto the videoconferencing software, headphone connection difficulties) and were addressed prior to beginning treatment to ensure limited technology-based interruptions when the week-long program began. Nonetheless, a study staff member was made available to help with technology-related difficulties during daily sessions (group or individual) should they arise. All families were asked to fully charge their devices before a session, or be seated in an area where their devices can be charging during sessions. All families in the current study had access to headphones, an electronic device with a camera (i.e., tablet, computer, smartphone), and the internet, however, it may be beneficial to have such materials available to families to borrow and return after sessions are completed to ensure technology barriers do not prohibit participation.

CASE SERIES PRESENTATION

The purpose of the case series is to provide an illustrative example of the Taming Tics Together protocol and modifications made to meet each family's individual needs. We will first present each case example, followed by the content used to address individual presenting concerns. Next, we present treatment outcomes outlined by symptom area and conclude with a brief overview of booster sessions and their content.

Participants

Please see Table 2 for a comprehensive overview of participant demographic characteristics.

Mike. Mike was a nine-year-old White, non-Hispanic male, who was being homeschooled at the time of intake. Prior to intake, he had not received any prior diagnoses. Given his tic symptom history, he met criteria for Tourette's Disorder. Additionally, at the time of intake, Mike met criteria for ADHD, combined presentation. To manage symptoms related to ADHD, Mike's caregiver reported having used an incentive system at home. No other history of inpatient, outpatient, or psychopharmaceutical treatment was reported.

Kevin. Kevin was a nine-year-old, Black, non-Hispanic male who was attending school remotely due to COVID-19 restrictions. Due to the wide disbursement of the patient's symptoms, he failed to meet criteria for any one anxiety disorder and was thus diagnosed with an Unspecified Anxiety Disorder. Given his history of tic symptoms, he was diagnosed with Tourette's Disorder. He had a history of receiving cognitive behavioral therapy for tic concerns and had a basic foundational understanding of CBIT strategies, however, he had primarily been taught to use breathing techniques to manage his tics. Caregivers reported that he had concluded treatment with his previous provider before enrolling in TTT. At the time of treatment, Kevin was prescribed Clonidine to manage difficulties with sleep.

Henry. Henry was an 11-year-old White, Hispanic male, whose school format was not reported at the time of intake. His caregivers reported a previous diagnosis of ADHD, combined presentation, and Tourette's Disorder. Diagnoses of Tourette's

Disorder and ADHD, combined presentation, were confirmed at intake. Caregivers also indicated a previous history of medication use to address ADHD symptoms (i.e., Guanfacine) but reported significant side effects that led to the discontinuation of those medications. Caregivers did not report a history of behavioral treatment for ADHD or tics. At the time of intake, Henry was prescribed Adderall to manage his symptoms.

Individual Session Content

Mike. The individual clinician met with Mike for a total of 90 minutes per day, with breaks taken as needed. During individual sessions, tic symptoms were addressed using the standard TTT protocol. To manage ADHD symptoms, a token-reward system and attention questions were used whereby Mike would earn points for exhibiting target behaviors (i.e., recognizing the urge to tic, practicing competing responses) as well as bonus points for answering attention questions related to TTT content (i.e., “what is the first step of creating a competing response?”) within the session.

Kevin. The individual clinician met with Kevin twice a day for a total of 120 minutes (two 60-minute sessions/day). The first session each day was designated for addressing tics, and the second for addressing elevated anxiety symptoms. The CBIT content built upon Kevin’s prior basic knowledge of CBIT; as such, the clinician and patient utilized session time to review and create competing responses for existing tics, as well as creating competing responses for pretend tics (relapse prevention) to ensure the continued practice of treatment strategies for the patient. Content to address anxiety included psychoeducation on emotions and anxiety, as well as a review of common cognitive distortions and practice using cognitive restructuring and problem-solving strategies.

Henry. The individual clinician met with Henry for a total of 90 minutes per day, with breaks as needed. Tic-related difficulties were addressed using the standard treatment protocol. Similar to Mike, modifications to the group-based point system were made to reduce his inattentive, hyperactivity, and impulsive symptoms. As such, attention questions were embedded within the intensive token-reward system to increase engagement within sessions.

RESULTS

Tic Symptoms and SUDs Ratings

The number of tics reported and addressed (i.e., competing responses were created) in treatment are presented in Table 3. Overall, all three children demonstrated reductions in tic SUDs on day 5 compared to day 1. Youth were successful in addressing at least four tics within the context of individual sessions. While several tics across each youth's tic hierarchy were not explicitly addressed, it is of note that SUDs ratings across several non-addressed tics also decreased (see Figures 5 - 7).

YGTSS Scores

Unfortunately, Mike was unable to be contacted for post and 1-month follow-up assessments, therefore, only Kevin's and Henry's YGTSS (see Figure 8), corresponding co-occurring diagnosis assessments (for a visual depiction of Kevin's results, see Figure 2), and overall satisfaction with treatment results are described below. YGTSS score descriptive ranges, as indicated by McGuire and colleagues (2021), were utilized. At baseline, Kevin obtained a YGTSS Total Tic Severity score (YGTSS-TSS) of 12/50 falling into the moderate range. At post, he received a YGTSS-TSS score of 11/50 and remained in the moderate range. However, at 1-month post-treatment, he received a

YGTSS-TSS score of 0/50, falling into the normal range. His YGTSS impairment (YGTSS-I) scores, at baseline, post, and 1-month following, were 10 (moderate), 0 (normal), and 0 (normal). YGTSS Global Tic Severity Scores were, at baseline (22; mild), post (11; mild), and 1-month follow-up (0; borderline).

At baseline, Henry obtained a YGTSS-TSS of 24/50, falling into the moderate range. At post, he received a YGTSS-TSS score of 8/50, falling into the mild range. Finally, at 1-month post-treatment, he received a YGTSS-TSS score of 4/50, falling into the borderline range. YGTSS-I scores, at baseline, post, and 1-month following treatment, were 36.5 (marked), 15 (moderate), and 0 (normal), respectively. YGTSS Global Tic Severity scores were, at baseline (60.5; marked), post (23; mild), and 1-month follow-up (4; borderline).

Co-occurring Symptoms

Per parent report on the RCADS, Kevin was exhibiting clinically elevated symptoms of separation anxiety and panic disorder, and borderline clinical symptoms in the overall anxiety and combined anxiety and depression scales at baseline. At the 1-month assessment, Kevin no longer exhibited clinical or borderline elevations across all domains (see Figure 9). Henry's presenting co-occurring difficulties at baseline, per parent report on the DBD-RS, were clinically elevated on ADHD-hyperactive/impulsive scale (8/9 items endorsed), with his mother endorsing 15/26 items across the ADHD and oppositional defiant disorder (ODD) subscales. At the 1-month follow-up assessment, he continued to exhibit clinical elevations on this scale, however, noted a reduction in symptoms (7/9 items endorsed) and a reduction in overall clinically elevated endorsed (13/26) items across the ADHD and ODD subscales.

Clinical Opinion on Improvements and Families' Treatment Satisfaction

Per an independent evaluator's report, Kevin received a score of 70 (some difficulty in a single area, but generally functioning pretty well), 90 (good functioning in all areas), and 90 on the CGAS at baseline, post, and 1-month follow-up, respectively. Henry received a score of 50 (moderate degree of interference in functioning), 95 (superior functioning), and 95 on the CGAS at baseline, post, and 1-month follow-up, respectively by the independent assessor.

Both Kevin's and Henry's parents completed the CSQ at the 1-month follow-up. Parents reported high levels of treatment satisfaction at the 1-month follow-up ($M=31.25$, $SD=.96$, $N=4$). Both families also indicated that the child-only group was helpful in that it provided an opportunity to meet others with similar difficulties and felt that the caregiver-only group was beneficial to provide detailed information about services and additional supports. Finally, caregivers indicated satisfaction with the one-to-one sessions, as the individual sessions provided additional clinical support for their child. Further, attendance rates for the child-only group, caregiver-only group, and one-to-one sessions were 100%, 93.33%, and 100%, respectively, for an overall attendance rate of 97.8% (one caregiver was unable to attend one of the caregiver-only sessions).

Booster Session Attendance

Two of three families chose to participate in booster sessions. Kevin's family participated in one booster session approximately 3-months post-treatment. At this time, Kevin and his individual clinician reviewed the process of awareness training and creating competing responses, brainstormed competing responses for two additional tics, and importantly, discussed telling his teacher about tics and having a "secret signal" as a

reminder to do competing responses while in class. Henry's family participated in one booster session approximately two weeks post-treatment. At this time, Henry and his individual clinician reviewed the process of the awareness training and creating competing responses. With his parents only, his clinician reviewed how to implement a daily reward system and use a reward menu to address concerns related to his ADHD symptoms.

DISCUSSION

The primary aim of the current study was to provide an overview of a newly developed intensive, internet-based group intervention for youth with TDs and common co-occurring diagnoses, as well as offer preliminary support for treatment acceptability and utility. Youth in the current study exhibited significant improvements across their tic symptoms, as indicated by self-reported SUDs ratings and independent evaluator's assessment at post- and 1-month follow-up on YGTSS scores. Specifically, per parent report, Henry exhibited an 83.3% reduction, and Kevin's exhibited a 100% reduction from baseline to 1-month following the end of treatment on the YGTSS-TSS. Henry's total number of ADHD symptoms also decreased following the end of treatment, and caregivers reported overall satisfaction with treatment, indicating that their needs were met in treatment. It is important to note that previous studies have suggested that youth with disruptive behaviors and anxiety have more difficulty engaging in treatment (Himle et al., 2012), particularly if conducted via telehealth, however, these studies have often not explicitly addressed the co-occurring diagnoses within the context of treatment to manage such behaviors. By incorporating and addressing co-occurring symptoms concurrently with TD symptoms, we were able to successfully address each youth's

symptoms (and subsequently reduce their comorbid symptom interference in the acquisition of tic symptom skills). Further youth were actively engaged in each component of treatment, as shown in the points received for group and individual session participation.

The evidence from the current study continues to fill an important gap in the TD treatment literature by providing preliminary evidence to simultaneously address tics and co-occurring symptoms and adds to the literature on telehealth CBIT. Importantly, IOP programs, such as TTT, present a promising avenue when attempting to increase access to treatment. Importantly, this case series builds upon qualitative feedback provided by a previously participating family (Dale et al., 2021, see Study 1) and highlights areas of improvement in the treatment protocol as a result. Both families who completed post-treatment assessments indicated that the treatment format and content were acceptable, each highlighted the value of engaging in a group-based format as a mechanism for meeting others with TDs, as well as providing and gaining access to peer and caregiver support. Further, attendance rates were high, suggesting that this format was easily accessible to families and engagement in treatment was high.

Given that access to providers trained in both CBIT and interventions for common co-existing diagnoses is limited, the current IOP format provides the unique opportunity for families to access treatment providers briefly and thus, may aid in reducing or eliminating the barrier of having limited access to specialty providers by conducting treatment via a virtual modality. Given the flexibility to conduct sessions at any time throughout the day (when offered during school breaks or over summer vacation), this format is flexible to accommodate differing schedules. TTT provides each family with

approximately four to five hours of treatment per day, for a total of roughly 20 to 25 hours of intervention services across the five days of treatment, with the possibility of an additional hour of treatment should families participate in booster sessions; this is comparable to the amount of time spent in treatment when CBIT and co-occurring diagnosis intervention sessions are conducted at separate times. Overall, our study's results suggest that a combined, intensive, telehealth group approach is likely an acceptable alternative to standard intervention modalities, while simultaneously reducing traditional barriers to accessing evidence-based treatment.

This study is not without limitations. First, the results are based on a small sample size and, therefore, may not be generalizable. Second, there is variability in coverage for intensive intervention services and telehealth services, therefore, this format may not be suitable or feasible for all families. Insurance companies and policymakers should consider extending coverage to intensive intervention formats, and telehealth-based intervention formats, and the combination of the two, to address long-standing barriers related to accessing evidence-based care. Third, the current study was conducted as an open-trial design. The next important step in evaluating the TTT treatment modality involves utilizing a randomized controlled trial to rigorously evaluate treatment effectiveness. Fourth, formal clinician/staff feedback was not collected regarding their perspective on the acceptability and feasibility of the program. Informal feedback from the clinicians indicated relative ease in scheduling individual sessions, implementation of strategies, and feasibility of a telehealth-based format. Future studies should incorporate standardized and formal feedback from staff members to understand perspectives from those implementing the treatment in addition to caregiver feedback. Finally, the follow-

up assessment period was extended to only 1-month following the end of treatment, which may not be sufficient time for families to implement, incorporate and effectively use strategies, particularly those associated with disruptive behaviors (i.e., implement behavioral charts and reward systems and provide sufficient time for evaluation of utility). However, our results suggest the continued implementation of treatment strategies at the 1-month follow-up assessment. Future studies should consider extending the post-treatment assessment time to further elucidate long-term symptom trajectories.

Notwithstanding its limitations, the current study provides preliminary evidence for an internet-based intensive intervention to address TD and common co-occurring symptoms concurrently. A telehealth-based, intensive format provides the opportunity for families to exclusively focus on the strategies taught in session while limiting the impact of weekly external responsibilities from impeding session attendance and may reduce the burden of and time lost due to travel to and from a clinic. Further, telehealth-based interventions may provide a more naturalistic setting for the in-vivo practice of skills across their typical home, school, and social environments, as well as provide parents the opportunity to access in-home feedback for parenting skill implementation and ways to support their child outside of the context of a standard treatment setting. Additionally, the group-based format likely offers an opportunity to gain support from other families with similar experiences, while also increasing the number of families who are able to gain access to specialized treatment at once.

TDs and co-occurring diagnoses are prevalent among youth in the United States and can have a significant impact on youth with TDs, their caregivers, and their family as a whole. Although the number of families accessing CBIT has increased in the last ten

(i.e., 6% to 36% of youth between 2008 and 2018; TAA 2018), access to CBIT remains limited. An integral next step is to evaluate existing non-traditional or alternative treatment formats, such as TTT, using more rigorous evaluative modalities (i.e., randomized controlled trial studies) to determine the continued feasibility, effectiveness, and overall acceptability. Further evaluations may also begin to aid in determining for whom videoconference-based intensive interventions may be more suitable. As such, an intensive, group- and internet-based CBIT format which integrates intervention strategies to address common co-occurring diagnoses is a promising additional treatment modality that is likely to reduce barriers to accessing evidence-based treatment, be more cost-effective, and reduce impairment associated with TDs and common co-occurring diagnoses for youth and their families.

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Figure 1. Taming Tics Together Manual Excerpt – Child Group, Session 1

Child Group Session 1:
Introductions to Group, Psychoeducation, and Introductions to Tic Hierarchies and Awareness

Who Attends: Children/Adolescents

Time: 60 Minutes

Materials: Psychoeducation Sheet
Hierarchies
Awareness Game

Objectives:

1. Introductions/orientation to activities and the group
2. Establish group rules
3. Provide children/adolescents with psychoeducation about tics
4. Introduce tic hierarchies
5. Introduce the awareness game

Outline:

Introductions

It is important to begin this session by building rapport and getting to know each child. At this point, your staff likely will not have met each child and vice versa, therefore, spend the first five minutes introducing yourselves, playing a quick group game (see *Ice Breakers* for ideas) and getting to know each other.

- Go around the room and have each person offer the following information:
 - Name
 - Age
 - Favorite ice cream flavor

Establishing Group Rules

Create a list of group rules with all participating children and counselors. Ask children for ideas about rules, but it is important that the following are included:

1. Confidentiality
2. Listening when others are speaking by putting your microphone on mute, or sitting quietly
3. Be respectful of everyone's thoughts and opinions, even if you don't agree
4. Have fun!

Psychoeducation

What are Tic Disorders and Tics (TDs)?

Something that happens in your brain that makes your body make movements, or your voice make sounds all of a sudden and happen over and over again. They happen not because you're making them happen, but because your brain is making them happen. These are called *involuntary behaviors* because you're not doing them on purpose!

You might want to use the following dialogue to introduce tics to kids (extracted from Woods et al., 2008):

[REDACTED]

What's really important is that:

- Your parents didn't do anything to make tics happen
- YOU didn't do anything to make tics happen
- Tics are no one's fault, they aren't things you learned to do, they don't happen on purpose, they are not a habit, and they aren't something you can control right now. **BUT** we're going to learn strategies to help manage them over this next week

Prompt: does anyone have a guess of where on your body tics usually show up first?

Answer: Tics usually begin with eyes, face, and neck regions, and sometimes start with really simple noises

Sometimes, tics involve one movement, for example, just one blink, but sometimes they can include lots of movements, like blinking and head shaking. Sometimes we can hold tics in for a little while, but usually, the tics find a way to sneak out or we let the tics out at a later time, like when we get home from school at the end of the day. Sometimes, we can't hold tics in, and they just happen.

Prompt: Raise your hand if you've ever had a tic happen before!

Allow kids to see that they aren't the only ones in the group with tics. Usually, there is some kind of feeling or sensation that comes before the tic. This is referred to as a *PREMONITORY URGE or SENSATION*. Premonitory urges are feelings that come *BEFORE* the tic. Some kids and teens say they feel something uncomfortable, pressure, tension, tickle, itch, or some type of feeling in their body that lets them know a tic is coming. What usually happens after the premonitory urge or feeling is that we do the tic, and then that feeling goes away! But, what happens is that the feeling only goes away for a little while. It comes back and makes you feel the feeling to do the tic again. Tics are often worse with strong emotions, like excitement or anxiety, when children are tired or fatigued, or when we pay a lot of attention to tics. Tics are often better during calm, focused activities or in inhibiting environments

Figure 2. Taming Tics Together Manual Excerpt – Child Group, Session 5

Child Group Session 5:
Creating competing responses as a group for future tics (relapse prevention) and
Goodbyes!

Who Attends: Children/Adolescents

Time: 60 Minutes

Materials: Certificates

Worksheet 1d. Tics, Tic Signals, and Competing Responses

Relapse Prevention as a Group

During this time, it will be important to create competing responses for tics that (1) may not currently be present but tend to wax and wane for this particular child and (2) “pretend” tics that might occur in the future. This will allow the child to practice each step of creating the competing response more independently.

Come up with a pretend tic (i.e., arm jerk, head nod, etc.) and have the child walk you through creating a competing response for that tic. Have each counselor pretend to have a tic (it will be helpful to choose at least 1 motor, 1 vocal, and 1 complex tic during this practice). Have counselors pretend that they do not know the steps of a competing response and are not even sure what a competing response is! Allow the children to collaboratively walk each counselor through the steps of creating a competing response. It might be helpful to use a clean copy of “Worksheet 1d. Tics, Tic Signals, and Competing Responses” to monitor these pretend or not currently occurring tics. As a group, walk through creating a list of competing responses for these tics.

NOTE: prior to this session meet with the other counselors to discuss tics that you (1) have not been able to create a competing response for by day 5 and/or (2) that each child has experienced in the past but is no longer experiencing. This will be helpful to keep in mind when “assigning” pretend tics to each counselor to use for the competing response practice.

Wrap up, Certificates and Goodbyes!

Prior to this session, you will have already created a certificate for each child to earn during the session. Briefly, have each counselor (i.e., the counselor that worked one-on-one with the child) describe all of the child’s hard work, and virtually present the child with a certificate with their superlative. Examples include “best sharer,” “awesome problem solver,” and “great team player.” It may also be helpful to assign each kid as a “Certified Tic Tamer” to wrap up their hard work for the week.

