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PILOT OPEN CASE SERIES OF VOICE OVER INTERNET PROTOCOL-DELIVERED ASSESSMENT AND BEHAVIOR THERAPY FOR CHRONIC TIC DISORDERS

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Abstract

Comprehensive Behavioral Intervention for Tics (CBIT) is an efficacious treatment for children with chronic tic disorders (CTDs). Nevertheless, many families of children with CTDs are unable to access CBIT due to a lack of adequately trained treatment providers, time commitment, and travel distance. This study established the interrater reliability between in-person and Voice over Internet Protocol (VoIP) administrations of the Yale Global Tic Severity Scale (YGTSS), and examined the preliminary efficacy, feasibility, and acceptability of VoIP-delivered CBIT for reducing tics in children with CTDs in an open case series. Across in-person and VoIP administrations of the YGTSS, results showed mean agreement of 91%, 96%, and 95% for motor, phonic, and total tic severity subscales. In the pilot feasibility study, 4 children received 8 weekly sessions of CBIT via VoIP and were assessed at pre- and posttreatment by an independent evaluator. Results showed a 29.44% decrease in clinician-rated tic severity from pre- to posttreatment on the YGTSS. Two of the 4 patients were considered treatment responders at posttreatment, using Clinical Global Impressions–Improvement ratings. Therapeutic alliance, parent and child treatment satisfaction, and videoconferencing satisfaction ratings were high. CBIT was considered feasible to implement via VoIP, although further testing is recommended.

Keywords

tic disorders, behavior therapy, voice over internet protocol, videoconferencing

Chronic tic disorders (CTDs), including Tourette Syndrome (TS) involve sudden, repetitive involuntary motor and/or vocal tics that have been present for longer than 1 year (<u>American Psychiatric Association, 2013</u>). CTDs are associated with impairment in physical, psychological, social, and family functioning and reduced quality of life (<u>Conelea et al., 2011; Cooper, Robertson, & Livingston, 2003; Cutler, Murphy, Gilmour, & Heyman, 2009;</u> <u>Storch et al., 2007</u>). Antipsychotic medications have typically been the first-line intervention for CTDs (<u>Gilbert,</u> <u>2006</u>). Although moderately effective for reducing tics, they are often associated with adverse side effects that limit their clinical use, including: weight gain, sedation, cognitive dulling, depressive symptoms, and neurological side effects (e.g., tardive dyskinesia, dystonia; <u>Scahill et al., 2006</u>). Behavior therapy has emerged as an effective nonpharmacological treatment option for reducing tics (<u>Cook & Blacher, 2007; Himle, Woods, Piacentini, &</u> <u>Walkup, 2006</u>), and has recently been recommended as a first-line treatment for tics in European and Canadian best-practice guidelines (<u>Steeves et al., 2012; Verdellen et al., 2011</u>).

Recently, several behavioral techniques, including psychoeducation, habit reversal training (HRT), functionbased assessment and intervention, self-monitoring, relaxation training, and behavioral rewards (Woods et al., 2008), have been combined to create a multifaceted treatment package called Comprehensive Behavioral Intervention for Tics (CBIT). CBIT was recently compared to supportive psychotherapy and education (PST) in two separate multisite RCTs in children (Piacentini et al., 2010) and adults (Wilhelm et al., 2012). Acute treatment for both groups involved 8 sessions over 10 weeks. Results of the child study (*n* = 126) demonstrated that 52.5% of the participants in the CBIT group were acute phase treatment responders, compared to 18.5% of the PST group. There were also significantly greater reductions in clinician-rated tic severity, as measured by the Yale Global Tic Severity Scale (YGTSS), in CBIT (30.8%) relative to PST (14.2%); reductions were similar in magnitude to those found in several RCTs of medications (Scahill et al., 2013). Additionally, decreases in the YGTSS impairment scores in the CBIT group (51.2%) were greater compared to PST (29.9%). Responder status was maintained through 6-month follow-up for 87% of the acute-phase CBIT responders.

Despite the efficacy of CBIT, many families of children with CTDs are unable to access the treatment. In a recent national survey examining treatment utilization in children and adults with chronic tic disorders (n = 672), treatment-seeking families cited several reasons for not pursuing a behavior therapy option to treat their child's

tics, including little time to attend weekly therapy, a lack of treatment providers in their region, and long distances to providers (<u>Woods, Conelea, & Himle, 2010</u>).

One way to increase access to treatment among underserved populations is through the use of videoconferencing (VC) technologies. The traditional VC model, such as that used by <u>Himle et al. (2012)</u>, typically involves two clinical or academic sites with specialty VC equipment connected via a dedicated high-speed connection. Although VC has been shown to be efficacious, feasible, and acceptable when used to deliver interventions for a range of psychological disorders (e.g., <u>Capner, 2000; Simpson, 2009</u>), traditional VC has only recently been applied to CTDs. In an initial pilot study, <u>Himle et al. (2010</u>) delivered CBIT via VC to three children with TS and found that all three showed significant reductions in tic severity (<u>Himle et al., 2010</u>). In a follow-up study, <u>Himle et al. (2012</u>) randomly assigned 20 children to receive CBIT delivered either via VC or traditional face-to-face delivery. Results showed significant pre to post reductions in clinician-rated tic severity (YGTSS) in both groups, with mean reductions of 33% and 27% for VC and face-to-face groups, respectively. No significant differences in mean reductions in tic severity were similar when using VC format. Additionally, both treatment modalities were rated as highly acceptable by parents and children, with no significant differences between groups. There were also no significant differences in parent and child-reported therapeutic alliance between groups (<u>Himle et al., 2012</u>).

