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An Initial Case Series of Intensive Cognitive–Behavioral Therapy for Obsessive–Compulsive Disorder in Adolescents with Autism Spectrum Disorder

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Abstract Obsessive-compulsive disorder (OCD) is prevalent among youth with autism spectrum disorder (ASD). Cognitive-behavioral therapy (CBT) with ASD-specific modifications has support for treating OCD in this population; however, use of intensive CBT in youth with ASD and severe OCD has not been tested. The current study examined the preliminary effectiveness of an individualized intensive CBT protocol for OCD in adolescents with ASD. Nine adolescents (aged 11-17 years) completed a regimen of intensive CBT (range 24-80 daily sessions) incorporating exposure with response prevention (ERP). Treatment materials, language and techniques were modified in accordance with evidence-based findings for this population. Seven of nine participants (78%) were treatment responders, and large treatment effects (d = 1.35 - 2.58) were obtained on primary outcomes (e.g., obsessive-compulsive

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symptom severity). Preliminary findings suggest that an intensive CBT approach for OCD is effective among adolescents with ASD.

Keywords Obsessive–compulsive disorder · Autism spectrum disorders · Cognitive–behavioral therapy · Intensive treatment · Adolescents

Introduction

Autism spectrum disorder (ASD), is characterized by impairments in social interaction and communication, as well as restrictive and/or repetitive patterns of behaviors and interests [1]. Relatively high prevalence rates have been observed among school-age youth in the United States, where ASD occurs in 1 in 68 [2], and the core ASD symptoms cause significant impairment across multiple domains of child functioning [3, 4].

Comorbid psychiatric conditions are common in youth with ASD, including attention-deficit/hyperactivity disorder, disruptive behavior disorders, and anxiety disorders [5], which exacerbate functional impairment beyond that attributed to core ASD symptoms [5, 6]. Obsessive-compulsive disorder (OCD), in particular, is commonly comorbid with ASD, with caseness occurring in approximately 37% of youth with ASD [7]. When OCD and ASD cooccur, overall impairment in school, peer relationships, and family domains frequently exceeds that reported among youth with either condition alone, as observed in anxiety disorders comorbid with ASD [8]. Moreover, youth with OCD and ASD exhibit higher rates of comorbidity with other anxiety disorders than do youth with OCD alone [9]. Cognitive-behavioral therapy (CBT) with exposure and response prevention (ERP) is the first-line intervention for youth with OCD, demonstrating superiority to pharmacological treatment [10, 11]. When OCD is present with comorbid psychiatric disorders, treatment and prognosis are complicated, with research suggesting diminished treatment response [12, 13].

Cognitive and communication deficits, limited abstraction capability, and inflexibility among youth with ASD represent additional challenges to implementation of CBT programs [14]. Nevertheless, modified CBT protocols have demonstrated effectiveness in reducing anxiety symptoms among youth with high-functioning ASD [15]. These protocols incorporated ASD-specific modifications, such as including special interests [e.g., 16], visual supports [e.g., 17, 18] reduced cognitive loading, enhanced behavioral components, increased family involvement [e.g., 16, 17, 19], and developmentally modified treatment language e.g., [20]. In addition, treatment duration was often expanded to address deficits in social skills, restricted and/or stereotyped interests, poor attention and motivation, disruptive behavior and school-based issues-all problems commonly reported among anxious youth with ASD [19, 21, 22].

Specific to adolescents with ASD, several RCTs tested the effectiveness of CBT for anxiety disorders inclusive of OCD. White et al. [23] compared the effectiveness of a multimodal anxiety and social skills intervention (MASSI) with a waitlist control condition in a small RCT in 30 adolescents (12-17 years) with autism, Asperger's syndrome (AS) or pervasive developmental disorders (PDD-NOS). Intervention included individualized modules of psychoeducation, coping and problem solving; and group sessions of social skills training. Significant group differences were observed in social functioning (d=1.03), but not for anxiety symptoms. Storch et al. [19] examined a developmentally scaled version of the Behavioral Interventions for Anxiety in Children with Autism-BIACA-CBT protocol [21] in 31 adolescents with autism, AS or PDD-NOS (11–16 years) who were randomly assigned to 16 weekly CBT sessions or an equivalent duration of treatment as usual (TAU). Findings reflected significant differences favorable to CBT in reductions of symptom severity (d range 0.79–1.30), treatment response (68.8 vs. 26.7%) and remission status (27 vs. 0%). Wood et al. [22] also examined the effectiveness of the BIACA program in an RCT where 33 adolescents with autism, AS or PDD-NOS (11-15 years) were randomized to 16 CBT sessions or a 3-month waitlist control. Results showed significantly greater reductions in anxiety severity (d=0.74) and treatment response for CBT as compared to waitlist condition (79 vs. 28.6%).