Award final points for each child that actively participated, reminding them that these points will go towards their overall in-session rewards.

Figure 3. Taming Tics Together Manual Excerpt – Individual Session 1

Individual Session 1:

Hierarchy creation and awareness game; (2) Relaxation (deep breathing); (3) introduce in-session rewards and brainstorm rewards

Who Attends: Children/Adolescents; parent(s) for 30 minutes

Time: 100 Minutes

Materials: Hierarchies

Awareness game

Reward chart template

Reward menu template

Relaxation – deep breathing instructions

Objectives:

1. Create an in-session reward chart
2. Brainstorm at-home rewards
3. Create tic hierarchies
4. Re-introduce and practice the awareness game
5. Assign at-home practice

Outline:

Rewards

It may be important to decide upon an in-session reinforcement reward system *prior* to beginning the session. Depending on the child's presenting symptoms, it may be important to create a more rapid reinforcement system (i.e., 1 point for each attempt to practice skills in session). See sample charts in the appendix for an example, however, rewards should include, at the very least,

1. Points for attending the session
2. Points for participation in session activities (may be awarded per activity, per 15-minute interval, etc.)
3. Points for completion of homework. It will likely be important to use increased points for homework completion to ensure compliance.

Depending on the child's age, you may choose to use a colorful chart or a contract (see appendix)

When creating the reinforcement and reward systems, keep in mind that each family will be using these materials (or a similar version) at home, therefore, make sure to keep these systems clear and S-I-M-P-L-E. While you create these systems in session, it may also be helpful to create a corresponding reward menu for at-home AND in-session participation. Parents will also learn about creating these materials during the parent group session, however, the introduction during this section to the child will be helpful to start their thinking about rewards.

Troubleshooting

If the child is having difficulty identifying tics, you may want to try modeling a tic and identifying it for the child. You may use the following dialogue:

To start, your counselor is going to pretend to have a tic, and it's going to be your job to point out when your counselor is doing the tic. You can do this by lifting one finger into the air like this [Show pointing index finger straight up into the air, as if you are holding up the number 1]. For this round, let's choose to catch a shoulder shrug tic [show the child what a shoulder shrug tic would look like]. Remember, whoever notices the tic first and raises a finger gets a point!

If the child is having difficulty identifying an urge:

Spend at least 35-50 minutes trying to practice identifying an urge. Attempt to ask about the urge in alternative ways, such as:

- *Do you get a feeling like you have to do the tic before it happens?*
- *Do you notice a feeling, like an itch, a tingle, or tickles in your body near where the tic happens?*
- *Do you have any thoughts that make you feel like you have to make the tic?*

Additionally, it may be helpful to use the language from the Premonitory Urge for Tics Scale (PUTS; Woods, 2005) to ask questions about the tics.

Finally, it may also be helpful to take a short break from the awareness game and try another, more exciting game and/or an even more boring game.

NOTE: today is only the first day of awareness training. Children will be assigned awareness practice for their evening homework, so it is okay if they leave the session without identifying an urge.

Relaxation Strategy # 1: Deep Breathing

Using "Instructions 2a. Deep Breathing" decide on the type of deep breathing you will use. You can ask the child if they would prefer to start deep breathing or balloon breathing, or if they would like to call it something else. Introduce deep breathing to the child and then begin practicing. Use about 5-10 minutes of the session to practice deep breathing. It may be helpful to set a 1-minute timer to practice deep breathing, then quickly check in about how the child feels, then do another round. If needed, tie in-session rewards to deep breathing practice as well.

NOTE: it is important that relaxation strategies are practiced **AT THE END OF THE SESSION**. Relaxation strategies are meant to help the child feel relaxed, which may result in fewer tics. Given that it is important for tics to occur during awareness practice, we want to make sure that relaxation strategies do not prevent tics from occurring during this session.

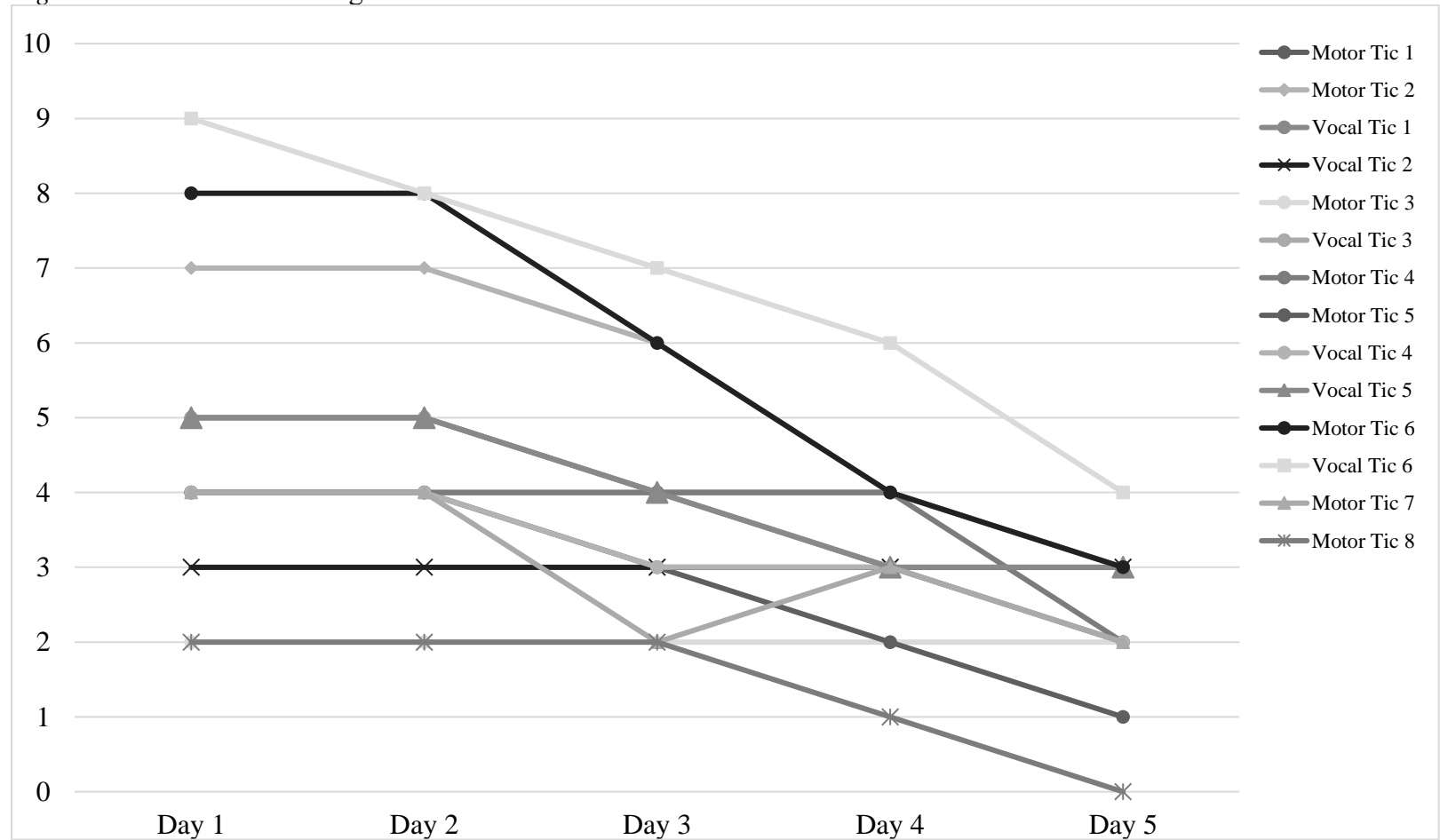
Figure 4. Peer Support Activity (Questions in a Jar)

Instructions: Prior to the session, create at least 10 questions to put inside a jar. Questions should ask about each child's experience with tics and how they handle certain situations. Go around the room and ask each child to pick a question, one at a time. The child who selects the question will answer first, but the other children will also have an opportunity to share their perspectives on that question. Move around the room until all questions have been asked. Sample questions are listed below:

1. **Have you ever told your friend about your tics? How did it go?**
2. **Have you shared this with a teacher? How did that go?**
3. **What do you do if you have tics in class and other people notice?**
4. **Do you parents ever say stop it to tics? How does that feel?**
5. **Has another child ever said something to you about your tics? What did you tell them?**
6. **Let's pretend a child who has tics is getting bullied at school because of their tics. What would you tell them to do? How can that child respond?**
7. **What is one thing from the past week that made you feel proud of yourself?**
8. **What strategies have you learned so far that might help you if you're feeling nervous?**
9. **What would you want others to know about you that has nothing to do with your tics?**
10. **What is something helpful a friend can say when you have a tic?**
11. **What could you say to someone if they don't know about tics?**



Figure 5. Mike's SUDs Rating Scores



NOTE. Mike did not attend post or 1-month follow-up assessments.

Figure 6. Kevin's SUDs Rating Scores

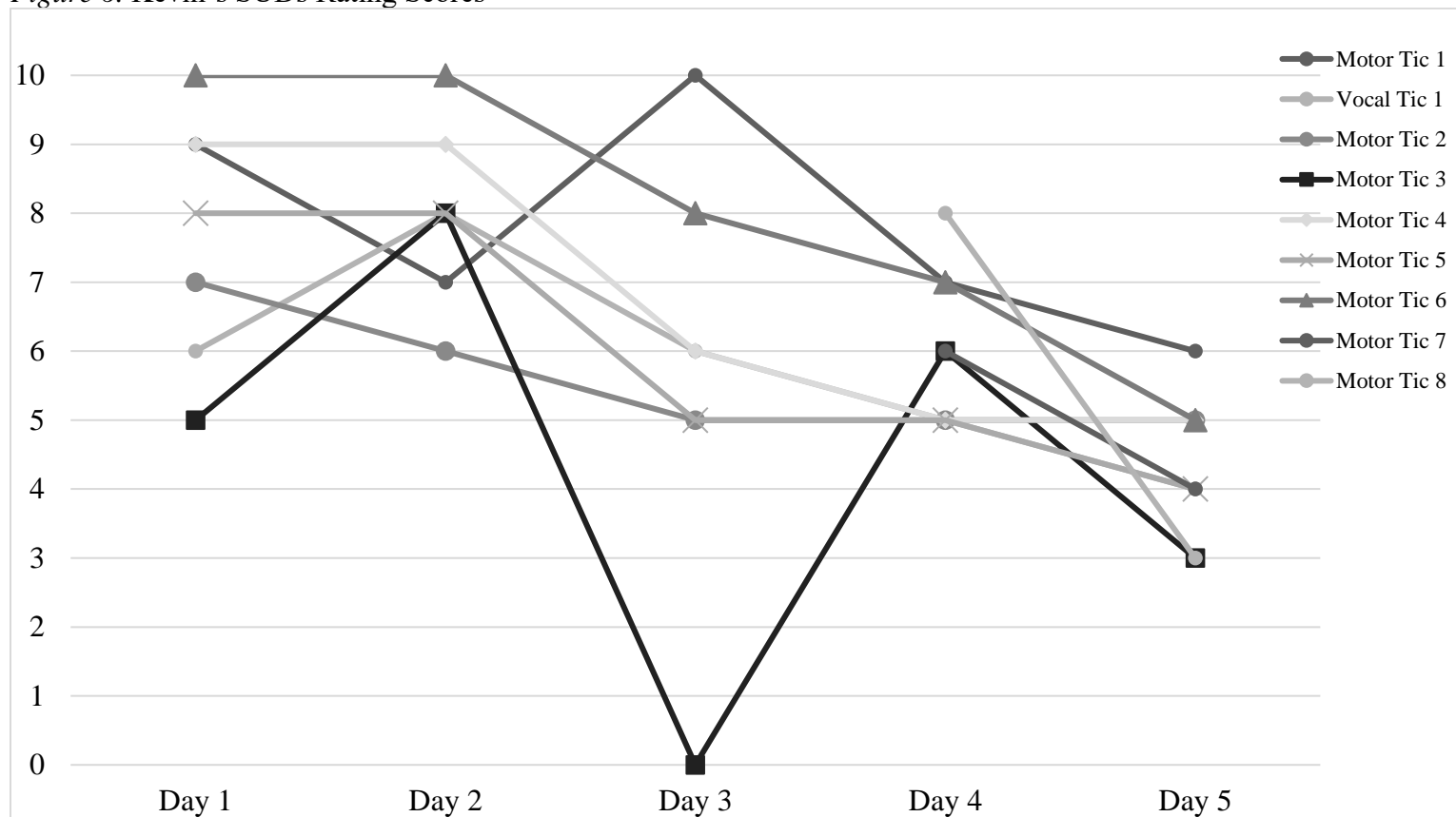


Figure 7. Henry's SUDs Rating Scores

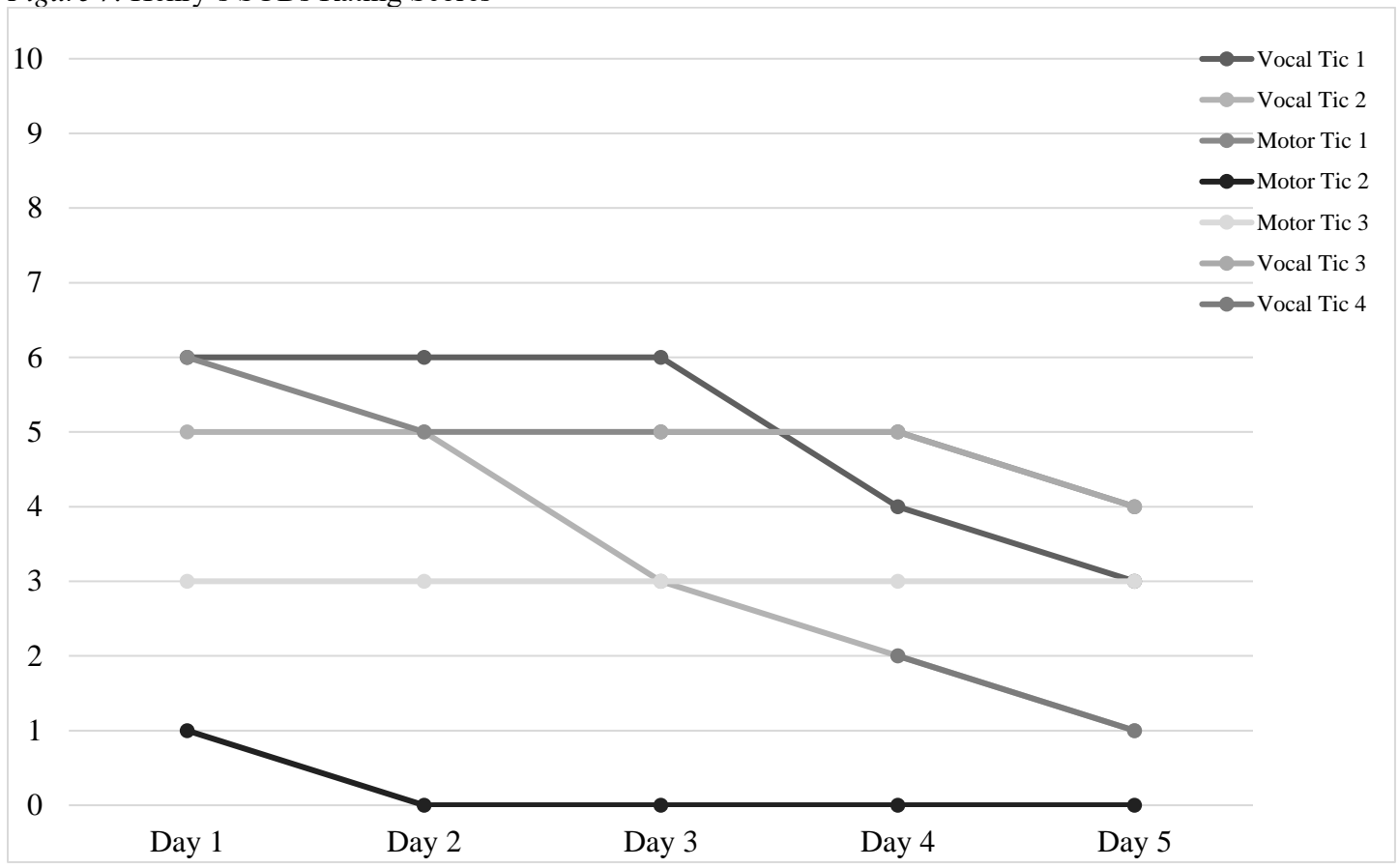
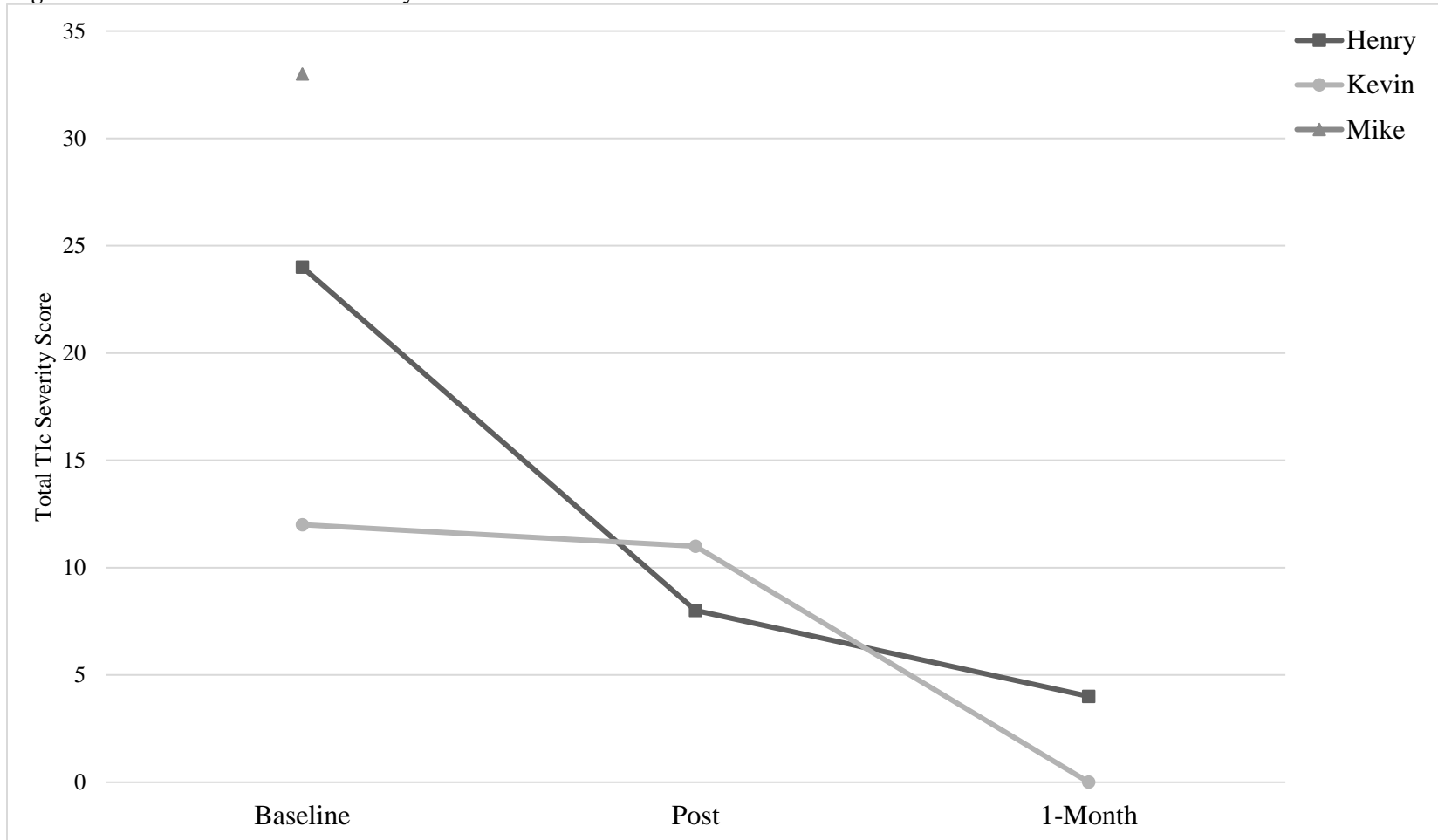
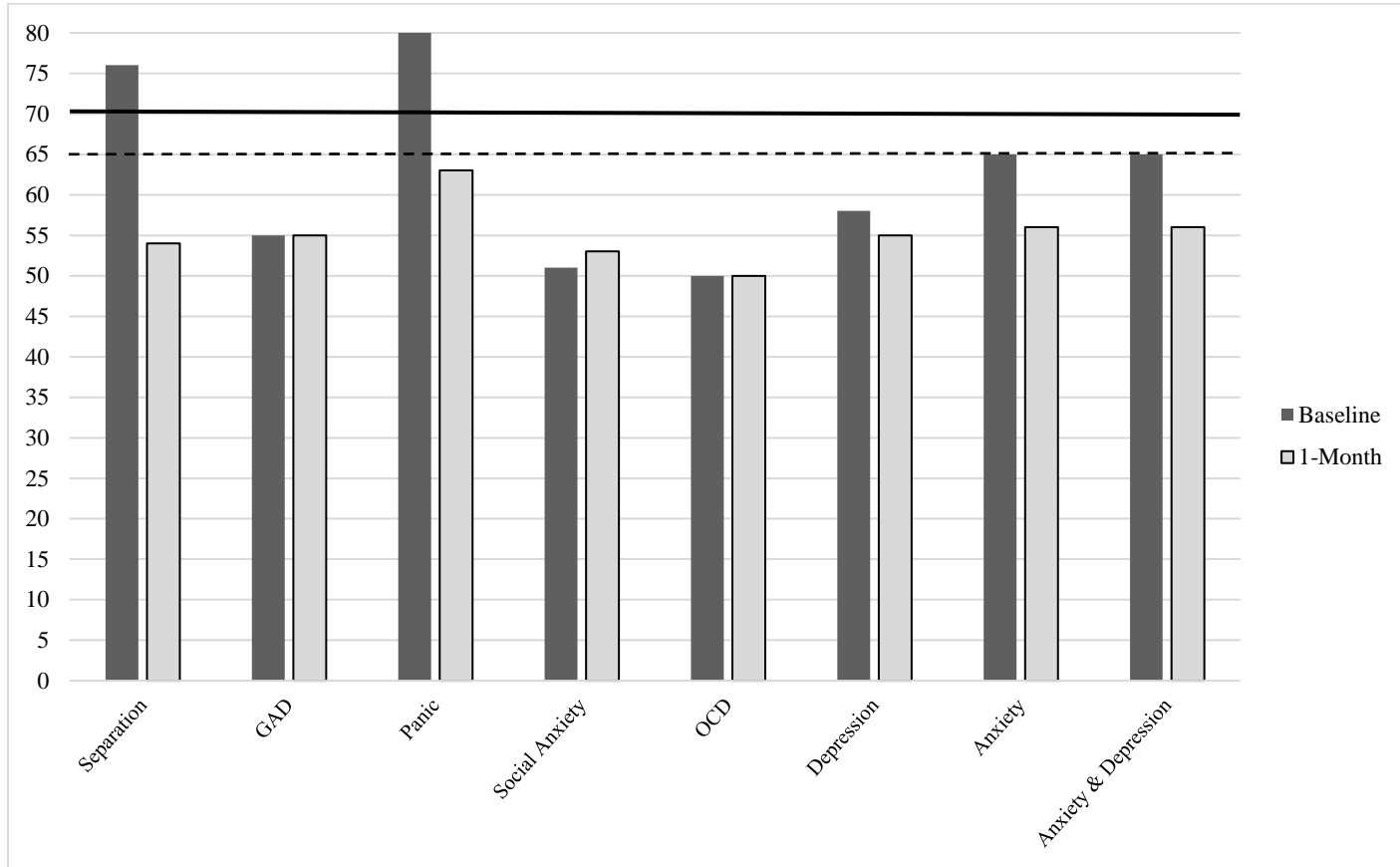