Despite its growing popularity, the traditional VC delivery model fails to address several important accessibility barriers. Treatment may be restricted to the locations where the equipment is installed and typically requires patients, and sometimes the therapist, to travel to a dedicated site to use the equipment. Another limitation is that the traditional VC model often requires the support of specially trained personnel at both VC sites to maintain the equipment, assist the therapist and client in connecting and using the equipment, and to provide logistical support to the patient (Hilty, Luo, Morache, & Nesbitt, 2002; Simpson, Bell, Knox, & Mitchell, 2005). Such reliance on support resources not only increases the cost of treatment, but also poses logistical barriers that may limit adoption of VC to deliver care.

A newer alternative to traditional VC delivery is the use of Voice over Internet Protocol (VoIP) telephony or webbased VC (e.g., Skype). This technology allows users to connect over the Internet through software using wired or wireless high-speed connections and a basic web camera. Delivering CBIT via web-based VC has several potential advantages over traditional VC delivery. First, it benefits patients, as services may be received from their own home computers, reducing costs, and potentially decreasing time missed from work and school to attend appointments. Second, a web camera is a lower cost alternative to the equipment needed for traditional VC. Third, web-based VC benefits clinicians, as they may be able to work directly from their office or be otherwise freed from the constraints posed by the physical stability of traditional VC equipment. Lastly, another advantage is that VoIP allows patients broader, direct access to experts in a particular treatment or field. Although both traditional VC and VoIP delivery provide access to treatment for those who live far away from knowledgeable treatment providers, traditional VC typically requires patients to drive to the nearest clinic that houses the equipment for treatment.

Despite the potential of VoIP to increase access to treatment, there are potential limitations. Specifically, call quality and reliability are highly dependent on the type and speed of the Internet connection (Kazemitabar, Ahmed, Said, & Habsullah, 2010) and may be influenced by certain computer specifications, including processor speed, random access memory, and hard drive disk space (Ramirez, 2011). Additionally, external technical support may not be readily available, particularly for consumers using VoIP in their homes. There is some research assessing the efficacy and feasibility of VoIP-delivered treatment, with promising findings. VoIP treatments for insomnia (Lichstein, Scogin, Thomas, DiNapoli, Dillon, & McFadden, 2013), social phobia (Yuen et al., 2013), OCD (Storch et al., 2011), substance abuse (King, Brooner, Peirce, Kolodner, & Kidorf, 2014), and

depression in older adults (<u>Choi et al., 2014a</u>; <u>Choi, Wilson, Sirrianni, Marinucci, & Hegel, 2014b</u>) have been associated with significant decreases in target symptoms. Additionally, high satisfaction (<u>Choi et al., 2014a; King et al., 2014</u>; Storch et al.; Yuen et al.) and a strong therapeutic alliance (King et al.; <u>Lichstein et al., 2013</u>; Yuen et al., 2013) have been observed. With respect to feasibility, patients have found the modality to be convenient, but technical difficulties (<u>Choi et al., 2014b</u>; <u>King et al., 2014</u>; <u>Lichstein et al., 2013</u>; <u>Yuen et al., 2013</u>) and challenges reading body language (<u>Lichstein et al., 2013</u>; <u>Storch et al., 2011</u>) are common. Despite preliminary evidence, research on the efficacy and feasibility of VoIP-delivered therapy is still rather limited; and the validity and reliability of administering clinician-rated measures via this modality have yet to be assessed. Therefore, two studies were performed as part of research testing the feasibility of using VoIP to assess and treat CTD in children. To gain preliminary data on the feasibility of assessing tic severity over VoIP, Study 1 assessed the interrater agreement between in-person and VoIP-administrations of a primary tic rating scale, the Yale Global Tic Severity Scale (Leckman et al., 1989). In Study 2, the feasibility, acceptability, and preliminary efficacy of VoIP-delivered behavior therapy for CTDs were evaluated.

Study 1

Method

Participants

Participants were 10 former and current patients of a university-based Tic Disorders Specialty Clinic recruited through completed and active studies. Participant ages ranged from 10 to 14 years (*M* = 12.39, *SD* = 1.36). Nine participants were male, and one was female. All participants were identified as Caucasian. Inclusion criteria included (a) residence in the clinic's home state (b) an age between 9 and 17, (c) DSM-IV-TR diagnosis of CTD (Chronic Motor/Vocal Tic Disorder or Tourette Syndrome), and (d) fluency in English. Exclusion criteria included (a) a DSM-IV-TR substance abuse or dependence or conduct disorder diagnosis within the past 3 months, (b) a lifetime DSM-IV-TR diagnosis of pervasive developmental disorder, mania, or psychotic disorder, (c) any serious psychiatric, psychosocial, or neurological condition requiring urgent treatment beyond that provided through the study, and (d) lack of a functional and accessible home computer, and high-speed (i.e., cable/DSL) Internet connection.