Given the high incidence and impairment associated with OCD comorbid to ASD [5], as well as differences in phenomenology and treatment implications [9, 14] interest in evaluating OCD-specific CBT for this population has increased. Promising findings have been reported in case studies [20, 24–29], a case control study [30], a quasiexperimental study [31], and two small RCTs [32, 33]. As in studies of anxiety among youth with ASD, protocol modifications included increasing parental involvement [e.g., 26–28], adding reward programs, simplified cognitive therapy [e.g., 26, 27], and language [20], as well as incorporation of child interests and use of visual prompts and role playing [e.g., 20, 28].

In adolescents with high-functioning ASD, case reports have demonstrated benefit targeting contamination, harm, order, hoarding [20, 26] and catastrophic obsessions in an adolescent with AS [24]. Only one RCT has been conducted in a sample comprised of adolescents and adults with comorbid ASD and OCD. Russell et al. [32] randomized 46 participants (14-65 years) to CBT or anxiety management condition (psychoeducation, relaxation, healthy habits, and problem solving). No ASD-specific modifications were reported. Non-significant differences were observed between CBT and control conditions with respect to effect size (d=1.01 vs. d=0.6), and treatment responder (30 vs. 10%) and remitter rates (20 vs. 15%). Murray et al. [30] compared the effectiveness of a 14-session CBT protocol for OCD among youth with and without ASD (AS, high-functioning autism, and PDD-NOS). Treatment included psychoeducation, externalizing techniques, ERP, and relapse prevention; no modifications for ASD were reported. Mean percentages of symptom reduction on Children's Yale-Brown Obsessive-Compulsive Scale-CY-BOCS—[34] were significantly greater in the OCD group $(M=48.2\pm22.2\%)$ as compared to the OCD+ASD group $(M=33.3\pm16.9\%)$. Similarly, percentages of responders and remitters for OCD group (73 and 46%, respectively) were superior to those among the OCD+ASD group (46 and 9%, respectively) although not of statistical significance. These findings, using a standardized CBT approach not including ASD-specific modifications, indicate the importance of including adaptations in CBT programs addressing OCD in adolescents with ASD [30].

Overall, mixed findings observed across studies may suggest the more resistant nature of OCD [30, 32] and anxiety [23] when they co-occur with ASD. Moreover, although medication augmentation is indicated for severe pediatric OCD cases [35], no evidence-based pharmacological treatment strategies exist for addressing significant OCD in ASD. Intensive CBT is recommended for children with severe functional impairments, since daily sessions involving massed ERP may result in faster improvements [36], allowing youth to rapidly resume school and normal activities. Modified weekly approaches for OCD are associated with promising findings among adolescents with ASD [e.g., 17, 19, 22, 26]. Nevertheless, access to CBT programs is often hampered by the availability of trained providers [37], particularly when ASD-specific modifications are required. An intensive approach for these interventions facilitates treatment access by allowing families to relocate for a shorter period of time than with weekly CBT. To the best of the authors' knowledge, CBT delivered in an intensive format has not been evaluated in youth with high functioning ASD and comorbid OCD. In addition, previous studies on OCD-specific CBT in patients with ASD have focused predominantly on childhood [25, 27-29] or adult samples [31, 32]. Examining OCD-specific intensive CBT in adolescents with ASD is of particular importance, since adolescence is a developmental period with idiosyncratic characteristics, and additional adaptations may be necessary to address their clinical needs; for example, materials should be practical and focused on relevant issues for adolescents, as well as respect emerging independence [19]. Also, adolescence is characterized by heightened vulnerability to internalizing problems [38, 39] which is increased in individuals with ASD [40].