Figure 8. YGTSS Total Tic Severity Score



NOTE. Mike did not attend post or 1-month follow-up assessments.

Figure 9. Kevin Parent-Report Revised Child Anxiety and Depression Scale (RCADS) Baseline and 1-Month Post T-Scores



NOTE. The dashed line indicates a borderline clinical threshold (T-score 65). The solid line indicates the clinical threshold (T-score 70).

Table 1. Sample Taming Tics Together Schedule

Session Type	Time/Duration	Monday	Tuesday	Wednesday	Thursday	Friday
Child Group Sessions	9:00-10:00 AM	Introductions and Rules	CBT Triangle & Identifying Emotions	Problem Solving Basics and Practice	Questions in a Jar	Creating CRs as a group (Relapse Prevention)
		Psychoeducation				
		Intro to Tic Hierarchy & Awareness Training	Movement (Scavenger Hunt)		Movement (Activity Stations)	Certificate Ceremony (Individual clinicians join)
Break, Preparation and Supervision	10:30 - 11:30 AM	Clinician Group Supervision	Clinician Group Supervision	Clinician Group Supervision	Clinician Group Supervision	Clinician Group Supervision
	11:30 AM - 12:00 PM	<i>Individual Session Transition/ Flex Time</i>	<i>Individual Session Transition/ Flex Time</i>	<i>Individual Session Transition/ Flex Time</i>	<i>Individual Session Transition/ Flex Time</i>	<i>Individual Session Transition / Flex Time</i>
	12:00 - 1:00 PM	<i>Lunch</i>	<i>Lunch</i>	<i>Lunch</i>	<i>Lunch</i>	<i>Lunch</i>
Individual Sessions	2 hours*	Introduce in-session reward and brainstorm 1-2 rewards	Comorbidity-specific content	Comorbidity-specific content	Comorbidity-specific content	Comorbidity-specific content
		Hierarchy & Awareness	Hierarchy, Awareness, and CR Practice for 1 st Tic	Hierarchy, Awareness, and CR Practice for 2 nd Tic	Hierarchy, Awareness, and CR Practice for 3 rd Tic	Hierarchy, Awareness, and CR Practice for 4 th Tic
		Relaxation: Deep Breathing	Relaxation: PMR	Relaxation: Mindful Eating	Relaxation: Body Scan	Review
						Relaxation: Coping Self-statements
Caregiver Group Sessions	6:00-7:30 PM	Introductions, Psychoeducation, Awareness Training, & Rewards	Introduction to CRs, Praise, & Planned Ignoring	Behavioral Chart & Rewards	Commands & Consequences and Caregiver Well-being	Review, Relapse Prevention, and School

NOTE. CR = competing response. CBT = Cognitive Behavioral Therapy. PMR = Progressive Muscle Relaxation *Sessions can occur any time between 11:30 AM and 5:00 PM. Sessions may occur in one 2-hour block or shorter, separate blocks.

Table 2. Participant Demographics and Overview

	<u>Mike</u>	<u>Kevin</u>	<u>Henry</u>
Age	9	9	11
Race (Ethnicity)	White (non-Hispanic)	Black (non-Hispanic)	White (Hispanic)
Diagnostic Profile	ADHD, Combined Tourette’s Disorder	Unspecified Anxiety Disorder Tourette’s Disorder	ADHD, Combined Tourette’s Disorder
School Format	Home School	Remote Schooling (COVID-19 Related)	Not reported
Medication (during treatment)	None	Clonidine	Adderall
Prior Treatment	No formal treatment; prior use of incentive systems	Cognitive Behavioral Therapy, Relaxation Strategies	None
Family Involvement in Treatment	Mother	Mother and Father	Mother and Father
Individual Session Format	One 90-minute session/day	Two 60-minute sessions/day	One 90-minute session/day

Table 3. Number of Tics Endorsed and Number of Corresponding Competing Responses Created During Treatment

Name	Number of Motor Tics Endorsed	Number of Vocal Tics Endorsed	Number of Tics Addressed
Mike	8	6	4
Kevin	8	1	5
Henry	3	4	4

NOTE: The number of tics and competing responses created correspond to the 5-days of the intensive program.

IV. STUDY 3: RANDOMIZED CONTROLLED TRIAL

INTRODUCTION

Tic disorders (TDs) are neurodevelopmental disorders characterized by sudden and involuntary motor movements (i.e., eye blinking, shoulder shrugging) and/or phonic (i.e., throat clearing, sniffing) tics and include several DSM-5 diagnoses, such as Tourette's Disorder, persistent (chronic) motor or vocal tic disorder, and provisional tic disorder (American Psychiatric Association [APA], 2013), among others. It is estimated that TDs impact between 1 and 3% of children (Robertson et al., 2017), and transient tics appear to impact up to 20% of children (Scahill et al. 2014). It is believed that tics generally have an onset between ages four and seven (Specht et al., 2011; Wolicki et al., 2019) with common estimates noting males experience symptoms at approximately four times the rate of females (e.g., Mol Debes et al., 2008; Roessner et al., 2007).

TDs often have a significant impact on youths' overall functioning. Importantly, they can have adverse effects on youths' functioning across several domains (i.e., social, familial, and academic functioning; Kurlan et al., 2001; Roessner et al., 2011; Specht et al., 2011), and lead to quality-of-life impairment (Dale et al., 2021, see Study 1; Storch, Lack et al., 2007). In a recent survey conducted by the Tourette Association of America [TAA] Impact Survey Working Group (2018), many youth noted that they experience some form of discrimination as a result of their TD symptoms, such as bullying or removal from an extracurricular activity or their classroom. Moreover, recent studies by Lavenstein and colleagues (2016) and Malek (2022) have suggested between 15 to 60 percent of children with TDs have also reported pain as a result of their tics. Overall, TDs likely impact children's and their family's daily activities, functioning, and quality of life.

Additionally, an overwhelming majority of children with a TD experience at least one co-occurring mental health diagnosis. Over 60% of children with a TD will also meet criteria for attention-deficit/hyperactivity disorder (ADHD; Centers for Disease Control and Prevention [CDC], 2009), half exhibit elevated anxiety symptoms, and a least one-third experience clinically significant obsessive-compulsive spectrum symptoms, with youth exhibiting symptoms of a TD, ADHD, and obsessive-compulsive disorder (OCD) simultaneously being referred to as experiencing the “Tourette [Disorder] Triad” (Kurlan, 2010). Estimates of co-occurring diagnoses are higher for youth diagnosed with Tourette's Disorder, indicating between 80 and 90% of children experience at least one co-occurring diagnosis (Hirschtritt et al., 2015; Wolicki et al., 2019) In fact, it has been suggested that individuals with a TD and a co-occurring diagnosis reported lower quality of life and interference in social relationships (Eapen et al., 2016). For example, it has been noted that co-occurring TDs and ADHD in childhood can result in more impairment than TDs alone (i.e., Lebowitz et al., 2012), such that these children are more prone to angry outbursts and other disruptive behaviors, as well as greater social impairment (Sukhodolsky et al., 2003). Parents of children with TDs and co-occurring diagnoses are also likely to experience increased caregiver stress or strain, with some studies attributing elevations to the co-occurring diagnosis-related symptoms (such as ADHD and OCD; i.e., Stewart et al., 2015), some to the TD-related symptoms and perceived social support (Schoeder & Remer, 2007) and some noting significant familial impact related to the combination TD and co-occurring diagnosis presentation (Wilkinson et al., 2001) as compared to families with a child exhibiting a TD alone. Further, Vermilion and colleagues (2020) reported parents of children with TDs noted lower quality of life as

compared to families without a child experiencing a TD. Taken together, both TD and co-occurring symptoms can have a significant impact on the family's overall quality of life and well-being. Given the increased likelihood of children with TDs to also meet criteria for ADHD, OCD, a disruptive behavior disorder, and/or anxiety or mood disorders, it is important that symptoms associated with these common co-occurring diagnoses be concurrently addressed during treatment, particularly if these symptoms are causing significant impairment.

COMPREHENSIVE BEHAVIORAL INTERVENTION FOR TICS

Several randomized controlled trials (RCTs) have indicated that Comprehensive Behavioral Intervention for Tics (CBIT) is effective in managing and/or reducing tic symptoms (e.g., Himle et al., 2012; Piacentini et al., 2010). CBIT is typically offered in a one-to-one, in-person format and involves several stages to treatment completion (i.e., Psychoeducation, Awareness Training, Habit Reversal Training, Relapse Prevention, and Relaxation). A recent meta-analysis found that individuals with a TD who engaged in a behavioral intervention, such as CBIT, were more likely to benefit from treatment, as compared to control interventions (McGuire et al., 2014). Further, results from this review indicated comparable treatment effects to those found with medication use (McGuire et al., 2014); however, reported minimal associated risk. Importantly, Wolicki and colleagues (2020) found that 84.4% of youth in their study who were being treated with medication, for example, aripiprazole, haloperidol, or pimozide, to manage their tics experienced side effects, resulting in approximately half of families discontinuing the medication. As such, the American Academy of Neurology (AAN; Pringsheim et al., 2019) recommends CBIT as a first-line intervention for TDs.

CBIT was originally designed to be conducted across roughly eight to 10-weeks, with each weekly session lasting approximately one hour. Despite substantial support for CBIT, significant issues remain that reduce the reach and impact of this intervention. First, there are a limited number of providers trained in this specialized, evidence-based treatment modality (Woods et al., 2010), therefore, obtaining a provider trained in CBIT is often an arduous task (Bhikram et al., 2021; Flancbaum et al., 2011; Pringsheim et al., 2019). Second, highly co-occurring disorders are often not explicitly addressed or targeted within the traditional intervention protocol (Coffey, 2015). As a result, families are often required to attend separate intervention sessions to address each category of symptoms (e.g., CBIT provider for TDs, Cognitive Behavioral Therapy (CBT) provider for anxiety), increasing the cost, duration of treatment, and time spent traveling to multiple providers. Therefore, an alternative treatment format that provides increased access to CBIT and that incorporates treatment strategies for both TDs and co-occurring diagnoses is necessary.

The use of internet-based (i.e., videoconferencing) interventions predates the COVID-19 pandemic and has shown preliminary efficacy. For example, Himle and colleagues (2010; 2012) evaluated a videoconference-based CBIT, initially within a case-series presentation (N=3), and then with a follow-up randomized pilot study (N=20). Results noted that scores from the videoconference on the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) were comparable to the initial pediatric CBIT trial (Piacentini et al., 2010) and families indicated acceptability with the videoconference format. Nonetheless, these studies did not explicitly address co-occurring symptomatology, and the follow-up study required families to participate within their

university setting; thus, comparison to an at-home telehealth format is limited. Further, Ricketts and colleagues (2016) evaluated an internet-based CBIT format with 20 youth, resulting in comparable reductions in tic severity in Piacentini and colleagues' (2010) initial CBIT randomized controlled trial.

Tichelper.com, an online, self-guided CBIT-based intervention, provides an alternative treatment format for families who are unable to access CBIT providers. This format allows families to complete online modules independently and at their own pace. Nonetheless, Tichelper.com may be less beneficial for more complex cases and does not address common comorbidities within its curriculum (Conelea & Wellen, 2018). Importantly, this format does not provide access to a live clinician, which may not be sufficient for some families. In addition, Andr n and colleagues (2019) also found that a semi self-guided online intervention (i.e., the clinician was available to answer questions via written message and phone) was an acceptable and feasible alternative to in-person Exposure with Response Prevention (ERP) or Habit Reversal Training (HRT) for families. While youth were eligible to participate if they experienced a co-occurring diagnosis, these symptoms, again, were not addressed in treatment. Most recently, Rachamim and colleagues (2020) found that a primarily parent-guided, internet-based CBIT intervention was feasible and reported improvements in tic severity and impairment. Importantly, additional studies have been conducted using internet-based CBIT protocols in adults, which show promise for this format (Haas et al., 2022; Jakubovski et al, 2016); however, many have not yet been evaluated within a pediatric sample.

Evaluation of group-based intervention formats has also begun to show promise. For example, Heijerman-Holtgreffe and colleagues (2020) evaluated an intensive, group-based ERP protocol for 14 youth ($M_{age}=11.69$ years) and with results indicating a 16% reduction in total tic scores on the YGTSS. While improvements in tic-related symptomatology were noted, the reductions indicated in this study are significantly lower than those reported in studies using CBIT intervention strategies. While group-based CBIT has begun to show preliminary evidence to support this format, most studies have incorporated weekly sessions, did not explicitly address common co-occurring diagnosis symptoms, and provided limited individualized support outside of traditional or standard CBIT strategies (i.e., individual competing responses created within the context of a group; Yates et al., 2016; Zimmerman-Brenner et al., 2021).

Non-traditional or alternative delivery formats, such as intensive interventions, are well-established, particularly as an intervention modality for several of the common co-occurring diagnoses of TDs. Importantly, intensive intervention formats have been shown to be a cost-efficient modality for providing multimodal interventions (i.e., Ehrenreich & Santucci, 2009; Fabiano et al., 2014;). Intensive modalities provide the option for children and families to access treatment for a finite time period, limiting the burden often associated with attending weekly treatment sessions. Recently, there has been increasing evidence to support intensive, group-based interventions, particularly for diagnoses that require specialized interventions that are not often widely or easily accessible. For example, Graziano et al. (2020) evaluated an intensive behavioral parent training (BPT) treatment format to address disruptive behavior disorders. BPT is a well-established and evidence-based behavioral intervention for ADHD and other disruptive

behavior disorders (for reviews, see Evans et al., 2018; Fabiano et al., 2009; Pelham & Fabiano, 2008). Results from this study found that intensive BPT was as effective as standard weekly BPT sessions. Further and notably, parents who experience high levels of stress were more likely to benefit from the abbreviated format than from the traditional BPT intervention modality. Additionally, Cornacchio and colleagues' (2019) evaluation of an intensive, group-based format for children with selective mutism was associated with high caregiver-reported treatment satisfaction, few reported barriers to treatment from caregivers, and clinically significant reductions in symptoms two months following the end of treatment. Storch and colleagues (2007) also found that individual, intensive CBT to treat pediatric OCD was as effective as weekly sessions, reporting comparable improvements three months following the end of treatment.

Overall, intensive intervention modalities are effective across a multitude of pediatric mental health illnesses, and this intervention format can likely be extended to CBIT as a mechanism to target TDs, co-occurring diagnosis, as well as quality of life and familial (e.g., parental stress, treatment maintenance) outcomes. While initial support for an intensive form of CBIT has begun to emerge, the literature is very limited. Blount and colleagues (2018) conducted a case series (N=5) study evaluating a brief, four-day intensive intervention, which resulted in tic-related improvements at the end of treatment. However, the population evaluated consisted of adults with TDs. To our knowledge, only three studies to date have evaluated the use of an intensive CBIT intervention format within an exclusively pediatric population. Blount and colleagues (2014) conducted a preliminary case series, predominately focused on pre-adolescents, utilizing a four-day intensive intervention. This study showed initial support for the utility of intensive CBIT

for reducing tic symptoms; however, the sample size (N=2) likely limits the generalizability. Kennedy et al., (2016) assessed the effectiveness of conducting eight sessions across two consecutive weeks with 10 youth ($M_{age} = 13.7$ years old). Results indicated significant improvements in the YGTSS-assessed total tic severity following treatment.