Materials

Skype was used to perform web-based VC. Skype, owned by Microsoft Corporation, is a peer-to-peer VoIP system, providing free video and voice calling, instant messaging, and file sharing between users (Skype Technologies S. A., 2013). Skype was chosen because of its popularity among consumers and ease of use (Garfinkel, 2005). Treatment was delivered from a private secure clinic room, using a Dell Optiplex GX 980 desktop computer with a 21.5-inch screen, Logitech c270 web camera, and a high-speed (54.0 megabytes per second) wireless local area network Internet connection available through the university. The Skype picture-in-picture function was used at all times so the therapist could monitor the participant's movements and body positioning. Participants used their home desktop or laptop computer, high-speed Internet connection, and a web camera to connect with the therapist. A low-cost Logitech c110 web camera was loaned to any family who did not already own one.

Measures

Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989)

The YGTSS, a clinician-rated tic severity measure, was administered. It allows for the assessment of tics using five categories: number, frequency, intensity, complexity, and impairment. Each item is rated on a scale of 0 to 5. The measure has independent subscales for motor and vocal tic severity, each ranging from 0 to 25, an overall

impairment scale ranging from 0 to 50, and an overall tic severity score: the sum of the motor and vocal subscales. When delivered face-to-face, the YGTSS has demonstrated acceptable psychometric properties, including good construct validity, internal consistency, and interrater reliability; and acceptable convergent and divergent validity (Leckman et al., 1989; Storch et al., 2005).

Procedure

A brief phone screen was conducted with interested participants to gauge study eligibility. A study evaluator, trained in the administration of the YGTSS, then traveled to the homes of eligible participants to complete the study procedures. During the visits, the evaluator sought parental consent and child assent using Institutional Review Board–approved forms, which highlighted risks of participation including, but not limited to, protecting session content from being overheard by family members and others within the home, and maintaining Internet network security and privacy during VC. The evaluator also assisted family members to download and set up the Skype software and to install a web camera for use during the study visit. Families were then connected via Skype to a primary rater located in a private room in the clinic. The primary rater, also trained to perform the YGTSS, administered the interview via web-based VC. The evaluator in the home, who was off-camera, served as the secondary rater. They observed the interview and independently rated the child's tics from within the families' homes. Participants were paid \$25 for completion of study procedures.

Results

The interrater agreement between in-person and VoIP administrations of the YGTSS was calculated by dividing the lower total score by the higher total score for each subject. Across all 10 participants the average agreement for the YGTSS motor subtotal was 91% (range = 71% to 100%). The YGTSS vocal tic subtotal average was 96% (range = 85% to 100%), and the total score (motor and vocal subtotals) was 96% (range = 88% to 100%).

Study 2

Methods

Participants

Participants were four Caucasian males, between the ages of 10 and 13, diagnosed with TS (see Table 1), who resided within 83.5 miles of the clinic on average. They were selected from the Study 1 sample, as they were the only four participants who had not yet received behavior therapy. Inclusion criteria were (a) residence in the clinic's home state (b) age between 9 and 17, (c) DSM-IV-TR diagnosis of CTD (Chronic Motor/Vocal Tic Disorder or Tourette Syndrome), (d) a Clinical Global Impressions – Severity score \geq 4 (moderately ill or worse), (e) a YGTSS Score \geq 14 and < 30 for TS or \geq 10 and < 20 for CMTD, (f) medication free or on a stable medication for the treatment of tics, OCD, ADHD, anxiety, and/or depressive disorder for at least 6 weeks, with no planned changes during the study period, and (g) fluency in English. Exclusion criteria were (a) a total YGTSS tic score > 30, considered high severity, selected to remain consistent with other behavior therapy exclusion criteria; for YGTSS scores > 30, a caseness panel discussed additional treatment options with the family, and determined the appropriateness of the patient's participation in the study), (b) a Wechsler Abbreviated Scale of Intelligence-Vocabulary (The Psychological Corporation, 1999) subtest T-score < 37 (low average range), (c) a DSM-IV-TR diagnosis of substance abuse or dependence, or conduct disorder within the past 3 months, (d) a lifetime DSM-IV-TR diagnosis of pervasive developmental disorder, mania, or psychotic disorder, (e) any serious psychiatric, psychosocial, or neurological condition requiring urgent treatment beyond that provided through the study, (f) four or more previous HRT sessions for TS, (g) lack of a functional and accessible home computer, and/or highspeed (i.e., cable/DSL) Internet connection, and (h) refusal to sign a release of information form for the child's local primary care physician, mental health professional, or neurologist.