In light of the potential benefits for adolescents with OCD and ASD, we present a case series of nine adolescents with significant OCD who participated in multimodal, CBT-based intensive outpatient (i.e., 3 h daily, 5 days/week) or partial hospitalization (6.5 h daily, 5 days/week) programs for OCD. We hypothesized that treatment would be associated with reduced OCD symptom severity and associate functional impairment. Also, recent meta-analysis observed that CBT produced significant improvements in secondary outcomes such as anxiety and depression in neurotypical youth with OCD [41]. Thus, it is expected that intensive CBT will also be effective in improving anxiety and depression in youth with OCD and comorbid ASD.

Method

Participants

Archival data were collected from 9 adolescent patients (aged 11–17, $M=14\pm 2$ years, 89% male) who were consecutively treated at a specialized OCD treatment program who presented with high-functioning ASD and a primary psychiatric diagnosis of OCD (i.e., the reason for presentation). Participants were Caucasian (89%) or Hispanic (11%). All participants had a primary diagnosis of OCD in conjunction with ASD. At intake, diagnoses were derived through consensus procedure, which combined all available clinical information made during clinical interviews with a board certified child/adolescent psychiatrist and extensive clinical interactions with two experienced psychologists, as well as records review [42]. Each of the nine participants was reviewed by a caseness panel consisting of these three clinicians; there was 100% agreement related to diagnoses of high-functioning ASD and OCD as the primary psychiatric disorder according DSM-IV-TR criteria. Beyond diagnosis, other inclusion criteria included: (1) Primary OCD in conjunction with ASD diagnosis; (2) clinically significant OCD symptoms as evidenced by a score of 16 or greater on CY-BOCS; and (3) English speaking and able to read. Participants were excluded if they presented with lifetime history of psychosis or endorsed substance abuse.

Procedure

This study involving archival records review was approved by the institutional review boards at the University of South Florida and Rogers Memorial Hospital. Participants were families who had completed a regimen of intensive psychotherapy consisting of either a partial hospitalization (6.5 h of treatment per day) or intensive outpatient program (3 h of treatment per day) at a multidisciplinary behavioral health center specializing in the treatment of OCD/anxiety disorders in youth with and without ASD. Pre-treatment measures were administered within 3 days of admission to treatment program. Post-treatment measures were administered on the final day of treatment. Clinician-rated measures were administered by unblinded trained graduatelevel evaluators familiar with the patient and family being treated.

Measures

Children's Yale-Brown Obsessive–Compulsive Scale— CY-BOCS—[34]

The CY-BOCS is a semi-structured clinician-administered interview assessing symptoms and severity for OCD in youth in the past week. The Symptom Checklist assesses current or past presence of 62 common obsessions and compulsions. The 10-item Severity Scale assesses duration, interference, distress, resistance, and control perceived for obsessions and compulsions. The CY-BOCS is the gold standard for assessment of pediatric OCD and demonstrates good psychometric properties in clinical samples in neuro-typical youth with OCD [43, 44], as well as among youth with OCD and ASD [45].

Columbia Impairment Scale—Parent & Child versions— CIS-C/P—[46]

The CIS is a psychometrically-sound 13-item self- and parent-report measure assessing multidimensional impairment in youth. Items are rated on a 5-point Likert scale from 0 (*no problem*) to 4 (*very bad problem*) according to experiences within the past week.

Pediatric Quality of Life Enjoyment and Satisfaction Questionnaire—PQ-LES-Q- [47]

This 15-item self-report measures quality of life in youth from 6 to 17 years across several domains over the past week. Items are rated from 1 (*very poor*) to 5 (*very good*); with higher total scores indicate greater quality of life. The PQ-LES-Q has demonstrated adequate psychometric properties [47].

Screen for Child Anxiety Related Disorders—SCARED— [48]

The SCARED is a 41-item self-report assessing anxiety symptoms experienced in the last 3 months in youth from 8 to 18 years. Items are rated using a 3-point Likert scale from 0 (not true or hardly ever true) to 2 (very true or often true). A total score and separate scores for Somatic Symptoms/Panic Disorder, Generalized Anxiety Disorder, Separation Anxiety, Social Phobia, and School Avoidance. Parallel child- or parent-report versions are available. Instructions were revised for the SCARED such that anxiety symptoms were rated at posttreatment based on the past 2 weeks. Psychometric properties are strong in children with anxiety disorders [48] and youth with high-functioning ASD [49].