Taken together, preliminary evidence for the utilization of intensive, internet, and group-based CBIT is promising. Nonetheless, current intervention formats often do not include components explicitly targeting common co-occurring diagnoses, despite high comorbidity rates with TDs (Coffey, 2015) and the well-known impact of both symptom sets on overall functioning. An integral next step is to evaluate an intensive intervention modality combining CBIT and evidence-based interventions for co-occurring symptomatology. This format may lead to a more rapid reduction in symptoms and a more suitable format for families of children experiencing TDs and co-occurring diagnoses, while also providing increased access to evidence-based interventions that are often geographically limited. The objective of this study is to evaluate the Taming Tics Together (TTT) treatment protocol using a randomized controlled trial design with an active (immediate) treatment group (I-TTT) and a 1-month waitlist control group (WL) using block randomization with a ratio of 2:1. Specifically, this study aims to (1) evaluate 1-month tic symptom trajectories and (2) evaluate 1-month co-occurring diagnosis symptoms, parental stress, and the overall impact on families' well-being. Further, the study aims to evaluate the overall satisfaction and feasibility of the current treatment protocol format following the completion of TTT. We hypothesize that families randomized to the I-TTT group will demonstrate (1) decreases in duration, frequency,

intensity, and severity of TDs, (2) maintenance of treatment gains through continued use of strategies learned throughout the intervention, and (3) will report reductions in tic-related interference in their global functioning and quality of life as compared to the WL group. Regarding co-occurring symptomatology, we hypothesize that the I-TTT group will demonstrate decreases in presenting co-occurring diagnosis symptom severity. Finally, we hypothesize that the I-TTT group will demonstrate (1) reductions in caregiver stress and (2) reductions in each family's well-being compared to the WL group at the 1-month follow-up.

OVERVIEW OF TAMING TICS TOGETHER

The TTT protocol was developed by Chelsea Dale, M.S., and Justin Parent, Ph.D., based on initial case study evaluations within an individual format (Dale et al., 2021, see Study 1) and a small group-based format (Dale et al., under review, see Study 2). Feedback from families and study clinicians was incorporated into subsequent studies to improve the treatment protocol and adapt the intervention to meet the needs of families (for details on prior case studies, see Dale et al., 2021(Study 1) and Dale et al., under review (Study 2)). TTT builds upon existing and well-established evidence-based interventions for TDs (e.g., Woods et al., 2008), and co-occurring disorders (e.g., BPT, Forehand et al., 2012; group-based CBT, Silverman et al., 1999). All interventions were adapted to be used within an intensive intervention context (for additional information, please see Dale et al., under review, see Study 2).

In its current format, TTT is a five consecutive day intensive intervention that can be conducted entirely via remote technologies (i.e., videoconference platforms). In the current study, HIPAA-compliant Zoom videoconferencing software was used for all

sessions. TTT offers four treatment components: (1) caregiver-only groups, (2) child-only groups, (3) individual child sessions, and (4) one optional 30-minute booster session at the 1-month follow-up. All child group-based strategies involve a TD-related component (i.e., psychoeducation about TDs, awareness, and habit-reversal training overview), transdiagnostic skills for co-occurring symptoms (i.e., emotion identification, problem-solving), and peer support content related to living with and managing tics (see Figure 1 for an example). All caregiver group content involves TD-specific skills and opportunities to practice using skills (i.e., practice creating competing responses for a variety of common tics), but also includes content related to co-occurring symptoms (i.e., differential attention, communication, emotion coaching) and caregiver support topics to help their children manage their TD and co-occurring diagnoses (i.e., talking to your child's school about diagnoses). Individual sessions are used to address each child's unique symptom set and create a "toolbox" of skills specifically designed to help the individual child (see Figure 2 for examples of group and individual session content).

METHODOLOGY

Participants and Recruitment

After receiving Institutional Review Board approval (IRB-20-0562), families who were currently on the Child and Family Well-being Clinic's (CFW) clinical waitlist were informed of the possibility of participating in a research study. Additionally, materials were sent to pediatricians, neurologists, and TAA chapters and support groups via email. Information regarding the program was also advertised through online formats (i.e., Facebook). Interested caregivers contacted the study center and spoke with a study team member, at which time the study was described in detail. If families indicated an interest

in participating in the study, a phone screen was conducted to determine preliminary eligibility. If families were deemed preliminarily eligible, they were then scheduled for an informed consent and screening appointment to determine eligibility and enroll. Families were eligible if (1) the child was between the ages of 7 and 17, (2) the child meet criteria for a Tic Disorder diagnosis, and (3) the family was available to participate in all sessions (i.e., child and at least one caregiver was available to attend all sessions), (4) the family spoke English, and (5) the child was experiencing elevations in co-occurring symptomatology as determined by at least subclinical elevations on intake assessment surveys or borderline clinical scores on diagnostic interview detailed below. Families were excluded if, (1) they were identified as having a psychiatric condition that was more impairing and requiring treatment before intervening on the TD, and (2) if a child met criteria for Autism Spectrum Disorder. These exclusion criteria were selected as these participants may benefit more from other services prior to enrollment in a group-based CBIT treatment. Figure 3 provides a CONSORT diagram detailing participant recruitment, participation, and retention at the 1-month follow-up assessment.

Twenty-one families were randomized to either the I-TTT group (n=14) or a 1-month waitlist (n=7). Two families did not participate in the intervention (reasons for not participating included: COVID-19-related interference and inability to contact to schedule sessions); however, both families completed the initial baseline assessment and were, thus, included in the final sample for analysis. One family required additional intervention sessions following the end of the five-day intervention. Given that their treatment protocol differed from the study protocol, the family was discontinued from study follow-up assessments. The final sample consisted of 20 children (13 I-TTT, 7 WL)

and at least one caregiver ($M_{childage}=12.54$ years; 50% male; 40.0% Hispanic/Latinx). In the current sample, 75.0% met criteria for Tourette Disorder, and 50% exhibited subclinical elevations in two or more co-occurring diagnoses. Caregivers reported that 60.0% were currently taking medication (i.e., over-the-counter, Psychoactive). It was indicated that 25.9% of families reported their household income fell below \$75,000 (median household income within the United States in 2020 was \$67,521 (U.S. Census Bureau, 2021)). Additionally, 75% of caregivers held at least a bachelor's degree (see Table 1 for complete demographic details). No differences among variables were noted between groups, with the exception of caregivers within the WL group were more likely to report holding a part-time employment position than parents in the I-TTT group, $\chi^2(1, N=20) 4.13, p=.042$ (see Table 2).

Procedure

Participants in both groups received the same intervention protocol; the only difference between each group was the timing in which they received the intervention. Families assigned to the WL group were offered enrollment into the next treatment group after one month on the waitlist. Specific assessments were completed at the following timepoints: baseline (prior to treatment), immediately post-treatment, and one month following the end of treatment. One primary caregiver completed parent- and self-report measures at each time point. Treatment acceptability and satisfaction measures were completed immediately following the end of treatment. Additionally, clinician-administered assessments were conducted at the baseline and 1-month follow-up (see Table 3 for a complete schedule of assessments).

Treatment groups were divided by age (i.e., 7- to 12-year-olds, 13- to 17-year-olds) to ensure content was provided at an age-appropriate level. Both groups completed all measures at the baseline assessment before starting the intervention, a post-treatment assessment immediately following the end of treatment, and an assessment 1-month following the end of treatment. Families assigned to the WL group completed the 1-month assessment twice (once prior to beginning treatment to determine any changes in symptoms and once one month after participating in TTT). Children were compensated with a \$10 electronic gift card for their participation in treatment; caregivers were offered a \$30 electronic gift card for completing assessments at all time points. Families were also offered an optional 30-minute booster session after completing the 1-month assessment. Intervention sessions, including the booster session, were offered free of charge.

Measures

Demographics and Caregiver Factors

Demographics. Caregivers completed a demographic form regarding their own race, ethnicity, education, marital status, and household income. Caregivers also reported on their child's demographic information.

Parental well-being. The Perceived Stress Scale (PSS; Cohen et al., 1983) is a 10-item self-report measure used to evaluate an individual's perception of stress, or to the extent that events in an individual's life are perceived as stressful. Items are rated on a 4-point Likert scale (0 = *Never* to 4 = *Very Often*). All items are summed to create a total perceived stress score (range 0-40), with higher scores indicating higher perceived stress. The alpha coefficient at baseline was .90. The Patient Health Questionnaire-4 (PHQ-4;

Kroenke et al., 2009) is a brief screening measure evaluating anxiety and depression symptoms. Items are rated on a 4-point Likert scale (0 = *Not at all*, 3= *Nearly every day*) and consist of two items related to anxiety and two items related to depression. Scores of three or greater on the depression and/or anxiety subscales indicate elevations within these domains and warrant further assessment. In the current study, caregivers completed the measure to briefly evaluate caregiver psychopathology within the internalizing symptom domains. The Total Score alpha coefficient was .90 at baseline.

Tic Disorder-Related Measures

The Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) is a semi-structured clinician-administered interview to evaluate past and current tic symptoms and is considered the gold standard for assessing tics in children and adolescents (Martino et al., 2017). The YGTSS is used to evaluate the frequency, intensity, complexity, and number, as well as the interference of motor and vocal tics, using a 6-point scale (range 0 to 5). An impairment score is also collected (range 0 to 50). In the current study, the YGTSS was collected at the baseline assessment and the 1-month follow-up, and the Total Tic Severity and Impairment scores were used as primary outcomes; however, the Global YGTSS score, which is a sum score of the YGTSS Total Tic Severity and YGTSS Impairment scores, was also analyzed. The YGTSS has been shown to be a reliable assessment of tics in children and adolescents (Storch et al., 2005).

The Premonitory Urge for Tics Scale (PUTS; Woods et al., 2005) is a 10-item self-report measure used to evaluate the severity of premonitory urges in children and adolescents with TDs. Nine items are used to calculate an overall severity score (range 9-36) and one item regarding the ability to stop or control tics. Scores above 31 indicate

extremely high intensity and likely severe impairment of the premonitory urge for tics. Items are endorsed on a four-point Likert Scale (1 = *not at all* to 4 = *very much*). The PUTS has been found to be correlated with tic severity as assessed on the YGTSS and shows high internal consistency for children ages 10 and older (Woods et al., 2005). In the current study, children ages 10 and older completed the PUTS at the baseline assessment and were used to aid clinicians in determining the child's current awareness of premonitory urges. The Subjective Units of Discomfort (SUDs) rating, as outlined in the treatment manual from Woods and colleagues (2008), was used to evaluate the amount of discomfort/distress associated with each current tic. Youth rate the level of discomfort/distress each tic causes on a scale from 0 (not present or not causing discomfort/distress) to 10 (causing significant discomfort/distress). While youth were allowed to endorse and rate as many tics as they were currently experiencing, a maximum of 10 tics were evaluated per participant to ensure that time in session was spent on treatment content. SUDs ratings were collected on each day of the five-day program (see Figures 4a and 4b for examples). When clinically indicated, the review of SUDs ratings during sessions was suspended.

Co-occurring Symptom Measures

The Kiddie Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL; Kaufman et al., 2000) is a well-established clinician-administered assessment used to conduct diagnostic evaluations. In the current study, the K-SADS-PL was used to determine diagnostic symptom presentations of all co-occurring diagnoses excluding in-depth evaluation of OCD symptoms, which were assessed using the Child Yale-Brown Obsessive-Compulsive

Scale (CY-BOCS; Scahill et al., 1997). The CY-BOCS is a clinician-administered assessment evaluating the severity of obsessions and compulsions, evaluating symptoms across five dimensions (time occupied by symptoms, interference of symptoms, distress, resistance, and degree of perceived control over symptoms). Overall severity scores range from 0 to 40, with scores between 14 to 24 falling within the moderate range, 25 to 30 within the moderate-severe range, while scores of 30 or more are considered severe (Lewin et al., 2014). Only caregivers and youth who endorsed obsessive-compulsive symptoms on the K-SADS-PL at baseline were administered the CY-BOCS at the baseline assessment. Information from the K-SADS-PL and CY-BOCS was used to inform individual session treatment planning.

The Revised Children's Anxiety and Depression Scale (RCADS; Chorpita et al., 2000) was used to evaluate broad child internalizing symptomatology. The RCADS is a 47-item youth- and caregiver-report, which encompasses six subscales (separation anxiety disorder, social phobia, generalized anxiety disorder, panic disorder, major depressive disorder, and obsessive-compulsive disorder). Informants endorse items on a scale from 0 (Never) to 3 (Always). The RCADS has shown good reliability within the context of a clinical sample (Chorpita et al., 2005). In the current study, the Total Anxiety and Depression subscales were used as secondary internalizing symptom outcomes. The alpha coefficient for complete data at baseline for the Depression subscale was .75. The Anxiety subscale alpha coefficient for complete data was .91 at baseline.

Externalizing symptomatology was assessed using the caregiver-report version of the Disruptive Behavior Disorder Rating Scale (DBD-RS; Pelham et al., 1992). The DBD is a 42-item questionnaire assessing symptoms across conduct (16 items), oppositional

defiant (8 items), and attention-deficit/hyperactivity disorders (9 inattention items and 9 hyperactivity/impulsivity items). Items are rated from 0 (not at all) to 3 (very much). In the current study, a raw score sum was calculated to determine changes from baseline to the 1-month follow-up. The alpha coefficients at baseline for Inattention, Hyperactivity/Impulsivity, and ODD raw sum scores were .76, .87, and .91, respectively.

Child Quality of Life and Functioning

To evaluate health-related quality of life across settings, caregivers completed the Pediatric Quality of Life Inventory, General Core Scales (PedsQL; Varni et al., 2001). Items are rated on a 5-point Likert scale from 0 (Never) to 4 (Almost Always). Items are then reverse scored and transformed to a scale from 0-100, with higher scores indicating better quality of life. The PedsQL incorporates several primary subscales, including evaluations of Physical, Emotional, Social, and School Functioning, as well as a secondary Psychosocial Health Summary score, which incorporates the Emotional, Social, and School Functioning scales. In the current study, only the Physical Functioning and School Functioning subscales were evaluated using caregiver ratings. Alpha coefficients for the Physical and School Functioning subscales at baseline were .74, and .74, respectively. The Tic Accommodation and Reaction Scale (TARS; Capriotti et al., 2015) is a 35-item questionnaire evaluating social reactions and accommodations to a child's tics across settings (i.e., at home, at school, in public settings). Items are rated on a 4-point Likert scale from 0 (not at all in the past week) to 3 (many times in the past week). Items can be summed to create three domain subscale scores: Attention, Aversive, and Escape. Additionally, a Total Score is calculated by summing all items within the scale. In the current study, the Total Score was used to evaluate tic-related

accommodation and reactions. The alpha coefficient for the Total Score was .84 at baseline. Due to the low reliability, the Aversive scale was removed from analyses. In the current study, caregivers independently completed the TARS.

Each child's overall global functioning and symptom severity were assessed using the independent evaluator-rated Children's Global Assessment Scale (CGAS; Shaffer et al., 1983) and the Clinical Global Impressions Scale-Severity (CGI-S; Guy & Bonato; 1970). The CGAS is rated by the independent evaluator on a scale from 0 to 100, with higher scores indicating greater functioning and less overall impairment. The CGI-S is rated on a 7-point scale from 1 (normal) to 7 (among the most extremely ill patients). The independent evaluator completed the CGI-S and the CGAS following the 1-month assessment. Additionally, parents were asked at the 1-month timepoint to report on any changes to their child's treatment (i.e., starting or stopping medication, starting or stopping behavioral interventions) regimen over the previous month.

Process Measures

The Burden of Treatment Participation (BTP) Scale evaluates the overall cost associated with participating in treatment. The BTP includes 10 items rated from 0 (strongly disagree) to 4 (strongly agree). Given that the intervention was offered free of charge, one item (i.e., "The cost of the treatment program was too high") was omitted to prevent artificial inflation of scores. Therefore, scores in the current study ranged from 0 to 36, with high scores indicating a greater burden associated with participating in the intervention. The BTP scale was administered immediately following the end of TTT.

The Treatment Acceptability Questionnaire (TAQ; Hunsley, 1992) is a 6-item assessment evaluating reactions to treatment. In the current study, caregivers were asked

to complete the measure, which assesses items such as the overall acceptability of treatment, side effects, the effectiveness of treatment, ethics of treatment, as well as the knowledge and trustworthiness of the clinicians. Items are rated using a 7-point Likert scale, resulting in a total score range of seven to 42, with higher scores indicating greater treatment acceptability. Items were modified slightly for the delivery to parents following the end of treatment. This measure has been utilized effectively in prior technology-based CBIT adaptation studies (Ricketts et al., 2016; Himle et al., 2012). The Client Satisfaction Questionnaire (CSQ; Larsen et al., 1979) was utilized to evaluate overall caregiver-reported satisfaction with the treatment program. The CSQ is an 8-item measure, in which items are rated on a 4-point Likert scale (total scores may range from 8 to 32), with higher scores indicating greater satisfaction. Caregivers were also provided with the opportunity to provide qualitative feedback (i.e., “Was the child group helpful?” “Was the parent group helpful?”) on the treatment in general. The TAQ and CSQ were completed immediately following the last day of TTT.

Treatment Integrity

At the conclusion of each treatment day, the clinicians who completed the individual session and the group leaders completed a daily session note, which included the duration of the session and content reviewed within the session. All clinicians attended daily supervision sessions with the Ph.D.-level licensed clinician, and treatment plans were reviewed prior to each session. Additionally, supervisors were available to join sessions with the primary clinician as needed. Given the flexible nature of the treatment protocol, guidelines for session progression were offered to clinicians within the TTT manual and broadly encompassed the inclusion of at least one tic-related skill

and one co-occurring diagnosis symptom-related skill. Finally, attendance was taken at each session. Families were considered present at the caregiver-only group meetings if at least one caregiver attended. All other sessions required the child's presence to be marked in attendance.

Data Analytic Plan

Analyses were conducted using the Statistical Package for Social Sciences, version 27 (SPSS, 2020), and R statistical software (R Core Team, 2012) lavaan package (Rosseel, 2012). For group difference analyses, Full Information Maximum Likelihood (FIML) estimation was conducted to account for missing data within an intent-to-treatment model, where possible. For primary analyses, we employed linear regression modeling to examine differences in tic symptoms and co-occurring symptom trajectories from the initial intake assessment to one month following the end of the intensive treatment. Hedges' g is utilized to evaluate the effect size of treatment on primary and secondary outcomes due to differing sample sizes within each group. Additionally, the Reliable Change Index (RCI; Jacobson & Truax, 1991), was used to evaluate clinically-significant change in TD symptoms. RCIs, which suggests that a value of greater than or equal to 1.96 indicates the effects attributable to treatment are not due to chance and are reliable. Given that interrater reliability within our study was not available, the reliability data from Storch and colleagues (2005) was used to calculate the RCIs for the YGTSS Total Tic Severity Score. Further, the reliability data from McGuire and colleagues (2018) was used to calculate the RCIs for the Global YGTSS. evaluate clinically significant changes in co-occurring symptom trajectories, we used the Reliable Change Index First, RCIs were created across all co-occurring outcomes (i.e., RCADS and DBD).