Table 1. Patient Characteristics								
		Participant 1	Participant 2	Participant 3	Participant 4			
	Gender	Male	Male	Male	Male			
	Age	13	13	11	10			
	Race	Caucasian (non-Hispanic)	Caucasian (non- Hispanic)	Caucasian (non- Hispanic)	Caucasian (non- Hispanic)			
	WASI-Vocab T- score	73 (very superior)	43 (average)	52 (average)	56 (average)			
	Mini-Kid Diagnosis	TS; ADHD-inattentive type	TS; ADHD-inattentive type	TS	TS			
	Medication	Vyvanse, Sertraline, Clonidine	None	None	None			

TS = Tourette Syndrome; ADHD = Attention Deficit Hyperactivity Disorder.

Materials

For a detailed description of computer equipment used by the treatment site and participants during the study period see <u>Table 2</u>.

Table 2. Computer Equipment and Features

	Participant 1	Participant 2	Participant 3	Participant 4	Treatment Site
Internet Connection	Wireless	Wireless	Wireless	Wireless	Wi-Fi
Web camera Type	Built-in	Built-in	Separate	Built-in	Separate
Web camera model	N/A	N/A	Logitech c110	N/A	Logitech 270
Additional Equipment	Headset	None	None	None	None
Computer Type	Laptop	Desktop	Desktop	Laptop	Laptop
Computer Location	Basement	Basement	Kitchen	Dining room/Kitchen	Office
Computer Age	1.5 years	7.5 months	7 months	3 weeks	12 months
Computer Make/Model	Gateway NV53	iMac12.2	Lenovo	Hewlett Packard Pavillion G7	Dell Optiplex 980
Operating System	Windows 7	Mac OS 10	Windows 7	Windows 7	Windows XP
Processor	AMD Athlon II	Intel Core i5	Intel R core i5	AMD a6	Intel Core i5
Processor Speed	2.0 GHz	2.7 GHz	2.5 GHz	2.7 GHz	3.2 GHz
Random Access Memory	4 GB	4 GB	6 GB	3.48 GB	3.49 GB
Free Hard Drive Space	199.0 GB	892.9 GB	376.0 GB	575.0 GB	217.0 GB

Measures

Mini International Neuropsychiatric Interview for Schizophrenia and Psychotic Disorders Studies for Children and Adolescents (MINI-Kid; <u>Sheehan et al., 1998</u>)

The MINI-Kid is a brief structured clinician-rated diagnostic interview assessing for 27 psychiatric disorders in children and adolescents. The interview has adequate to excellent interrater and test-retest reliability and good to excellent convergent validity for most diagnoses (<u>Sheehan et al., 2010</u>).

Wechsler Abbreviated Scale of Intelligence (WASI; The Psychological Corporation, 1999)

The WASI is a psychometrically acceptable (<u>Axelrod, 2002; Canivez, Konold, Collins, & Wilson, 2009; Saklofske,</u> <u>Caravan, & Schwartz, 2000</u>) measure of intellectual functioning for individuals ages 6 to 89 years. The vocabulary subtest was administered during the screening assessment.

YGTSS (Leckman et al., 1989)

The YGTSS, described in Study 1, was administered by an independent evaluator via web-based VC.

Clinical Global Impression-Severity and Improvement Scales (CGI-S and CGI-I; Guy, 1976)

The CGI-S and CGI-I are clinician-rated scales assessing global severity (-S) and improvement (-I) in the patient since the baseline assessment. Clinicians rate the patient's severity using the following anchors: 0 (*not assessed*), 1 (*normal, not at all ill*), 2 (*borderline ill*), 3 (*mildly ill*), 4 (*moderately ill*), 5 (*markedly ill*), 6 (*severely ill*), and 7 (*extremely ill*). Clinicians assess the patient's improvement using the following ratings, which were slightly modified from the original scale: 0 (*not assessed*), 1 (*very much improved*), 2 (*much improved*), 3 (*improved*), 4 (*minimally improved*), 5 (*no change*), 6 (*minimally worse*), 7 (*much worse*), and 8 (*very much worse*). The scales have good concurrent validity and are sensitive to change (Leon et al., 1993).

Child's Perception of Therapeutic Relationship (Kendall et al., 1997)

The Child's Perception of Therapeutic Relationship scale is a 10-item measure, assessing the child's perceptions of the quality of the therapeutic relationship. Total scores range from 8 to 40, with higher scores reflecting a more positive perception of the therapeutic relationship. The instrument displays adequate internal consistency and reliability (Elvins & Green, 2008).

Treatment Acceptability Questionnaire (TAQ; Hunsley, 1992)

The TAQ is a 6-item measure assessing patient and parent reactions to treatment. Items covering acceptability of the treatment, side effects, ethics and effectiveness of the treatment, and knowledge and trustworthiness of the treatment provider were rated by parents on a Likert scale of 1 to 7. Total scores on the measure range from 7 to 42, with higher scores reflecting greater acceptance. The questionnaire displays good internal consistency and test-retest reliability, and some indication of concurrent validity (Hunsley, 1992).