Pediatric Item Bank for Depression of the Patient-Reported Outcomes Measurement Information System— PROMISTM—[50]

The pediatric PROMIS was developed to assess several health domains for youth aged 8–18 years. The generic health domains include physical function, pain, fatigue, emotional distress, and social function. Within the emotional distress domain, the PROMIS pediatric 14-item bank for depressive symptoms assesses negative mood, anhedonia, negative perceptions about self and negative social cognition in the past 7 days. The PROMIS was validated by Irwin et al. [50], finding good psychometric properties.

Clinical Global Impression-Severity—CGI-S—[51]

The CGI-S is a single-item clinician-rating scale assessing anxiety-related symptom severity and associated impairment. Severity ratings range from 0 (*no illness*) to 6 (*extremely severe*). It is one of the most used measures in treatment studies in children and adolescents with OCD [52].

Treatment

All participants completed a regimen of intensive Cognitive-behavioral therapy (CBT) incorporating exposure with response prevention (ERP) within either an intensive outpatient program (3 h a day of ERP, 5 days a week) or partial hospitalization program [6.5 h of treatment per day (4–5 h of ERP per day), 5 days a week]. Due to the nature of presenting symptoms (i.e., severity) and other environmental factors (i.e., family dynamics, financial considerations, geographic locale, etc.), duration of the treatment ranged from 24 to 80 days (M=46.5±20.9) across participants. Treatment rationale and techniques were the same for the two intensive modalities; there were no substantive differences in the protocol between modalities other than time. CY-BOCS severity at pretreatment was similar for participants in each program (M_{PHP} =31.5±8.5, M_{IOP} =32±4.2).

Consistent with standard care CBT with ERP for pediatric OCD, intensive treatment for all youth included psychoeducation, hierarchy development, ERP, homework (up to 60 min per day), and relapse prevention. During the initial day of treatment, children and their parent(s) were provided with psychoeducation about OCD and the Cognitive–behavioral treatment model. During the first and second days, a fear hierarchy was developed with exposures to relatively easy triggers occurring during the second day of treatment and becoming progressively more difficult over the course of treatment.

A number of modifications to this treatment approach were made to individualize care to youth with ASD. All procedures were tailored for use in youth with comorbid ASD based on best practice and research evidence [19, 21]. First, increased attention was devoted to affective education; specifically, assisting youth with identifying physiological cues associated with anxiety (i.e., shallow rapid breathing, discomfort in various areas of the body, etc.), and with techniques to determine environmental triggers for said cues. This served a dual purpose, wherein participants received assistance with increasing their experiential understanding of anxiety, and information was gathered throughout the affective education process to further inform other aspects of treatment (i.e., hierarchy generation, coping skill-building, etc.). Second, therapy emphasized concrete therapeutic exercises (i.e., in vivo exposure activities) while reducing focus on cognitive techniques (i.e., thought challenging, Socratic questioning, etc.). Of particular importance to this point, exposure activities incorporated age-appropriate self-care (i.e., personal and sleep hygiene, organization, etc.) where possible. Deficits in self-care and organizational adaptive skills are commonly observed in individuals with ASD [e.g., 53]. These deficits may negatively affect youth in coping with feared situations further increasing anxiety and failure experiences [21]. Third, the treatment planning process was deliberately expanded to incorporate individual participants' restricted and/or stereotyped interests.

Beyond individualization, the treatment process also included modifications targeting more global barriers to treatment common to youth with ASD. First, parent/caregiver participation was increased; specifically, additional psychoeducation, behavioral management training, problem solving training, and modeling/assistance with reward system use were provided. Second, treatment metaphors and literature (handouts) were modified to use developmentally appropriate language. Third, manipulatives were used to reduce abstraction of vital treatment components (i.e., use of "Feelings Thermometer" with developmentally scaled markers in place of traditional subjective units of distress scale (SUDS), simple charting system to track purpose, steps and completion of behavioral exposure tasks, etc.). Fourth, appropriate problem solving and coping steps were explained and modeled in a simplified and repetitive fashion so as to maximize retention and skill-building.

For all participants, treatment concluded with activities and education related to relapse prevention, transfer of care to appropriately trained outpatient providers, and relapse prevention for patients and their families. Treatment was delivered by graduate (masters and doctoral) level therapists with at least 1 year of experience working with youth. Regular clinical supervision with licensed clinical psychologists was provided.