Next, the RCI from the most impairing symptom set for each youth participant, based on parent report, was then aggregated into a primary co-occurring symptom severity RCI outcome to determine the overall clinical significance of treatment on the most severe co-occurring outcomes. RCIs were calculated for secondary outcomes on complete data only. Finally, exploratory outcomes within the quality-of-life domain (i.e., PedsQL, TARS) were evaluated using a linear regression model on the subscales indicated above.

RESULTS

Sample Symptom Presentation.

Participants were equally distributed across groups by sex (six males and seven females within the I-TTT group, four males and three females within the WL group). Overall, each participant in the current sample met criteria for a TD, and exhibited elevations and/or subclinical symptoms on at least one co-occurring diagnosis (see Table 4). Three participants met criteria for a provisional tic disorder, and all were female. Only one participant met for a chronic motor tic disorder (male participant); all remaining participants met criteria for Tourette's Disorder. The average ages of reported onset of motor and vocal tic symptoms were approximately age 6 and age 8, respectively. Caregivers of female participants reported a slightly higher mean age onset of motor and vocal tics (approximately age 7 and age 9, respectively) compared to caregivers of male participants (approximately age 6 and age 8, respectively). The I-TTT and WL groups reported the same age of onset for motor tics (approximately age 6); however, the WL group reported a slightly older age of onset for vocal tics (approximately age 9) compared to the I-TTT group (approximately age 8). Additionally, the I-TTT group reported higher scores across all YGTSS outcomes at baseline. At the 1-month follow-up, no families

within the complete sample reported changes to treatment in any form (i.e., changes to medication, modifications to behavioral intervention) between the final day of treatment and the 1-month follow-up assessment.

All female participants endorsed elevations in internalizing symptoms, compared to 70% of male participants, whereas 70% of male participants endorsed externalizing symptoms compared to only 30% of female participants. Across both groups, roughly the same percentage of participants endorsed elevations in internalizing symptoms in the immediate treatment (84.62%) and waitlist group (85.71%). Importantly, a higher percentage of participants in the I-TTT group endorsed externalizing symptoms, OCD, and ADHD (53.85%, 30.77%, and 53.85%, respectively) compared to the WL group (42.86%, 14.29%, and 42.86, respectively). Given that teacher-report was not collected, new diagnoses of ADHD were not offered; however, prior diagnoses were noted.

Primary Outcomes

Effect sizes using Hedges' g on primary YGTSS outcomes were calculated using completed data at the 1-month follow-up for between-group evaluations. Additionally, effect sizes evaluating within I-TTT-group changes from baseline to the 1-month follow-up (see Table 5 for all effect size calculations). Hedges' g was selected due to the difference sample sizes of the immediate treatment and waitlist groups. Examining within-group effects, there was an improvement on all of the primary YGTSS outcomes for the I-TTT group, ranging from small to medium effect sizes. Improvements were also noted within the WL group on the YGTSS-I and Global YGTSS scores; however, increases in scores on the YGTSS Total Tic Severity were observed.

When comparing the effect of treatment between groups, the difference between the I-TTT and WL groups on YGTSS Impairment scores was small, indicating greater improvement in the WL group at the 1-month follow-up. Group differences on the YGTSS Global score (.22) were small at the 1-month follow-up, indicating lower scores overall within the WL group than in the I-TTT group. All other subscales had a very small effect size. Notably, however, the I-TTT group exhibited a larger average point decrease across all YGTSS subscales as compared to the WL (see Table 6). Additionally, the I-TTT group's scores across all YGTSS subscales (i.e., Impairment, Total Tic Severity) at baseline were higher than those of the WL. Differences between the I-TTT group on the YGTSS Impairment and the Global YGTSS scale were nonsignificant. However, scores on the YGTSS Total Tic Severity indicated marginal significance ($b = -4.731$, $SE = 2.72$, $p = .08$), such that allocation to the I-TTT group was associated with decreases in YGTSS Total Tic Severity scores at the 1-month follow-up compared to the control condition (see Figures 5 and 6). Importantly, 30% of participants in the I-TTT group ($N=10$) dropped one clinical severity category (i.e., from marked severity to moderate severity; see McGuire, Piacentini, and colleagues (2021) for categorical cutoff scores). At the 1-month follow-up, 30% of participants (3 out of 10 participants) within the I-TTT group who completed the assessment appointment indicated clinically significant improvements on at least one tic symptom outcome, compared to 16.7% ($n=1$) of the WL group.

Within-group evaluations of the I-TTT group were also conducted to assess possible differences in outcome by age and sex (see Table 7). The effect of age on the YGTSS Total Tic Severity scores was marginally significant ($b = 6.79$, $p = .082$), such

that older age was associated with higher YGTSS Total Tic Severity at the 1-month follow-up. The effect of age on the YGTSS Impairment score, however, was significant ($b = 14.20, p < .05$), such that older age was associated with more impairment at the 1-month follow-up assessment. Differences in the Global YGTSS scores by age were nonsignificant. The effect of sex on YGTSS Total Tic Severity scores was also significant ($b = 6.85, p < .05$), such that female sex was also associated with higher scores at follow-up. Differences in all other YGTSS outcomes were nonsignificant. Additionally, the average number of tics reported on tic hierarchies during individual sessions was 9.5 ($SD = 6.79$), with an average of 5.58 ($SD=3.06$) motor and 3.92 ($SD=5.37$) vocal tics endorsed throughout the five days of TTT.

Secondary Outcomes

Examining effect sizes within the I-TTT group on the RCADS indicated a small (.46) effect on depressive symptoms and a medium (.72) effect on anxiety symptoms compared to a small (.22) and large (.96) effect on anxiety and depressive symptoms within the WL group. The effect of treatment on ADHD-Hyperactive/Impulsive symptoms was small within the I-TTT group and was very small for all other outcomes. Effect sizes for externalizing symptoms within the WL group indicated worsening of symptoms, with a small negative effect for ADHD-Hyperactive/Impulsive symptoms and a very small effect for all other outcomes (see Table 5).

The effect of the group on total raw scores on ADHD-Hyperactivity/Impulsivity items was marginally significant ($b = -1.61, p = .06$), such that the I-TTT group was associated with fewer symptoms. All other outcomes were not significantly different at the 1-month follow-up (see Figures 7 through 11). Within-group evaluations of the I-TTT

group were also conducted to assess possible differences in co-occurring symptom outcomes by age and sex. The effect of age on total raw scores on ODD items was significant ($b=-2.02$, $p= 0.56$), such that older age was associated with fewer ODD symptoms at follow-up. All other co-occurring diagnosis symptoms were nonsignificant by age. The effect of sex on RCADS depression outcomes was significant ($b = 7.71$, $p <.01$), such that females were more likely to experience depressive symptoms than males at 1-month follow-up. All other co-occurring diagnoses were not significantly different by sex. Additionally, 43% of participants within the I-TTT group (3/7) reported clinically significant improvements on at least one secondary outcome ($RCI >1.96$) and 14.3% ($n=1$) of participants reported a negative change on a secondary outcome. Due to low retention rates on secondary outcomes based on caregiver reports, only complete data were used to calculate effect sizes. The effect size for the RCADS Anxiety and Depression outcomes were medium (.40) and small (.20) and the DBD ODD, ADHD-Hyperactivity/Impulsivity outcomes were medium (.44 and .33, respectively), indicating improvement in the I-TTT group. The effect size for ADHD-Inattention was large (.70), however, and corresponded with WL improvement.

Of the families within the I-TTT group who completed the 1-month follow-up assessment (10 out of 13), 45.45% indicated an improvement in their CGI-S score, whereas 27.27% indicate no change; 18.18% ($n=2$) noted an increase in severity at the 1-month follow up. However, 100% of participants noted improvement on the CGAS, with an average change of 21.4 points, indicating improvement in their child's global functioning.

Exploratory Outcomes

Additional outcomes evaluating the impact of tics were conducted. There were no significant differences reported on the subscales of the TARS. Additionally, marginally significant differences were reported on the PedsQL School Functioning subscale, such that parents in the I-TTT group reported higher school functioning at the 1-month follow-up as compared to the WL group. No differences were found on the Physical Functioning subscale. Regarding exploratory parent-related outcomes, no significant differences were found in parental stress or parental internalizing symptoms between groups (see Table 8).

Treatment Acceptability and Feasibility

The average caregiver rating for overall treatment acceptability on the TAQ was 37 (SD= 6.46), indicating high treatment acceptability overall. These findings were also consistent with prior videoconference-based CBIT studies (i.e., Himle et al., 2012). Caregiver treatment satisfaction ratings on the CSQ were also high, with a mean rating of 28.6 (SD=3.53, range = 21-32). The average overall costs and burdens endorsed on the BTP was 2 (SD=2.98), with only two parents reporting needing to take time off (range 30 to 60 minutes per day) from work to participate in TTT. Within the I-TTT group, 100% attendance was met at each of the three daily treatment sessions (child group, parent group, and individual sessions). Overall, 46.2% of participants within the I-TTT group (6/13) participated in the optional 30-minute booster session following the one-month assessment. No correlation was found between booster attendance and either the TAQ ($r = 0.39, p = 0.26$) or CSQ ($r = 0.30, p = 0.40$).

DISCUSSION

Summary of Current Study

The current study evaluates the feasibility, acceptability, and preliminary efficacy of Taming Tics Together, a novel, five-day telehealth-based intensive intervention for youth with TDs and co-occurring symptomatology, using an RCT design comparing an immediate treatment vs. a 1-month waitlist control group. Results suggest trending significance on improvements in tic-related symptomatology, namely tic symptom severity and some improvement in co-occurring child symptomatology. Overall, caregivers reported high satisfaction and acceptability of treatment, and the costs and burdens associated with treatment were low. Further, an extremely high (100%) treatment attendance rate across all session formats was attained.

The current study found mean YGTSS Total Tic Severity score reductions of 4.53 points at the 1-month follow-up within the I-TTT group. This point reduction is slightly below those reported by Piacentini and colleagues (2010; 7.6-point reduction) and other internet-based interventions, such as Himle and colleagues (2012; 7.8-point reduction), Ricketts and colleagues (2016; 6.5-point reduction), and Rachamim and colleagues (2020; 6.6-point reduction), all which entailed weekly sessions across at least eight weeks. Importantly, the post-assessment YGTSS for the aforementioned four studies was conducted immediately following the end of the treatment protocol, whereas the current study's post-assessment YGTSS was conducted one month following the end of treatment, therefore, comparisons may be limited.

Recent literature, however, suggests that an emphasis on changes in tic symptoms alone may not be sufficient to account for the overall impact of TDs on quality of life and

impairment (i.e., Bloch & Leckman, 2009; Conelea et al., 2013; McGuire et al., 2015). Therefore, a crucial next step is to evaluate and integrate intervention strategies to best meet the diverse needs of children and families experiencing TDs. Notably, previous studies have indicated that youth with disruptive behaviors or anxiety experience more difficulties engaging with treatment content (i.e., Himle et al., 2012), particularly when the intervention is delivered via videoconference. These studies often have not explicitly aimed to address co-occurring symptoms or implemented significant modifications to manage such behaviors within the context of treatment. TTT incorporates a rapid reinforcement system (point system) tailored to meet the needs of each group as well as each individual child's needs. On the first day of each group, the point system is introduced, with points awarded for attendance, participation, and engagement with session content. Once competing responses are established within the individual sessions, children are awarded points for (1) noticing they had a tic (such as by raising a finger to indicate to the group leaders they noticed a tic) or for attempts to use a competing response. Caregivers are informed of the use of the point system, and small prizes (i.e., small games, gift cards, extra screen time, or special activities) are discussed within the first caregiver session. Additionally, a tiered reward system may be implemented as needed, per the individual clinician's judgment. Further, specific or labeled praise (i.e., "Great job catching that tic!", "Thank you so much for staying on camera.," or "I love how you raised your hand before answering!") is used throughout the child group and individual treatment sessions. Thus, additional emphasis on managing co-occurring or related symptomatology is important in reducing the overall impairment of TDs.

In the current study, 43% of participants who completed the 1-month follow-up noted clinically significant improvements in co-occurring diagnosis symptoms. Further, a small effect of treatment was noted on internalizing symptoms, as well as ODD and ADHD-hyperactive/impulsive symptoms within the I-TTT group. Findings also suggested an increase in inattention symptoms at the 1-month follow-up for the I-TTT; however, this is possibly a reflection of tic symptom awareness and/or impairment of tic-related symptoms, rather than ADHD symptoms. For example, attempts to utilize skills learned during treatment, such as managing tics and/or managing co-occurring symptoms, may present as ADHD-like inattention or distraction (Erenberg, 2005; McGuire et al., 2014), however, would likely be captured as ADHD symptomatology on our current screening measures. Overall, the findings from the current study suggest that, in part, co-occurring symptomatology can be effectively managed and addressed concurrently with TD-related symptoms. Nonetheless, several important differences in treatment outcomes were identified in participant characteristics that warrant further discussion.

In our current sample, differences based on biological sex on TD-related outcomes were identified, such that females may have fewer improvements in an abbreviated treatment format. The literature on the impact of sex on TD symptoms is limited and somewhat inconsistent (i.e., Garris & Quiggs, 2021; Lichter & Finnegan, 2015); however, possible differences in the trajectory of tic symptoms in girls may highlight the need for alternative or adapted treatments to address their presenting concerns. Garris and Quiggs' (2021) review suggested females appear to have a later onset of tics, a later peak tic symptom severity, are more likely to experience more

complex tics, and experience more psychosocial impairment related to their tics. Importantly, Garris and Quiggs (2021) found that females have a higher chance of tic severity increasing into adulthood and lower chances of tic remission into adulthood, suggesting a higher likelihood of chronic symptom presentations. During the COVID-19 pandemic, a notable increase in more rapid-onset tics emerged predominately in females (Radhakrishnan et al., 2022). Our current findings support the notion of possible sex differences in TD symptom expression, as within-group analyses of the I-TTT group noted higher motor tic interference and total tic severity scores in female than male participants. Garris and Quiggs (2021) also noted that females are more likely to experience comorbid OCD and internalizing symptoms and are less likely to experience ADHD symptoms compared to their male counterparts. The sample in the current study was somewhat consistent with this finding, as 70% of males had a prior diagnosis of ADHD or exhibited elevations (compared to 30% of females). Additionally, in our current sample, females were more likely to endorse elevations in depressive symptomatology than males.

Overall, given that tic and co-occurring symptoms outcomes for adolescent females appear to be poorer than for their male counterparts, in addition to the possible negative impact of COVID-19-related stressors, adaptations to interventions for this population are necessary and further evaluation is warranted. While some adolescent females have experienced a less common or “traditional” onset of symptoms (i.e., rapid onset in adolescence), treatment recommendations still include the use of CBIT, psychoeducation, and CBT, with an additional emphasis on contextual factors that may be contributing to symptom severity (McGuire, Bennett, et al., 2021; Pringsheim et al.,

2021). Given that stress has also been associated with increases in the severity of both TD and co-occurring symptoms (Ruhrman et al., 2021), emphasis on increasing access to evidence-based intervention is integral. As such, possible important adaptations to TTT may be warranted, particularly for adolescent females with a later onset of tics. For example, adolescent females may benefit from additional time spent in treatment, such as extending TTT to two weeks, or may benefit from breaks in treatment sessions to allow for at-home practice (i.e., two consecutive sessions, followed by a day off at home, then two sessions again, followed by the weekend, etc.). Further, continued tailoring of treatment to meet the family's needs and simultaneously reduce the distress associated with the rapid onset of tics is integral to treatment success.

The current study was conducted within the context of the COVID-19 pandemic, at a time when varying degrees of restrictions and lockdowns were in place, and many elementary, middle, and high schools were in the midst of determining the educational format for the upcoming school year. The COVID-19 pandemic has also undoubtedly increased mental health concerns in children and adolescents and has limited access to traditional, in-person treatment formats (i.e., Comer, 2021). Further, a recent review indicated that adolescents with prior mental health concerns may be even more vulnerable during the pandemic due to a variety of risk factors (i.e., changes to care, difficulties related to isolation/confinement, anxiety or worries related to COVID-19; Guessoum et al., 2020). For example, a recent study conducted in Italy indicated that over half of youth participants reported changes in the severity of their tics during the COVID-19 lockdown, and was associated with more difficulties with remote learning (Termine et al., 2022). As such, consultation with the child's or adolescent's school may be of

additional benefit, as impairment across social and academic functioning may be alleviated by providing support across settings. According to a recent report released by the CDC (Radhakrishnan et al., 2022), overall mental health condition visits to emergency departments increased for the majority of children and adolescents during the COVID-19 pandemic; however, adolescent females (ages 12-17) experienced the largest increases in the proportion of visits for TDs in 2020, 2021 and 2022. This likely supports the notion that the pandemic's impact on the mental health of youth may be especially strong and interfering for teenage females.

Given the possible differences in onset of tic symptoms and trajectories, we also explored if TTT was more or less effective based on child age. In the current study, younger children in the I-TTT group noted lower total tic severity scores and less tic-related impairment than older participants at the 1-month follow-up. These improvements may be related to less time for reinforcement of the premonitory urge and tic expression cycle, coupled with external factors, such as reactions and/or accommodations from peers, caregivers, or teachers. Additionally, younger children in the current study appeared to follow a more "traditional" tic symptom onset, and most had not yet experienced several complex and/or severe tics. As such, strategies used within treatment, for example, competing responses, paired with reinforcement systems, or interventions that are delivered in a more developmentally appropriate manner (i.e., Bennett et al., 2020) may be more effective for younger children. Evaluation of CBIT in younger populations has been shown to be preliminarily effective and may provide skills and strategies to children and parents to understand and manage impairing tics (Bennett et al., 2020). Despite our knowledge that the age of worst tic is often reported between ages 7

and 15 (see Leckman et al., 2006), limited studies have been conducted evaluating the efficacy of early interventions in younger children. Nonetheless, early intervention may aid in reducing tic-related impairment and possible parental distress associated with their child's tics. Further, given that younger children may experience tics for a shorter duration when they first present to treatment, these differences in improvement may highlight the impact and importance of early intervention, and may suggest that a briefer intervention format may be more suitable for younger children.

Access to providers with the ability to offer both CBIT and interventions for common co-occurring diagnoses is limited. As such, TTT's intensive format affords the unique opportunity for families to access treatment in an abbreviated manner which, in turn, likely significantly reduces accessibility barriers, may eliminate the need for children to miss school (if conducted during the summertime), and reduce cost (i.e., travel costs, time off from work) through a videoconference platform. In the current study, families received at least one and a half hours of individual intervention each day, in addition to two and a half hours of group-based intervention per day, for a minimum of 20 hours of intervention across the five days of treatment. This is consistent with the previous case series evaluation of TTT (Dale et al., under review, see Study 2) and is comparable to the average amount of time spent in standard, weekly CBIT, and co-occurring diagnosis sessions when they are conducted separately. As such, a large dose of treatment provided over a brief period of time may be more suitable and feasible for some families. Additionally, individual sessions were conducted at any time throughout the day (outside of the time groups were scheduled) and groups were offered during school summer vacation (between June and August), which likely provides the flexibility

to accommodate families' schedules. As such, this study provides preliminary support that TTT is a possible alternative to in-person or other standard treatment modalities, may reduce traditional barriers to evidence-based intervention, and warrants further assessment.