Client Satisfaction Questionnaire (CSQ; Larsen, Attkisson, Hargreaves, & Nguyen, 1979)

The CSQ is an 8-item measure designed to assess patient satisfaction with health-care service quality and type, the extent to which the service met patient needs, willingness to recommend the service to others, satisfaction with the degree of help, the extent to which the service helped the patient manage problems more effectively, overall satisfaction, and willingness to return for future services. Individual items are rated on a scale of 1 to 4, with total scores ranging from 8 (*low satisfaction*) to 32 (*high satisfaction*). The CSQ possesses high internal consistency (Cox, Brown, Peterson, & Rowe, 1982; Larsen et al., 1979; Roberts & Attkisson, 1983), and excellent concurrent validity (Larsen et al., 1979; Nguyen, Attkisson, & Stegner, 1983).

Videoconferencing Satisfaction Questionnaire

This 12-item parent and child-report measure assesses satisfaction with the VC aspect of treatment, including such domains as the therapeutic relationship and therapist competence, comfort using the equipment, audio and visual quality, the ability of VC to address treatment barriers, and preference for VC relative to in-person treatment. It was adapted from the Telemedicine Satisfaction Questionnaire (<u>Nelson, Barnard, & Cain, 2003</u>). Items are rated on a Likert scale of 1 (*strongly disagree*) to 7 (*strongly agree*), with total scores ranging from 7 (*low VC satisfaction*) to 84 (*high VC satisfaction*).

VOIP Call Quality Ratings

The severity and type of technical difficulties during VC treatment and assessment sessions were tracked during treatment sessions, using questions adapted from Yuen (2010). Ratings were qualitative, and no numerical values were assigned to the descriptors. Severity ratings included: "None," "Insignificant" (session quality not affected), "Minimal" (session quality minimally affected), "Moderate" (session quality moderately affected), "Major" (session quality majorly affected), and "Severe" (could not complete the session). Staff who endorsed the presence of technical difficulties were asked to indicate the type of technical difficulties by selecting from four categories: sound quality (e.g., choppy, echoing, soft, delayed), video quality (e.g., blurry, freezing, choppy, delayed, dropped calls), unable to hear sound, and unable to see video. All relevant types of technical difficulties could be indicated for each session.

Procedure

Participants were recruited through a Tic Disorders Specialty Clinic. Six participants were screened via the phone to gauge eligibility. All were eligible, but two declined to participate. The first declined due to concerns that receiving treatment via VoIP would not be hands-on enough compared to in-person treatment, and the second declined due to a lack of interest in receiving treatment within the context of a research study. Those agreeing to participate were mailed packets including Institutional Review Board-approved consent and assent forms (which addressed home and Internet privacy issues like those used in Study 1), in addition to standard psychology clinic forms (i.e., consent for treatment, clinic background form, acknowledgement of receipt of privacy practices, email permission), instructions for downloading Skype, and guidelines for maintaining privacy when using web-based VC. Consent forms were then reviewed and discussed via telephone with interested participants. Also, a web camera was mailed to any participant who did not already own one, for use during the study period. Next, a baseline assessment was conducted by an independent evaluator via Skype using the measures describe above. The independent evaluator was a master's-level doctoral student who received training and supervision in administration of the clinician-rated assessments. The purpose of the evaluation was to (a) confirm eligibility, (b) determine initial tic severity, (c) establish the presence of comorbid conditions, and (d) document the type of VC and computer equipment being used by participants for the study. After eligibility was confirmed, a CBIT parent workbook (Woods et al., 2008) was mailed to families.

Treatment was provided by two master's-level therapists each with 2 years' experience performing CBIT. Therapists were trained and supervised according to procedures in the original CBIT trial (<u>Piacentini et al., 2010</u>). Treatment consisted of 8 weekly sessions and included several components. Families followed along with the protocol using a parent workbook, listing treatment procedures, homework assignments, and blank homework sheets. Treatment was conducted with both the child and parent together, with flexibility allowed for older adolescents.

In Session 1, the therapist provided psychoeducation about tics, created a hierarchical list of tics to treat, discussed tic-related impairment, and formed a behavioral reward program wherein the patient received points towards rewards for participating in treatment. In session 2 function-based assessment and interventions and HRT were conducted for the first tic on the hierarchy (typically the most bothersome tic). Function-based strategies involved identifying and minimizing any antecedents and consequences to ticcing associated with higher tic severity; HRT involved awareness training, competing response training, and social support. In awareness training, the child was taught to identify when tics or premonitory sensations occurred. This was done by having the child operationally define the tic and any premonitory urges, and instructing the child to practice noticing the tics as they occurred in session. In competing response training, the child selected a behavior that was (a) physically incompatible with the tic, (b) able to be held for 1 minute or until the urge to tic had subsided, and (c) socially discreet. This "competing response" was then done for 1 minute or until the urge dissipated contingent upon the occurrence of the tic or urge/signal to tic. In the social support component,

parents were trained to provide prompts and praise for use of competing responses. As in Session 2, the therapist targeted a new tic using function-based interventions and HRT each week in Sessions 3 through 8. Sessions 4 and 5 featured instruction in relaxation techniques, including relaxed breathing, and progressive muscle relaxation. Sessions 7 and 8 included a discussion of relapse prevention strategies.