Data Analysis

Primary and secondary outcome measure scores were computed, presenting data separately for each child on primary outcomes. To test treatment differences at pre- and posttreatment we used the nonparametric Wilcoxon signed rank test.

Results

Characteristics of Participants

Table 1 presents participant demographics, diagnoses, pharmacological treatments, and scores in primary outcome variables. Pretreatment mean scores for CY-BOCS total (31.6 ± 7.5), Obsession (15.2 ± 4.4), and Compulsion (16.4 ± 3.2) appear in Table 2 below. At admission, OCD-related impairment as measured by the CGI-Severity included two (22%) participants with extremely severe symptoms, six (67%) with severe symptoms, and one (11%) with moderate-severe symptom severity. Regarding obsessive-compulsive symptoms, all participants endorsed multiple obsessions and compulsions. Most common obsessive symptoms were aggressive obsessions (e.g. fear harm will come to self or to others) endorsed by seven participants (78%), followed by miscellaneous obsessions (e.g. intrusive sounds or words) by six participants (67%), magical thoughts and superstitious (e.g., unlucky numbers or words, being turned into someone else, etc.), and religious obsessions (e.g., fear of offending religious object) by five participants (56%), contamination obsessions (e.g., concerns with dirt, germs, or illnesses) by four participants (44%), and sexual obsessions (e.g., content involves homosexuality), hoarding obsessions (e.g., fear of losing possessions), and somatic obsessions (e.g., concern about aspect of certain body part) by two participants (22%). Within compulsive and ritualistic behavior, checking (e.g., checking that did not harm others), repeating (e.g., rereading, erasing and rewriting), miscellaneous compulsions (e.g., need to ask or confess), and rituals involving other people (e.g., reassurance seeking) were endorsed by six participants (67%), washing compulsions (e.g., excessive and ritualized handwashing), counting (e.g., counts backwards) and magical games (e.g., stepping over certain spots on the floor) by five participants (56%), and ordering compulsions (e.g., need for symmetry) by three participants (33%).

With respect to specific social and academic impairment at baseline, only two participants (22%) were enrolled in public school at time of admission. Of the remaining seven participants, three (43%) had been expelled from their assigned school-whether due to anxiety-based behaviors, or severity of ASD-related behaviors-within 30 days prior to admission; three (43%) were being home-schooled by family members, and one (14%) had withdrawn from their assigned school more than one year prior to treatment admission. Five participants (56%) received previous interventions with incomplete response. These included medication regimen in 3 cases (33%), weekly CBT in 3 cases (33%), daily 1-hour CBT in 1 case (11%), daily intensive 3-hour CBT in one case (11%), bibliotherapy in one case (11%), talk therapy in one case (11%), and religious counseling in one case (11%).

Primary Outcome Measures

Table 2 presents means and standard deviations for outcome measures and Wilcoxon signed rank test. Significant decreases were observed from pre- to post-treatment on the CY-BOCS total (14.1±8.0, d=1.86), Obsession (6.0±5.0, d=1.35), and Compulsion scores (8.1±4.1, d=2.50), as well as for the CGI-Severity (1.6±1.0, d=2.58). Mean change scores in primary outcomes for each intensive modality were 13.0±8.9 for CY-BOCS total and 1.6±1.1 for the CGI-Severity in PHP program (n=7), and 18.0±1.5 for CY-BOCS total and 1.5±0.7 for the CGI-Severity in IOP program (n=2).