Limitations

The current study is not without limitations. First, the current study only evaluated a 1-month follow-up period. Given the waxing and waning nature of tics, as well as the time often required to implement, incorporate, and effectively use the strategies consistently that were learned throughout treatment, additional and/or longer follow-up periods (i.e., 3, 6, 9, and/or 12 months) would be beneficial to determine longer-term outcomes. Additionally, results should be viewed with caution when compared to the initial CBIT study (Piacentini et al., 2010) given the different follow-up time points (i.e., an assessment was conducted immediately following the end of treatment). This is key as outcomes immediately following the end of treatment compared to one month later (with no support from a clinician or option for booster sessions in between) likely differ and possibly underrepresent improvements made immediately after treatment. Second, YGTSS assessments were not conducted immediately following the end of treatment due to staff limitations and to ensure that independent evaluators remained masked to participants' treatment condition. Future studies may benefit from additional assessments immediately following the end of treatment. Third, the current study allowed for one optional, 30-minute booster session following the completion of the 1-month follow-up. In the initial CBIT study (Piacentini et al., 2010), participants were offered three additional monthly booster sessions, which may have contributed to improvements.

Future studies should evaluate the use of additional booster sessions following the immediate end of treatment (i.e., two-week check-in meeting) to determine their utility, as it is possible some youth may require more than one week of intensive intervention and would benefit from more frequent and/or additional follow up. Further evaluation for whom a one-week compared to a longer intensive intervention format is most effective is also needed. Fourth, the current study sample was small (N=20) and, therefore, may not be representative of the U.S. population of youth and families with TDs. As such, findings may not be generalizable to the population at large, and further, larger-scale studies are warranted. However, preliminary findings indicated marginal significance on TD-related outcomes, suggesting the possibility of moderate improvements associated with treatment when evaluating TTT in a larger sample.

Additionally, the sample was recruited within the context of the COVID-19 pandemic, at which time, most intervention services were conducted via videoconference. While high satisfaction and acceptability of treatment were noted, suggesting that the treatment platform may be suitable for use, future studies would benefit from the inclusion of a technology-specific assessment regarding attitudes towards videoconference-based intervention to further elucidate the acceptability of this treatment format. Fifth, 1-month assessments were conducted around the beginning of the school year, at a time when worries and stress are often elevated. These concerns may also have been exacerbated by the COVID-19 pandemic, as many youth were returning to school in person for the first time in over one year, or were uncertain of their school's educational format for the upcoming school year. Future studies should consider the use of an additional assessment at the "school year follow-up," that is, an assessment conducted

when the school year was underway (i.e., end of October/early November), as this may allow for more time to adjust to using skills learned in treatment within the context of school, public, and social settings. Sixth, the WL control group scores across all YGTSS outcomes were less severe than those of the I-TTT group (see Table 7). Given the small sample size, this may, in part, impact between-group findings and may not truly reflect the possible impact of treatment. Therefore, a larger sample is likely needed to further elucidate group differences and the overall effect of treatment. Seventh, the current study only enrolled families that spoke English due to limited bilingual supervision and clinicians. Future studies should consider offering TTT in other languages, as well as carefully considering effective translation and adaptations to the treatment protocol, as a possible mechanism to improve access to care for families who do not speak English fluently. Eighth, formal treatment fidelity is not available for the current study. Future studies would benefit from recording sessions and tracking fidelity formally using a coding system.

Finally, retention on the secondary, caregiver-report outcomes was low. The current study consisted of a two-part assessment, which involved a videoconference visit to conduct clinician-administered assessments, and then an email with a link to caregiver-reported questionnaires. Low response rates on these measures at the 1-month assessment may be, in part, due to the remote and independent format of questionnaire completion, in contrast to in-person visits where forms may be completed during the visit and study staff members can quickly review forms for any missing items.

Conclusions

Notwithstanding its limitations, the current study has several strengths, including preliminary pilot data supporting the feasibility, acceptability, and utility of TTT to address TD and common co-occurring diagnosis symptoms in an intensive format. The videoconference and intensive format may provide families with an opportunity to focus their full attention on strategies taught within each session. Given its abbreviated nature, intensive treatment formats may aid in limiting or reducing the impact of weekly responsibilities by impacting session attendance, and likely reduce the burden of and time lost due to travel required for in-person/in-clinic treatment. Importantly, videoconference-based interventions may provide a more naturalistic setting for in vivo practice of skills for youth, and in-home feedback for parenting skill implementation, particularly in the context of the COVID-19 pandemic. Additionally, the group-based format may offer both the caregivers and their children the opportunity to learn from others with similar experiences, gain support from each other, and give youth exposure to practicing using skills within the context of a virtual group, which became a particularly important and unforeseen benefit as a result of remote learning during the pandemic. Importantly, the group format also allows more families at a time to access a specialty treatment provider.

This study provides pilot data to preliminarily support the use of TTT, particularly for younger children, but also has the unique flexibility to be adapted to better fit the needs of children and adolescents presenting with TDs. For example, females, especially those exhibiting a more rapid onset of tics, may benefit from additional support and a longer duration of treatment. Future iterations may consider evaluating the utility of

extending TTT into a two-week format, which may allow not only for additional practice utilizing skills but also an extended period of time to generalize skills and more effectively problem solve using skills when more severe or intense symptoms are present. Alternatively, additional brief booster sessions at a higher frequency, such as one time per week for the first month, may also suffice to provide additional support and continued practice, without committing to an additional week of treatment. Nonetheless, the current study sample was small, and further assessment of the current protocol within a larger RCT is also warranted.

TTT provides the opportunity for a person-centered approach to effectively tailor treatment in order to manage each family's unique presenting concerns, as well as aid children and their caregivers in building a supportive network of families with similar experiences. TTT is well-positioned to be able to provide alternative avenues to accessing CBIT and treatment for co-occurring symptoms, which have become increasingly more needed as a result of the COVID-19 pandemic and its associated stressors. TDs and co-occurring diagnoses are prevalent among youth in the United States and can lead to significant impairment across domains. Despite an increase in the number of families receiving CBIT over the past decade (i.e., 6% to 36% of youth between 2008 and 2018; TAA 2018), access to trained providers remains limited. While initial evidence suggests that telehealth intensive combined interventions may be an effective treatment modality to simultaneously address tic and co-occurring symptoms, further large-scale randomized controlled trial studies are needed to determine their effectiveness, feasibility, and overall acceptability. In particular, studies evaluating for whom this treatment is most effective are integral to determining treatments that are suitable for each family. Nonetheless, TTT,

an intensive, group- and internet-based CBIT presents a promising treatment modality that is likely to increase access to treatment, be more cost-effective, and improve the quality of life for youth with tics and common co-occurring diagnoses.

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Figure 1. Peer Support Activity (Questions in a Jar)

Instructions: Prior to the session, create at least 10 questions to put inside a jar. Questions should ask about each child's experience with tics and how they handle certain situations. Go around the room and ask each child to pick a question, one at a time. The child who selects the question will answer first, but the other children will also have an opportunity to share their perspectives on that question. Move around the room until all questions have been asked. Sample questions are listed below:

12. **Have you ever told your friend about your tics? How did it go?**
13. **Have you shared this with a teacher? How did that go?**
14. **What do you do if you have tics in class and other people notice?**
15. **Do your parents ever say stop it to tics? How does that feel?**
16. **Has another child ever said something to you about your tics? What did you tell them?**
17. **Let's pretend a child who has tics is getting bullied at school because of their tics. What would you tell them to do? How can that child respond?**
18. **What is one thing from the past week that made you feel proud of yourself?**
19. **What strategies have you learned so far that might help you if you're feeling nervous?**
20. **What would you want others to know about you that has nothing to do with your tics?**
21. **What is something helpful a friend can say when you have a tic?**
22. **What could you say to someone if they don't know about tics?**



Figure 2. Sample Schedule

	<u>Day 1</u>	<u>Day 2</u>	<u>Day 3</u>	<u>Day 4</u>	<u>Day 5</u>
Child Group	<ol style="list-style-type: none"> 1. Introductions and Rules 2. Psychoeducation 3. Introduction to Tic Hierarchies & Awareness 	<ol style="list-style-type: none"> 1. CBT Triangle 2. Identifying Emotions 	<ol style="list-style-type: none"> 1. Problem Solving 	<ol style="list-style-type: none"> 1. Questions in a Jar 2. Activity Stations 	<ol style="list-style-type: none"> 3. Relapse Prevention 4. Review
Parent Group - Child	<ol style="list-style-type: none"> 1. Introductions and Rules 2. Psychoeducation and Awareness Training 3. Brief Overview of Rewards 	<ol style="list-style-type: none"> 1. Praise 2. Planned Ignoring/Ignoring reassurance-seeking 3. Intro to Competing Responses & Awareness Training 	<ol style="list-style-type: none"> 1. Identifying Physiological Sensations/ Cues for Anxiety 2. Sensational Exposure Introduction 	<ol style="list-style-type: none"> 1. Parental Self-Care 2. Detective Thinking/Flexible Thinking 3. Thinking Traps 	<ol style="list-style-type: none"> 1. Relapse Prevention 2. Review
Teen Group	<ol style="list-style-type: none"> 1. Introductions and Rules 2. Psychoeducation 4. Introduction to Tic Hierarchies & Awareness 	<ol style="list-style-type: none"> 1. Cognitive Triangle 2. Behavioral Activation 3. Values 	<ol style="list-style-type: none"> 1. Questions in a Jar 	<ol style="list-style-type: none"> 1. Effective Communication 	<ol style="list-style-type: none"> 1. Relapse Prevention 2. Review
Parent Group - Teen	<ol style="list-style-type: none"> 1. Introductions and Rules 2. Psychoeducation & Awareness Training 3. Overview of Rewards 	<ol style="list-style-type: none"> 1. Cognitive Triangle 2. Behavioral Activation 3. Parental Self-Care 4. Praise 	<ol style="list-style-type: none"> 1. Emotional Behaviors 2. Detective Thinking 3. Thinking Traps 	<ol style="list-style-type: none"> 1. Effective Communication 	<ol style="list-style-type: none"> 1. Relapse Prevention 2. Review
Individual Session	<ol style="list-style-type: none"> 1. Rapport Building 2. Tic Hierarchy/SUDs 3. Awareness Training 4. Psychoeducation for Co-occurring Diagnosis 5. Relaxation: Deep Breathing 	<ol style="list-style-type: none"> 1. Tic Hierarchy/SUDs 2. Awareness Training (Tic 1) 3. Competing Response Creation (Tic 1) 4. Co-occurring Diagnosis Hierarchy 5. Relaxation: Progressive Muscle Relaxation 	<ol style="list-style-type: none"> 1. Tic Hierarchy/SUDs 2. Awareness Training (Tic 2) 3. Competing Response Creation (Tic 2) & Practice 4. Exposure for Co-occurring Diagnosis 5. Relaxation: Body Scanning 	<ol style="list-style-type: none"> 1. Tic Hierarchy/SUDs 2. Awareness Training (Tic 3) 3. Competing Response Creation (Tic 3) & Practice 4. Exposure for Co-occurring Diagnosis 	<ol style="list-style-type: none"> 1. Relapse Prevention 2. Review 3. Relaxation: Coping Thoughts