Between sessions, patients and parents completed tic-monitoring homework wherein 3 to 4 times per week they independently tallied each occurrence of a targeted tic during a 15- to 20-minute interval. Self-monitoring homework was reviewed at the beginning of each session by having the parent and child read it to the therapist from the workbook. In between sessions, parents supported their children by reminding them to use the competing response when they noticed the child had forgotten, and praising the child when they observed that the child had correctly used a competing response contingent on tic occurrence.

Following treatment, the independent evaluator performed a postassessment (i.e., YGTSS, CGI scales) via Skype. Also, participants and parents were prompted via e-mail to complete Internet questionnaires (i.e., Child's Perception of Therapeutic Relationship, TAQ, CSQ, and Videoconferencing Satisfaction Questionnaire) on a secure Internet surveying website (<u>Qualtrics Labs, Inc. software, 2013</u>). Participants were reimbursed a total of \$100 for completion of both baseline and posttreatment assessments.

Results

Andrew¹

Andrew was a 13-year-old male diagnosed with TS and ADHD-Inattentive type. At the time of the study, he was taking several medications for TS and ADHD, including Vyvanse, Sertraline, and Clonidine. His WASI-vocabulary *T*-score was 73, which fell in the very superior range. At baseline, Andrew received a YGTSS total tic severity score of 32, with tic-related impairment falling in the moderate range, and a CGI-S score of 5 (markedly ill; see Table 3). Andrew entered treatment with several tics, including: eye blinking, squinting, nostril flaring, ear wiggling, toe cracking, coughing, throat clearing, sniffing, repeating words, and emphasizing certain syllables in words. There were several tics he repeated a certain number of times, including making a half smile, knuckle cracking, retracing over his own writing, evening up, hyperextending his elbow, wrist movements, and shoulder shrugs. He experienced tic-related impairment in the form of feelings of embarrassment and stress, and interference in sports. At the final assessment, his total YGTSS score had decreased by 6 points to a score of 26, and he endorsed tic-related impairment in the minimal range. His CGI-S score was 4 (*moderately ill*), and his CGI-I score was 3 (*improved*).

	Baseline				Post				
	YGTSS		CGI-S	YGTSS			CGI-S	CGI-I	
	Motor	Vocal	Total		Motor	Vocal	Total		
Participant 1	17	15	32	5	16	10	26	4	3
Participant 2	10	12	22	4	6	8	14	3	1
Participant 3	12	8	20	4	14	8	22	4	5
Participant 4	14	13	27	5	8	5	13	3	1

Table 3. Baseline and Posttreatment Clinical Outcomes

Note. CGI-Severity scale: 1 = normal, not all ill; 2 = borderline mentally ill; 3 = mildly ill; 4 = moderately ill;

5 = markedly ill; 6 = severely ill; 7 = extremely ill CGI-Improvement: 1 = very much improved; 2 = much improved;

3 = improved 4 = minimally improved; 5 = no change;

6 = minimally worse; 7 = much worse; 8 = very much worse.

During treatment, a few challenges occurred. Andrew's most bothersome tics, which were more complex, did not occur during session. Andrew became irritated by his mother's use of verbal prompts to perform competing responses, as his younger siblings began to mimic her in his presence. This was temporarily resolved when his mother used nonverbal prompts instead. Of note: During half of the sessions, the patient's mother was near the computer but, due to the need to care for Andrew's younger siblings, did not join the session until the last 10 to 15 minutes. She had reported that she enjoyed joining the sessions at the end for a recap, as she felt it made her son feel more mature to not have a parent sitting over his shoulder while doing therapy.

Ben

Ben was a 13-year-old male with TS. He also met criteria for ADHD-Inattentive type, and his WASI-vocabulary *T*-Score was 43 (average). His YGTSS total severity score at baseline was 22, his tic-related impairment score fell in the mild range, and his CGI-S score was 4, indicating moderate symptoms. Four tics were targeted, including sniffing, a neck movement, holding his breath, and eye blinking/darting. Ben's tics were not particularly bothersome to him. His mother attributed this to a lack of awareness of the frequency with which he performed them; however, she perceived Ben's sniffing tic to be annoying, as it was frequent. Ben noted his tics were worse when he was stressed, bored, or tired. Allergies exacerbated his sniffing tic, and it was discovered that his tics occurred more often when they were being discussed. During the final assessment he received a YGTSS total tic score of 14, and endorsed no tic-related impairment. His CGI-S score was 3 (*mildly ill*) and his CGI-I score was 1 (*very much improved*).

Due to the mild and infrequent nature of the tics, performing and conveying the importance of function-based assessment and intervention was difficult. During sessions, Ben sat at the computer with his mother or father sitting behind him in the background.