1Female, 11 years old ASD, OCD, UMD, UAD2Male, 12 years old ASD,OCD,GAD, UTD3Male, 13 years old ASD,OCD,GAD, UTD4Male, 13 years old ASD, OCD, MDD5Male, 13 years old ASD, OCD, UMD6Male, 16 years old ASD, OCD, UMD		(treatment hours)									
				Total		Obses- sion	Com sion	Compul- sion	I		
				Pre P	Post I	Pre Post	st Pre	e Post	t Pre	Post	t -
		PHP (266.5)	NA	17 8		7 5	10	ŝ	4	б	Remitter
		PHP (234)	NA	38 8		19 5	19	\mathfrak{c}	9	7	Remitter
		PHP (156)	Pre: clomipramine, divalproex sodium, hydrox- yzine pamoate, lurasidone Post: clomipramine	39 2	24]	19 12	20	12	ŝ	4	Responder
		PHP (487.5)	Pre: fluvoxamine Post: lorazepam, clomipramine, fluoxetine	29 2	25]	14 11	15	14	5	4	Non responder
		PHP (520)	Pre: sertraline Post: aripiprazole, guanfacine, sertraline	39 2	24	20 10	19	14	5	ю	Responder
		PHP (344.5)	NA	35 2	21 1	16 12	19	6	9	5	Responder
7 Male, 16 years old ASD, OCD, MDD		PHP (364)	Pre: aripiprazole, escitalopram, hydroxyzine pamoate Post: aripiprazole, fluoxetine	24 2	20	10 13	17	7	ŝ	4	Responder
8 Male, 17 years old A	Male, 17 years old ASD, OCD, UAD, ADHD IOF (87)	IOP (87)	NA	29 1	10	14 5	15	5	5	4	Responder
9 Male, 14 years old ASD, OCD, GAD		IOP (75)	NA	35 1	18	18 10	17	×	5	б	Responder

 Table 1
 Participant demographics, diagnoses, and scores in primary outcome variables

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 Table 2
 Means, standard

 deviations and results for
 Wilcoxon signed rank test in

 outcome measures
 Notes and test in

Outcome	Pretreatment Mean (SD)	Posttreatment Mean (SD)	Ζ	р
CY-BOCS Total $(n=9)$	31.67 (7.57)	17.56 (7.04)	-2.67	0.008
CY-BOCS Obsessions $(n=9)$	15.22 (4.44)	9.22 (3.31)	-2.37	0.018
CY-BOCS Compulsions $(n=9)$	16.44 (3.24)	8.33 (4.30)	-2.67	0.008
CGI-S $(n=9)$	5.11 (0.60)	3.56 (0.88)	-2.75	0.006
CIS Child $(n=6)$	23.00 (4.82)	13.33 (6.50)	-2.20	0.027
CIS Parent $(n=6)$	30.00 (7.72)	23.33 (6.31)	-2.02	0.043
PQ-LES-Q $(n=6)$	46.83 (8.49)	54.33 (9.67)	2.00	0.046
SCARED Child $(n=6)$	39.00 (10.41)	28.50 (15.00)	-2.02	0.043
SCARED Parent $(n=6)$	45.66 (16.17)	36.16 (13.83)	-2.20	0.028
PROMIS Child $(n=9)$	40.56 (13.26)	29.56 (9.76)	-2.38	0.017

CY-BOCS Children's Yale-Brown Obsessive–Compulsive Scale, *CGI-S* Clinical Global Impression–Severity, *CIS* Columbia Impairment Scale, *PQ-LES-Q* pediatric quality of life enjoyment and satisfaction questionnaire, *SCARED* screen for child anxiety related disorders, *PROMIS* Pediatric Item Bank for Depression of the Patient-Reported Outcomes Measurement Information System

To allow comparisons with previous studies, we used a CY-BOCS score reduction of 35% or greater, and a CY-BOCS total score of 12 or less, to define treatment response and remission, respectively [54]. At post-treatment seven (78%) participants met criteria for responder status and three (33%) achieved remission.

Secondary Outcome Measures

Results from secondary measures are also shown in Table 2. Functional impairment on CIS, decreased significantly for child (9.7 ± 7.1) and parent-report (6.7 ± 5.3) . For quality of life, the PQ-LES-Q score showed significant increase (7.5 ± 6.9) . Significant decreases were observed on the SCARED reported by parents (10.5 ± 11.9) and children (9.5 ± 5.6) . Depressive symptoms showed significant reductions on the PROMIS (11.0 ± 9.2) .

In terms of social and academic impairment, of the seven participants who were not enrolled in public school at baseline: one of the three previously expelled participants was successfully enrolled and reintegrated into a public school, one was enrolled in a private/therapeutic school, and the third was homeschooled by family members; two of the three participants being homeschooled at admission remained in homeschooling upon discharge from treatment, and the third was enrolled and reintegrated into a public school; the participant who had been withdrawn from school for more than a year was successfully reintegrated into a private/therapeutic school. Both of the participants who had been in public school prior to treatment successfully returned to their assigned school upon discharge from treatment.