Figure 3. CONSORT Diagram

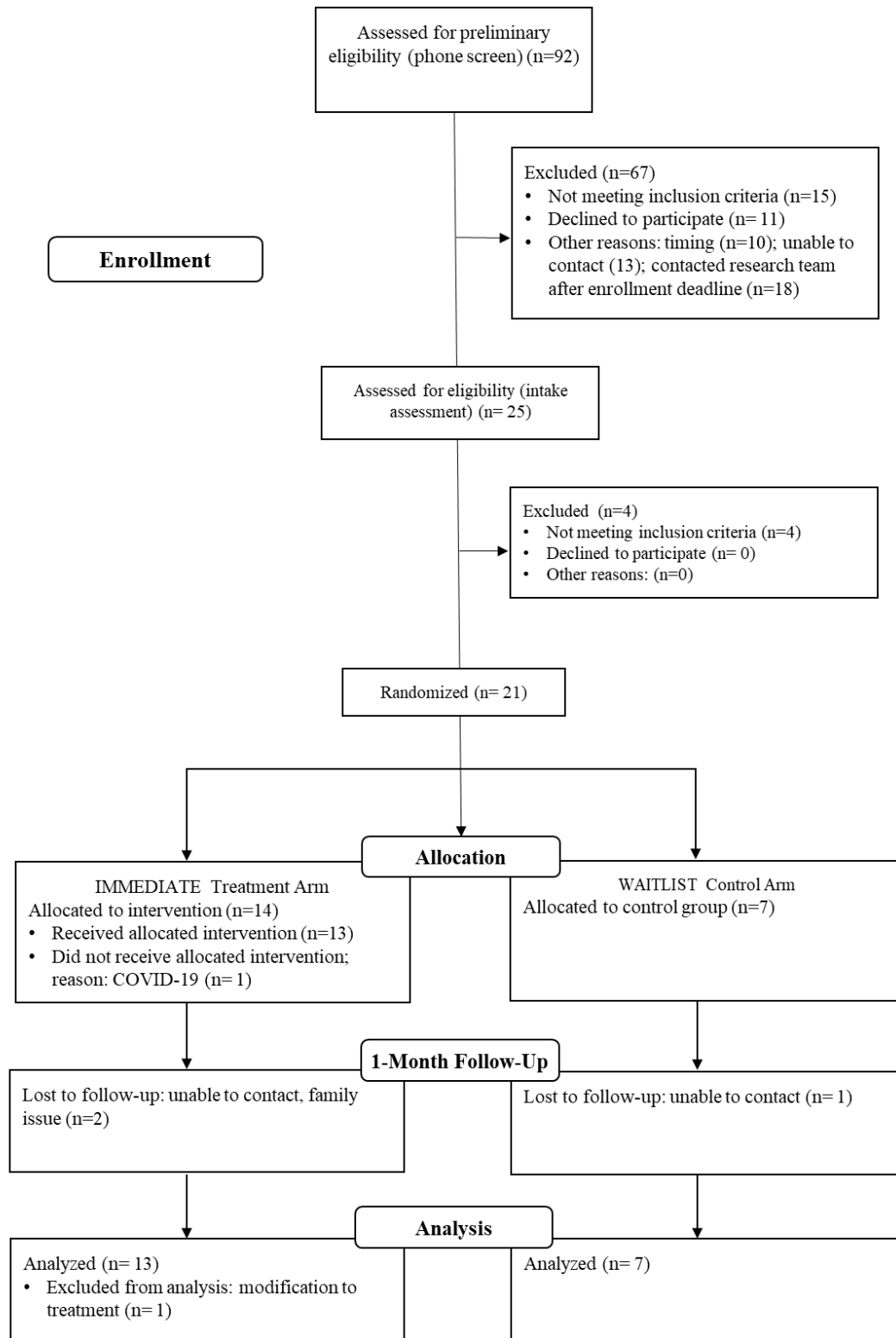


Figure 4a. Sample SUDs Ratings, Younger Group

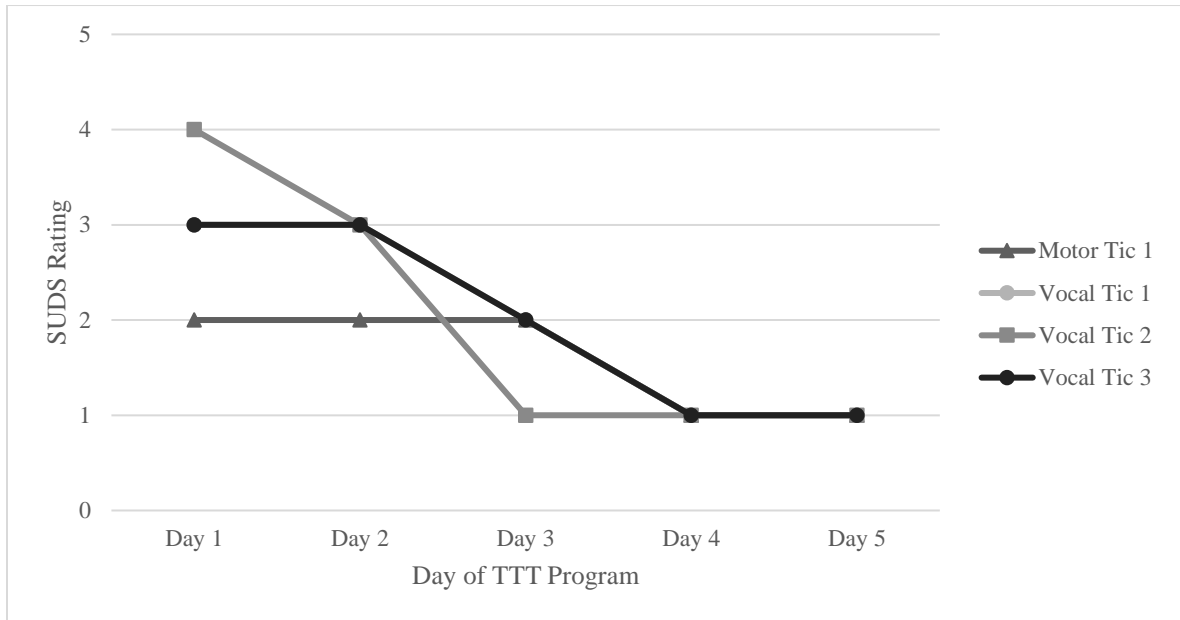


Figure 4b. Sample SUDs Ratings, Older Group

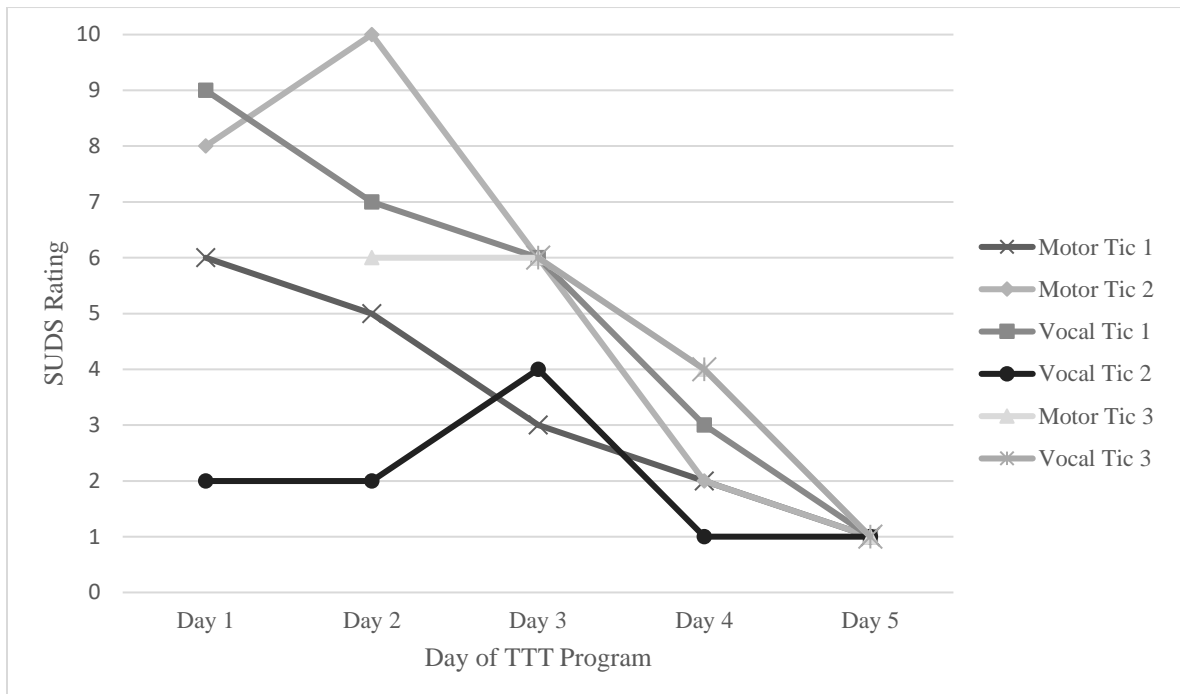


Figure 5. Yale Global Tic Severity Score, Total Tic Severity Scores by Group

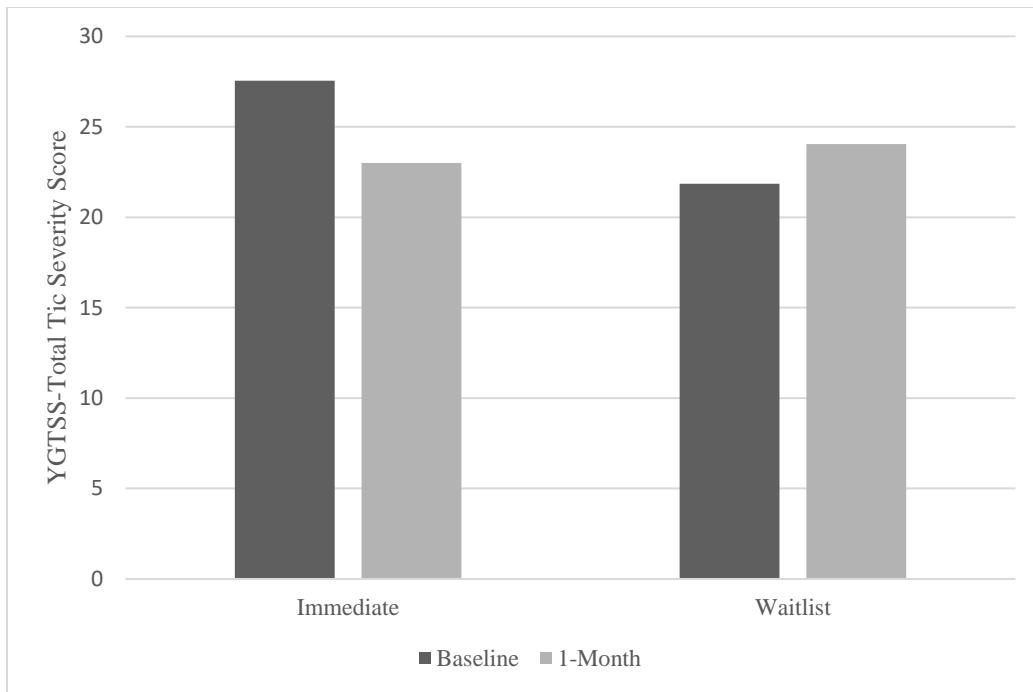


Figure 6. Yale Global Tic Severity Scores, Impairment Scores by Group

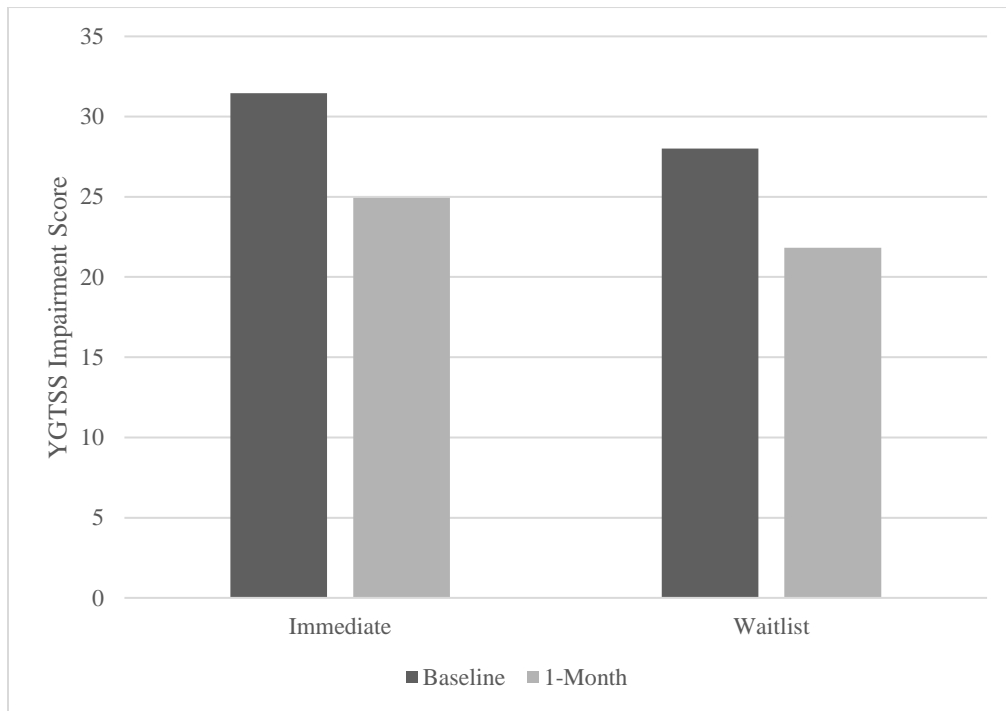


Figure 7. The Revised Children's Anxiety and Depression Scale, Anxiety Raw Scores by Group

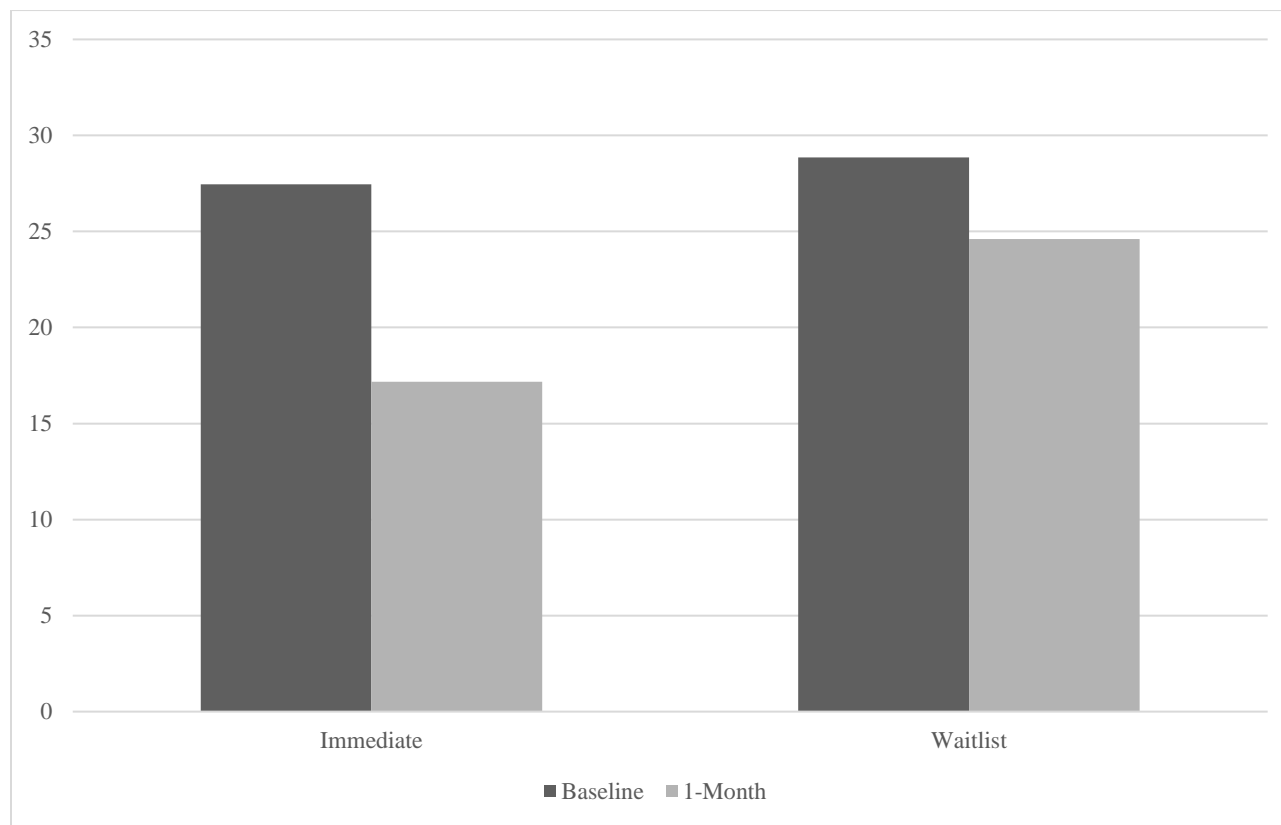


Figure 8. The Revised Children's Anxiety and Depression Scale, Depression Raw Scores by Group

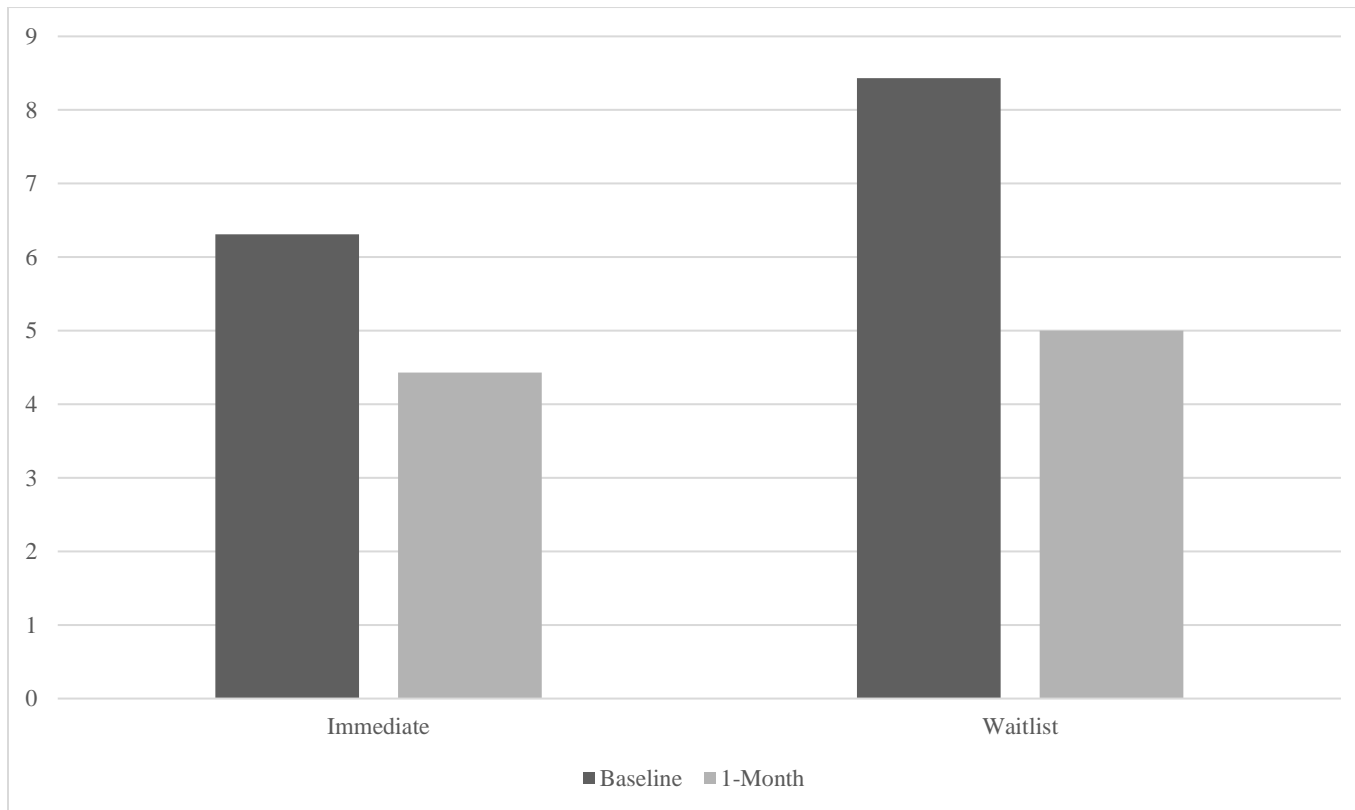


Figure 9. Disruptive Behavior Disorder Scale, Oppositional Defiant Disorder Raw Scores by Group

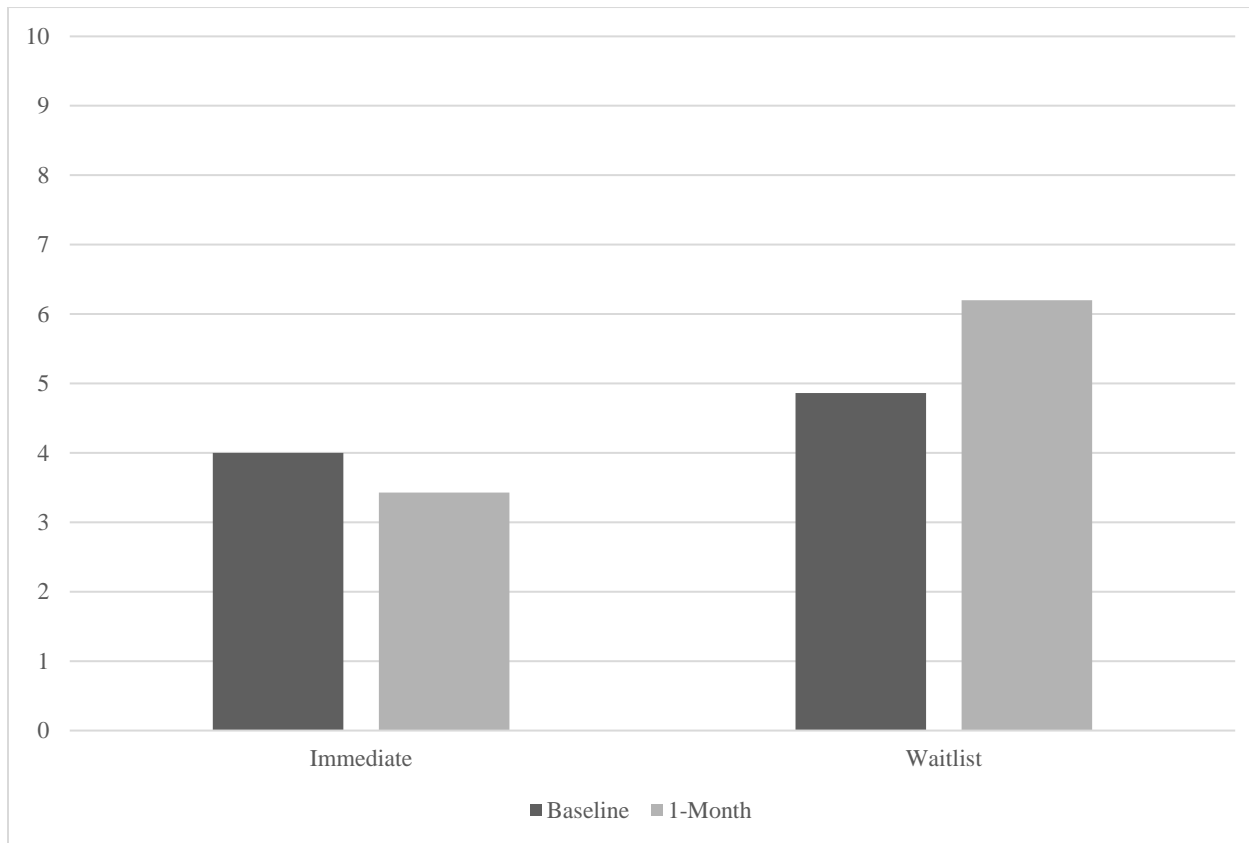


Figure 10. Disruptive Behavior Disorder Scale, Attention-Deficit/Hyperactivity Disorder – Inattention Raw Scores by Group

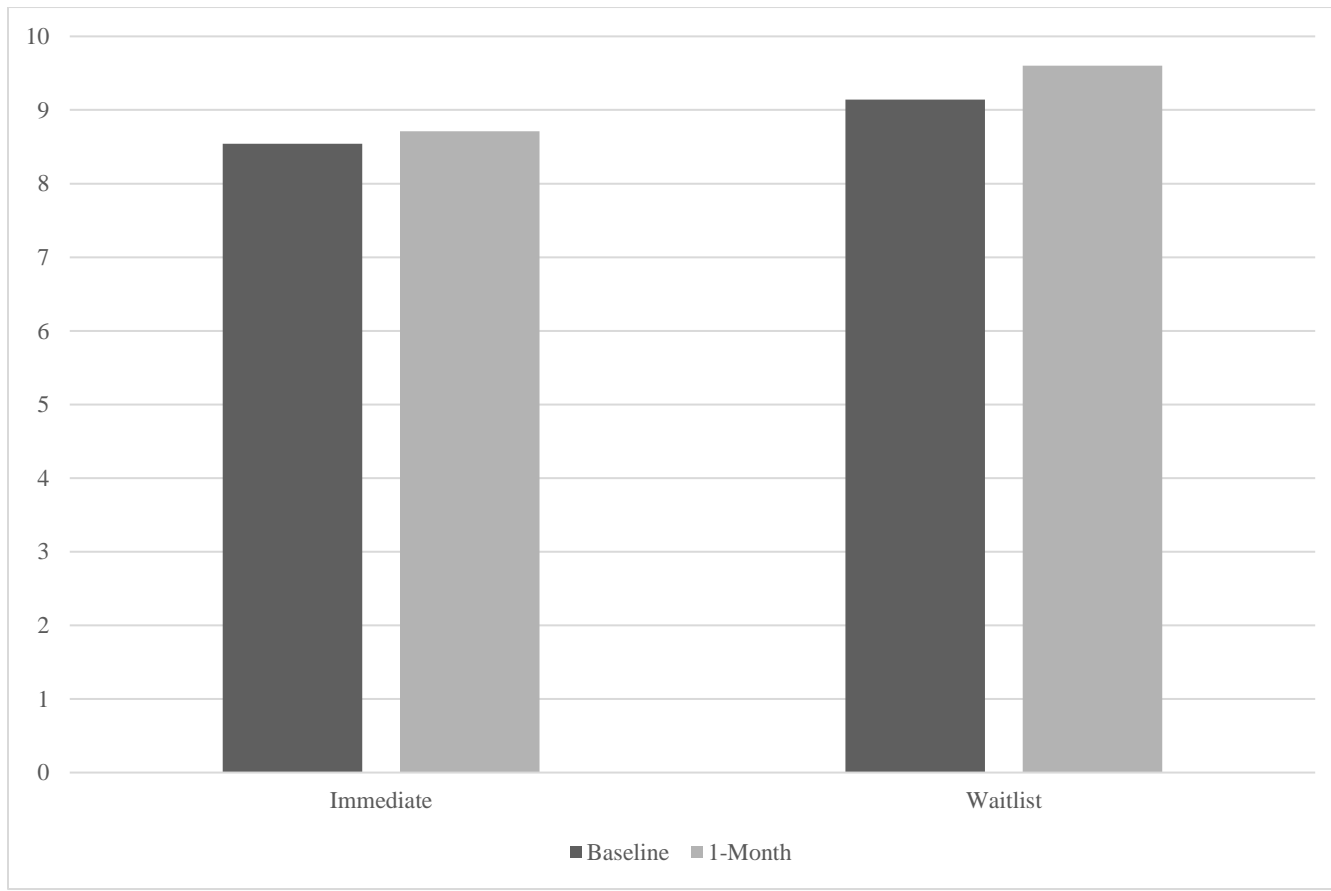


Figure 11. Disruptive Behavior Disorder Scale, Attention-Deficit/Hyperactivity Disorder – Hyperactive/Impulsive Raw Scores by Group