Craig

Craig was an 11-year-old male who resided with his parents, twin sister, and older sister. He met criteria for TS based on the MINI-Kid interview, and reported a history of reading difficulties. His WASI-vocabulary score was 52 (average). His YGTSS total tic severity score was 20, his tic-related impairment fell in the minimal range, and his CGI-S score was 4, indicating moderate illness severity. He reported some tic-related impairment in the form of personal annoyance from having tics and receiving unwanted questions from peers. Craig entered treatment with several tics, including jaw movements, turning his head upwards, skipping then tapping his hip with his palm, shoulder rolling, throat clearing, sniffing with a nose scrunch, and spinning his whole body to either side. He indicated that head-turning upwards happened more often when outside on sunny days, and the skipping-tapping tic occurred more frequently when he was inside his home, walking around or washing his hands. At the postassessment his YGTSS total tic score increased to 22, and his tic-related impairment score remained in the minimal range throughout the course of treatment. He received a CGI-S score of 4 (*moderately ill*), and a CGI-I score of 5 (*no change*).

Several issues occurred in treatment. Homework compliance was fairly poor. Also, Craig often fidgeted and had a playful demeanor in sessions. In the initial sessions he played with the web camera frequently, rotating it so that he and his mother shifted in and out of viewing range, and turning the video feed on and off. This behavior steadily decreased, especially after he was asked to bring a fidget toy to session. He also enjoyed using the Skype instant-messaging feature randomly to chat during sessions, which was somewhat distracting. Treatment ended after the fourth session because of a virus on the family computer. The problem took several weeks to be properly fixed, then the family was lost to contact for several more weeks.

David

David was a 10-year-old male, residing with his parents, twin brother, and older brother. He met criteria for TS, based on the MINI-Kid interview, and his parents also indicated he had mild anxiety. His WASI-vocabulary score was 56 (average). At baseline his YGTSS total tic severity score was 27, and his tic-related impairment was moderate. He received a CGI-S score of 5. His tics, which occurred most often during school hours, included a throat noise, head nod, hard wink, quick breath, and squeak. His head nod caused physical pain and interrupted activities, his vocal tics interfered with speaking, and he received unwanted attention and questions about his tics from peers. Overall, most tics were reduced from the beginning of treatment, with the exception of the quick breath tic. David also reported less distress and fewer tic hassles. At the postassessment, his YGTSS total tic score had decreased to 13, and he endorsed no tic-related impairment. His CGI-S score was 3 (*mildly ill*), and his CGI-I score was 1 (*very much improved*).

Treatment raised some issues. It was difficult to hear David's vocal tics over VoIP, making awareness training and competing response training challenging. However, his mother helped to identify tic occurrence during awareness training, and prompted and praised David's use of physically incompatible responses during competing response training. Additionally, David's siblings occasionally interrupted sessions, as they were curious about what was happening. During one session the family conducted the session from David's bedroom, which was unusual. Throughout treatment, David attended sessions with his mother or both parents.

Therapeutic Alliance and Acceptability

Averaged across all four participants, the mean patient rating for the Child's Perception of Therapeutic Relationship scale was 32.5 out of 40 (SD = 1.73; range = 31-35), suggesting a strong therapeutic alliance and was similar to that found among patients receiving VC-delivered CBIT (<u>Himle et al., 2012</u>). The mean parent treatment acceptability (TAQ) rating was high, with an average of 39 out of 42 (SD = 2.94; range = 35-42), and was also similar to the mean parent TAQ rating (35.5) found among those receiving VC-delivered CBIT (Himle et al.). Parental treatment satisfaction (CSQ) ratings were strong, with an average of 29.25/32 (SD = 2.75; range = 26-32), as were child treatment satisfaction (CSQ) ratings (M = 30.75/32; SD = 1.50; range = 29-32). In regard to satisfaction with VC, the mean patient and parent ratings were high, with respective scores of 73 (SD = 5.35; range = 68-79), and 74.25 (SD = 5.74; range = 67-81) out of 84.

VoIP Call Quality

In 71% of the 28 total treatment sessions across participants, no technical difficulties occurred. "Insignificant" technical difficulties occurred in 17.9% of sessions, "Minimal" technical difficulties occurred in 7.1% of sessions, and "Moderate" difficulties occurred in 3.6% of sessions. In regard to type of technical difficulties, 21.4% were related to sound quality (excluding complete inability to hear sound), and 10.7% were related to video quality (excluding complete inability to see video feed). In no treatment sessions were staff completely unable to see video feed or hear sound.

Discussion

In Study 1, the interrater agreement between in-person and VoIP administrations of the YGTSS indicated high reliability between modalities. In Study 2, the preliminary efficacy, feasibility, and acceptability of VoIP-delivered CBIT were assessed. Results showed a 29.44% average decrease in YGTSS tic severity from pre to post, which is similar to the 31% decrease found in the original CBIT trial in children (Piacentini et al., 2010) and in line with the respective ratings of 33% and 27% found in both VC and in-person modalities in the randomized comparison by Himle et al. (2012). Two of the four participants (50%) were classified as treatment responders at posttreatment. This is similar to the response rate (52.5%) in CBIT delivered face-to-face (Piacentini et al.); however, as this was a case series with a small sample size, comparisons should be made cautiously. The

therapeutic alliance and patient and parent satisfaction ratings were high, and the intervention was feasible with respect to technical issues, as 71% of therapy sessions were free of technical difficulties, and 75% of assessment sessions had "none" or "insignificant" technical difficulties.