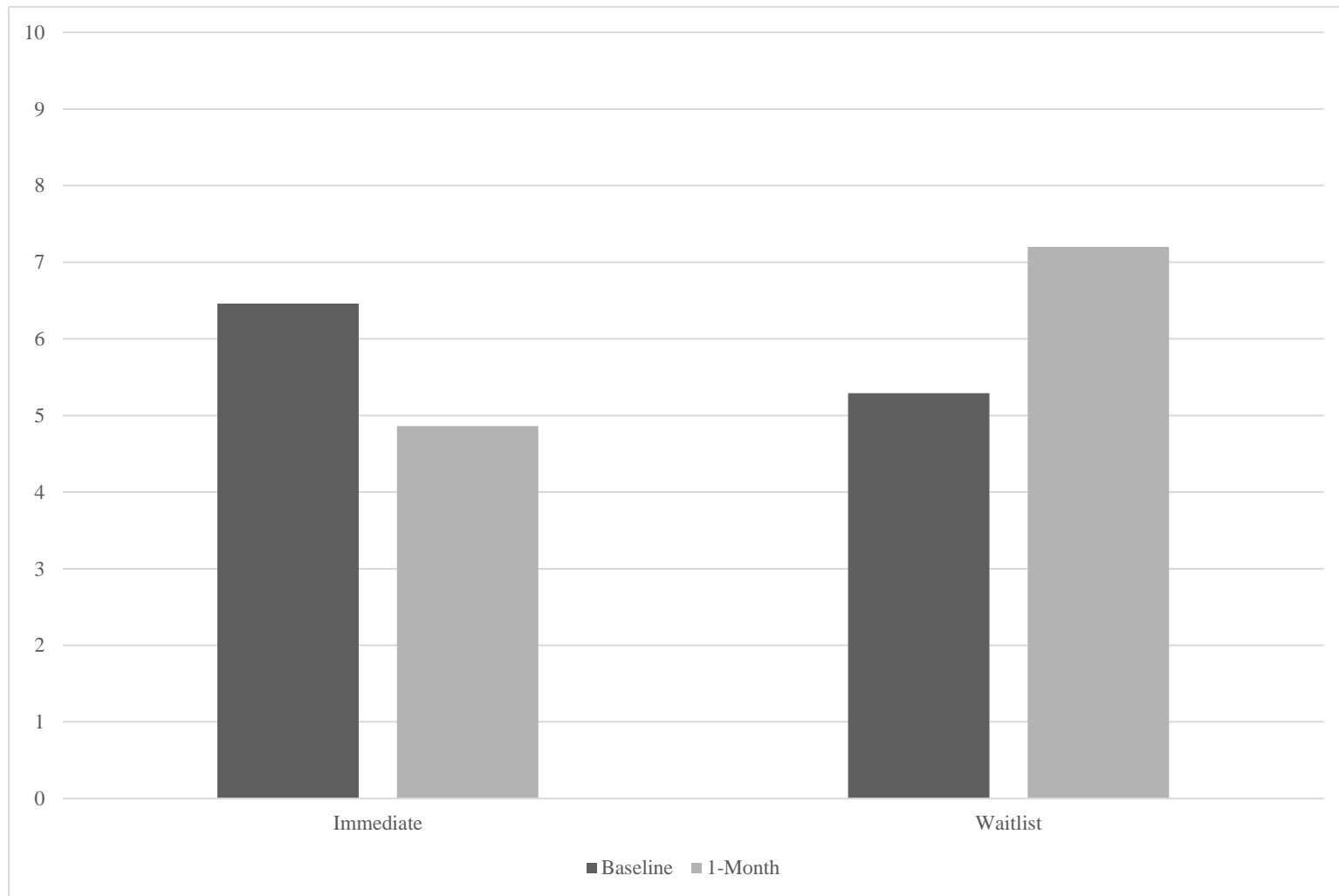


Table 1. Child and Caregiver Demographic Characteristics

<i>Child</i>	
Age <i>M</i> (<i>SD</i>)	12.54(2.44)
Sex (% Male)	50
% Taking Medication	60
Grade (%)	
Elementary School (3-5 th Grade)	45
Middle School (6-8 th Grade)	35
High School (9-12 th Grade)	20
Ethnicity/Race (%) ^a	
Asian	5.0
White	80.0
Hispanic/Latinx	40.0
Native American/Alaska Native	5.0
<i>Primary Caregiver</i>	
Age	41.90(4.66)
Sex (% Female)	95
Ethnicity/Race (%)	
Asian	5.0
White	80.0
Hispanic/Latinx	25.0
Marital Status ^a	
Married/Domestic Partnership	95.0
Separated	5.0
Employment Status ^b	
Employed – Full Time	50.0
Employed – Part Time	10.0
Homemaker	25.0
Self-Employed	10.0
Unable to Work	10.0
Household Income ^a	
\$20K-\$34,999	10.0
\$35K-\$49,999	5.0
\$50K-\$74,999	10.0
\$75K-\$99,999	15.0
\$100,000+	55.0
Did not report	5.0
Degree ^a	
High School or Equivalent	10.0
Some College, No Degree	10.0
Associate Degree	5.0
Bachelor's Degree	50.0
Master's Degree	25.0

Note: -- = Not Endorsed; Full Time Employment = 40+ hours/week; Part Time Employment = up to 39 hours/week; ^a = Only categories that were endorsed are included in the table; ^b = Parents were able to endorse more than one status

Table 2. ANOVA of Key Variable by Group at Baseline

	F	p
<i>Child Variables</i>		
Sex ^a	0.22	0.64
Age	0.05	0.82
Grade ^a	6.30	0.51
Ethnicity – Asian ^a	1.95	0.16
Ethnicity – White ^a	4.95	0.48
Ethnicity- Latinx ^a	0.59	0.44
Ethnicity- Native American ^a	0.567	0.45
Medication ^a	0.59	0.44
PUTS	0.00	0.99
<i>Caregiver Variables</i>		
Age	0.18	0.68
Sex	1.96	0.16
Ethnicity – Asian ^a	1.95	0.16
Ethnicity – White ^a	0.50	0.48
Ethnicity – Latinx ^a	0.07	0.80
Marital Status ^a	1.95	0.16
Highest Degree ^a	5.93	0.20
Job Status – Employed full time ^a	1.98	0.16
Job Status – Employed part time ^a	4.13	0.04*
Job Status – Homemaker ^a	0.07	0.80
Job Status – Self-Employed ^a	1.20	0.27
Job Status – Unable to Work ^a	0.22	0.64
Household Income ^a	5.130	0.27
<i>IE Assessment Variables</i>		
CGI-S ^a	2.23	0.53
CGAS ^a	11.21	0.59
Number of Motor Tic (YGTSS) ^b	0.42	0.53
Number of Vocal Tics (YGTSS) ^b	0.42	0.52
Frequency of Motor Tics (YGTSS) ^b	0.20	0.66
Frequency of Vocal Tics (YGTSS) ^b	0.50	0.49
Intensity of Motor Tics (YGTSS) ^b	0.14	0.72
Intensity of Vocal Tics (YGTSS) ^b	0.88	0.36
Complexity of Motor Tics (YGTSS) ^b	2.05	0.17
Complexity of Vocal Tics (YGTSS) ^b	0.65	0.43
Interference of Motor Tics (YGTSS) ^b	3.12	0.09
Interference of Vocal Tics (YGTSS) ^b	0.03	0.86
Tic Impairment (YGTSS) ^b	0.48	0.50
Motor Tic Severity Total (YGTSS) ^b	1.51	0.24
Vocal Tic Severity Total (YGTSS) ^b	0.52	0.50
Total Tic Severity Score (YGTSS) ^b	1.14	0.30
Total Yale Global Tic Severity Score (YGTSS) ^b	1.03	0.32

NOTE: * = $p < .05$; Only the ethnicities that were endorsed by parents are reported in the table.
YGTSS = Yale Global Tic Severity Scale; Clinical Global Impressions Scale – Severity, CGAS = Children’s Global Assessment Scale; PUTS= Premonitory Urge for Tics Scale; IE = Independent Evaluator; ^a = chi-square was calculated instead of an ANOVA, as this is a categorical outcome. ^b = all YGTSS outcomes were caregiver-report.

Table 3. Schedule of Assessments

Assessment Name	Baseline	Daily	Immediate Post	1-Month Follow-Up
Yale Global Tic Severity Scale (YGTSS)	X			X
Demographics	X			
Treatment Changes				X
Perceived Stress Scale (PSS)	X			X
Patient Health Questionnaire (PHQ)	X			X
Premonitory Urge for Tics Scale (PUTS)	X			
Subjective Units of Discomfort (SUDs)		X		
The Kiddie Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL)	X			
The Revised Children's Anxiety and Depression Scale (RCADS)	X			X
Disruptive Behavior Disorder Rating Scale (DBD-RS)	X			X
Pediatric Quality of Life Inventory, General Core Scales (PedsQL)	X			X
Tic Accommodation and Reaction Scale (TARS)	X			X
Children's Global Assessment Scale (CGAS)	X			X
Clinical Global Impressions Scale-Severity (CGI-S)	X			X
Burden of Treatment Participation (BTP)			X	
Treatment Acceptability Questionnaire (TAQ)			X	
Client Satisfaction Questionnaire (CSQ)			X	

Table 4. Diagnostic Characteristics

	Overall (N=20)
Tic Disorders (%)	
<i>Tourette Disorder</i>	75.0
<i>Chronic Motor Tic Disorder</i>	5.0
<i>Chronic Vocal Tic Disorder</i>	--
<i>Provisional Tic Disorder</i>	15.0
Disruptive Behavior Disorders (%)	
<i>ADHD</i>	50.0
<i>ODD</i>	10.0
<i>Elevations (Subclinical)</i>	--
Anxiety and Mood Disorders (%)	
<i>GAD</i>	20.0
<i>Separation</i>	5.0
<i>Social Anxiety</i>	20.0
<i>MDD</i>	15.0
<i>Elevations (Subclinical)</i>	35
Obsessive Compulsive Spectrum and Related Disorders (%)	
<i>OCD</i>	25.0
<i>Trichotillomania</i>	5.0
<i>Elevations (Subclinical)</i>	5
Number of Co-Occurring Subclinical Elevations/Diagnoses (%)	
1	50
2	15
3	30
4	5

Note: ADHD=Attention-Deficit/Hyperactivity Disorder; ODD=Oppositional Defiant Disorder; GAD=Generalized Anxiety Disorder; OCD=Obsessive Compulsive Disorder; MDD= Major Depressive Disorder. Some participants had elevations in more than one co-occurring diagnosis.

Table 5. Means, SDs, and Effect Sizes for Primary and Secondary Outcomes

	Baseline M(SD)	1-Month M(SD)	Within Groups (Pre-Post Change)	Between Groups
			Hedges' <i>g</i>	Hedges' <i>g</i>
<i>Primary</i>				
YGTSS Total Tic Severity				
Immediate	27.54(10.71)	23.01(11.56)	.38	
Waitlist	21.86(12.50)	24.04(8.99)	-.18	-.09
YGTSS Impairment				
Immediate	31.46(11.08)	24.94(12.02)	.55	
Waitlist	28(9.88)	21.82(6.71)	.68	.28
YGTSS Global Score (Total Tic Severity + Impairment)				
Immediate	59.0(18.40)	48.54(23.90)	.47	
Waitlist	49.86(17.96)	43.59(14.77)	.35	.22
<i>Secondary</i>				
RCADS – Anxiety				
Immediate	27.46(15.53)	17.17(6.91)	.72	
Waitlist	28.86(15.82)	24.60(24.11)	.20	-.40
RCADS – Depression				
Immediate	6.31(4.75)	4.43(2.23)	.46	
Waitlist	8.43(3.82)	5.00(3.00)	.90	-.20
DBD – ODD				
Immediate	4.00(5.30)	3.43(4.32)	.11	
Waitlist	4.86(7.13)	6.20(7.43)	-.17	-.44
DBD – ADHD-Inattention				
Immediate	8.54(4.47)	8.71(5.50)	-.03	
Waitlist	9.14(4.41)	9.60(6.19)	-.08	.70
DBD- ADHD-Hyperactivity/Impulsivity				
Immediate	6.46(5.85)	4.86(4.45)	.28	
Waitlist	5.29(5.62)	7.20(8.76)	-.25	-.33

NOTE: Missing data were accounted for using Full Information Maximum Likelihood (FIML) and the 1-month follow-up for YGTSS outcomes. Due to the limited sample size, secondary outcomes were calculated using only complete data at both time points.

Negative effect sizes indicate improvement in the immediate treatment group.

Hedges' *g* was calculated using the following equation: $(M_2 - M_1) / SD_{pooled}$, with $M_2 = I$ -TTT and $M_1 = WL$

Table 6. Model Results for Tic and Co-Occurring Diagnosis Outcomes

	B (SE)	<i>p</i>
Group – 1 Month		
YGTSS – Total Tic Severity Score	-4.73(2.72)	.081
YGTSS – Impairment Score	2.71(4.59)	.555
YGTSS – Global	-2.69(6.50)	.679
Group – 1 Month		
Anxiety	-2.92(7.65)	.703
Depression	-0.35(1.45)	.808
Group – 1 Month		
ADHD – Hyperactivity/Impulsivity	-1.61(0.86)	.060
ADHD – Inattention	0.53(2.45)	.829
ODD	-0.75(0.76)	.325

NOTE: Baseline to 1-Month stability was estimated but is not depicted here. Groups were coded such that negative scores indicate improvement in the I-TTT group (I-TTT coded 1, WL coded 0). YGTSS= Yale Global Tic Severity Scale; ADHD = Attention Deficit/Hyperactivity Disorder; ODD = Oppositional Defiant Disorder

Table 7. Within-Group (I-TTT) Tic and Co-Occurring Diagnosis Outcomes by Age and Sex

	<i>B</i> (SE)	<i>p</i>
Age – 1 Month		
YGTSS – Total Tic Severity Score	6.79(3.91)	.082
YGTSS – Impairment Score	14.20(6.74)	.035
YGTSS – Motor Tic Interference	0.23(0.64)	.722
YGTSS – Vocal Tic Interference	1.12(0.72)	.096
YGTSS – Global	15.12(11.12)	.174
Age – 1 Month		
Anxiety	-1.19(5.57)	.830
Depression	-0.74(0.92)	.423
Age – 1 Month		
ADHD – Hyperactivity/Impulsivity	0.43(0.98)	.660
ADHD – Inattention	-1.37(3.95)	.729
ODD	-2.02(1.06)	.056
Sex – 1 Month		
YGTSS – Total Tic Severity Score	6.85(2.71)	.011
YGTSS – Impairment Score	7.21(6.50)	.264
YGTSS – Motor Tic Interference	0.99(0.56)	.080
YGTSS – Vocal Tic Interference	-0.37(.71)	.597
YGTSS – Global	11.50(8.01)	.152
Sex – 1 Month		
Anxiety	-12.98(18.23)	.477
Depression	7.71(1.03)	.000
Sex – 1 Month		
ADHD – Hyperactivity/Impulsivity	-1.32(1.05)	.209
ADHD – Inattention	-4.27(5.93)	.472
ODD	-1.15(1.44)	.424

NOTE: Within-group coding of sex indicated female participants as 2 and male participants as 1. Within-group coding of age indicated older participants as 2 and younger participants as 1. The sample size for all outcomes was N=13, as FIML was used to account for missing data.

Table 8. Quality of Life, Impairment, and Parent-Related Outcomes

	<i>B (SE)</i>	<i>p</i>
Group – 1 Month		
<i>Parent Report</i>		
TARS – Total Score	-1.720(1.91)	.367
PedsQL – Physical Functioning	-6.42(6.30)	.308
PedsQL – School Functioning	22.51(11.90)	.058
Group – 1 Month		
PHQ-4	-1.05(0.95)	.268
PSS	-1.32(2.64)	.616

NOTE: PSS = Perceived Stress Scale; PHQ-4 = Patient Health Questionnaire, 4 item;
PedsQL = Pediatric Quality of Life; TARS= Tic Accommodation & Reaction Scale

VITA

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EDUCATION AND AWARDS

- | | |
|---------------------------|---|
| 2022 – 2023
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| 2021 – Present | Doctoral Candidate in Clinical Psychology
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| 2021 – Present | Dissertation Year Fellowship
University Graduate School, Florida International University
Miami, Florida |
| 2021 | Leonard Krasner Student Dissertation Award
Association for Behavioral and Cognitive Therapies (ABCT) |
| 2021 | Graduate Student Seed Funds
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| 2021 | Honorable Mention, 2021 Student Achievement Award –
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| 2017 – 2020 | M.S., Psychology
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SELECTED PUBLICATIONS AND PRESENTATIONS

Acosta, J., Dale, C.F., DiMarzio, K. & Parent, J. (2019, March). Youth sleep problems and parenting behaviors: The roles of warmth, behavioral control, and emotion socialization. Presented at the International Convention of Psychological Science (ICPS), Paris, France.

Dale, C.F., Acosta, J., McGregor, E., & Parent, J. (2019, March). The longitudinal association between youth screen time and behavioral problems: the role of sleep disturbances. Presented at the Society for Research in Child Development Biennial Meeting, Baltimore, MD.

Dale, C. F., DiMarzio, K., Morris, S. J., Ramos, M. C., Acosta, J., & Parent, J. (under review). Taming Tics Together: An intensive telehealth-based intervention for youth with tics and co-occurring diagnoses. [Manuscript submitted for publication]. Department of Psychology, Florida International University.

Dale, C., Ortiz, J., Cadet, G., Gallat, A., DiMarzio, K., Morris, S., Ramos, M., Acosta, J., & Parent, J. (2022, February). Evaluation of an intensive telehealth intervention for youth with tics and co-occurring diagnoses: A case series. Poster to be presented at the Miami International Child & Adolescent Mental Health (MICAMH) Conference, Miami, FL. (Conference canceled)

Dale, C., Parent, J., Forehand, R., DiMarzio, K., Sonuga-Barke, E., Long, N., & Abikoff, H. (2021). Behavioral Parent Training for preschool ADHD: Family-centered profiles predict changes in parenting and child outcomes. *Journal of Clinical Child and Adolescent Psychology*. Advance online publication. <https://doi.org/10.1080/15374416.2020.1867987>

Dale, C., Ramos, M., & Parent, J. (2021). Intensive behavioral therapy for tics and co-occurring ADHD: A case report. *Cognitive and Behavioral Practice*. Advance online publication.

DiMarzio, K., Parent, J., Forehand, R., Champion-Thigpen, J., Acosta, J., Dale, C., & Compas, B. (2021). Parent-child role confusion: Exploring the role of family processes in the context of parental depression. *Journal of Clinical Child and Adolescent Psychology*. Advance online publication. <https://doi.org/10.1080/15374416.2021.1894943>

McKee, L. G., DiMarzio, K., Parent, J., Dale, C., Acosta, J., & O'Leary, J. (2021). Profiles of emotion socialization across development and longitudinal associations with youth psychopathology. *Research on Child and Adolescent Psychopathology*. Advance online publication. <https://doi.org/10.1007/s10802-021-00829-6>.