Despite the promising findings, several notable issues emerged during the VoIP delivery of CBIT. First, one participant's family computer was infected with a virus just prior to the fifth treatment session. After taking the computer to be fixed, the family was still unable to use Skype and needed to take the computer in a second time. Following this, the family was lost to contact for several weeks, and treatment was not reinitiated. Second, variable sound quality occasionally interfered with treatment delivery, especially during awareness training, competing response training, and relaxation training. During awareness training, it was sometimes challenging to bring certain tics to the child's attention because they were more difficult to hear. Third, child engagement and sustained attention was occasionally difficult to manage during sessions. If a child is off-task during a session, the clinician may feel less control over the situation over VoIP than in a face-to-face session. Although this could suggest externalizing diagnoses like ADHD may moderate treatment response, of the two participants who had a comorbid diagnosis of ADHD, one was classified as a responder, with a rating of "very much improved" at posttreatment, and the other was not but still received a rating of "improved."

A fourth issue involved the review of homework at each session. Parents and patients read aloud selfmonitoring homework to the therapist at the beginning of each session, and the therapist took notes. This is in contrast to face-to-face sessions, in which the therapist is able to view homework firsthand. A fifth issue involved the restricted range of view of the patient as a result of the equipment setup (i.e., the computer placed on top of a table, desk, or cart). In contrast, traditional VC formats using dedicated VC systems allow for full view of the body or remote control of movable cameras. In the current VoIP model, therapists only saw the patient's face and top half of their body. This made addressing tics involving the lower extremities more challenging. A related difficulty involved eye contact. For one therapist, giving the appearance of making eye contact with the patient often required looking into the web camera instead of at the computer screen; however, doing so made it challenging for the therapist to see what was on screen.

A sixth issue was reduced parental participation during sessions in some cases. Despite parents being present for all sessions, their participation levels varied. Reasons are unclear, but it is possible that the nature of in-home VC-delivered treatment allows parents to multitask during sessions. Additionally, receiving treatment in the home can invite interruptions by other family members.

Based on these issues, the following are suggestions for improving treatment quality for home-based VC. For therapists who need to track weekly progress using a self-report symptom questionnaire, it may be helpful to have parents return such documents as email attachments in order to preserve document formatting for easy viewing and scoring. To enhance awareness training and competing response training, it may be beneficial to utilize parents to identify tics during sessions, so the therapist can call them to the child's attention and remind the child to use the competing response. To address the problem of restricted view of tics, it may be helpful to begin treating a facial tic or other tic involving the upper body first, so the child develops a routine before working on a tic involving the lower extremities. Asking the child to back up from the computer to observe tics or to see if the competing response is being performed correctly can also be helpful. It is also important to remind parents to sit in front of the computer with their child during sessions and avoid multitasking, especially if their child is young. This should also aid children who have difficulty sustaining attention.

Solving eye contact issues is challenging, but it may be helpful if the therapist sits in a chair that allows their eyes to be aligned with or above the web camera so that looking down at the screen will be less noticeable, or sits far enough away from the camera, thus decreasing the ability of the patient to discriminate subtle differences in direction of gaze. If the therapist is unable to have such a setup then they should focus on viewing the patients

on screen, and only occasionally look up into the web camera. Explaining this issue to the patient, prior to treatment, would be beneficial.

The study also raises some ethical considerations. Delivering treatment while patients are in a home setting introduces potential privacy issues with respect to family members interrupting sessions and hearing content. Additionally, maintaining Internet network security and privacy during VoIP sessions is a potential concern. Prior to study participation, families were made aware of these issues in the consent process, and received VC guidelines to address these concerns (e.g., conducting sessions in a computer room or room with closeable doors, logging off from Skype following each session, using a firewall or antivirus program, etc.); however, these issues do remain limitations. Furthermore, a general concern regarding distance treatments involves the handling of psychiatric emergencies. In the present study, no psychiatric emergencies occurred, and this issue was addressed preemptively by having families provide contact information and sign a release of information for a local health care provider in the event of an emergency prior to beginning treatment.

Other general design limitations also warrant mention. One major limitation involves the open trial design. A second is that this was a pilot study with four subjects, limiting the generality of our conclusions. Another limitation is that one of the six families phone screened for eligibility declined participation because they deemed the treatment modality unacceptable. Also, the need for a computer and high-speed Internet access may be considered a limitation, as it may be perceived as displaying a bias towards families of higher socioeconomic status. Finally, we did not include a follow-up assessment so we are unable to make conclusions regarding maintenance of gains.

Generally, web-based CBIT was effective and feasible and has the potential to further increase access to behavior therapy for families. Despite the positive outcomes, use of VoIP for therapy purposes may pose some logistical barriers with respect to policy issues for some clinics, depending on the institutional setting. Additionally, use of VoIP-delivered treatment has not been addressed within most states' licensing laws. Future studies should assess the efficacy of web-based VC delivery of CBIT relative to a waitlist or active control group. It would also be interesting to compare it to traditional VC delivery and face-to-face delivery in terms of efficacy and acceptability.

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