Discussion

These results provide preliminary support for the effectiveness of personalized intensive CBT for adolescents with ASD and comorbid OCD who have severe symptoms and/ or have been incomplete responders to past treatment. Consistent with previous studies treating OCD in youth with ASD [e.g., 26, 30, 32] participants showed improvements on clinician-rated OCD measures with large effect sizes (d=1.35-2.58). It is worth highlighting the high level of symptom acuity with which almost all participants in the current study presented, having a high mean CY-BOCS score (31.6 ± 7.5) and 89% of the sample presenting with severe or extremely severe impairment ratings on the CGI-S.

At post-treatment, 78% of participants were classified as responders, a larger percentage than those reported in previous studies using the CY-BOCS reduction criterion. Russell et al. [32] reported 30% response rate including adults in the sample, and Murray et al. [30] reported a 46% response rate using a standard CBT protocol; neither included exclusively treatment resistant and/or severely ill patients. Similarly, the percentage of remitters was greater in our study (33%) than those reported by Russell et al. [32] and Murray et al. [30] (20% and 9%, respectively) and similar to the percentage of remitters (30%) in a previous study on intensive treatment for youth with severe OCD [36]. Greater severity of OCD symptoms may have allowed for greater opportunities for symptom decline. Also, in the current study, the intensive CBT program was adapted for use in adolescents with ASD and OCD, and included modifications to address issues unique to this population. For instance, affective education allowed youth to recognize physiological cues associated to anxiety, obsessive thoughts, and to link them to environmental triggers. In this way, standard procedures of OCD treatment (e.g., hierarchy development) could be implemented adequately. Likewise, incorporating restricted interests into treatment activities (e.g. movie characters, determined objects) facilitated treatment adherence. Finally, considerable family involvement was also included to guide the home-based treatment activities, reduce family accommodation and promote youth's engagement. Also to account for ASD-related difficulties, families were required to collaborate in hierarchy development, rating anxiety, etc. This, as well as, the extent of massed ERP training included in the intensive program could be possible explanations for results observed.

Nevertheless, seven participants did not achieve remission and one was classified as non-responder. We believe this may reflect the severe presentation of participating youth, as well as some data that youth with ASD and anxiety/OCD may experience more modest symptom reductions relative to youth without ASD [e.g., 55]. This may be due to limited insight and motivation, difficulties in recognizing and rating anxiety, and limited flexibility to change certain behaviors. Increasing insight into the disorder and treatment protocol through psychoeducation, and using reward programs may be effective in optimizing the intervention in these cases [27]. Also, considerable amount of exposure practice is needed to obtain optimal results in the treatment of severe OCD cases, given that ERP is the primary active ingredient of the intervention [56]. An intensive modality facilitates massed practice of ERP at sessions and as homework, may facilitate generalization of gains. Despite parents being trained to implement homework exposure tasks and to reduce accommodation, distress and disruptive behavior exhibited by youth may have challenged consistent application of therapeutic concepts/skills. Involving parents adequately to promote exposure practice and reduce family accommodation is an effective way of enhancing treatment efficacy [57]. Finally, the goal of treatment at the partial hospitalization and intensive outpatient levels tends to differ from more standard outpatient care in that the former focuses on achieving sufficient improvement to allow for return to day-to-day functioning with further improvements-and ideally remission-made in outpatient care.

Secondary measures showed reductions in functional impairment and improved quality of life which is consistent with others [19, 23, 24, 26]. These findings suggest that reduction in OCD symptoms fosters improved social functioning in adolescents with ASD. Significant reduction of self- and parent-report of anxiety and depression was also observed. Although other findings showed that self-reports of anxiety [19, 23, 32] and depressive symptoms [32] did not reflect significant differences relative to control groups in youth with comorbid ASD, it is possible that the intensive nature of treatment in the current study increases patient and family focus on the intervention process and changes in anxiety and mood symptoms, given the improvements observed in previous studies of intensive treatment for OCD in anxiety [e.g., 58] and depressive outcomes [e.g., 59].

There are several limitations in the current study. First, we did not include an active control condition. Second, the small number of mostly male participants who were attending an intensive treatment program limits generalizability. It will be important for future research to involve larger and more diverse samples. Third, treatment duration was individually determined and not the same for all participants. Further, we could not examine differences in treatment effects as a function of program status. On balance, the purpose of this report was to describe the preliminary effectiveness of intensive CBT in adolescents with ASD and severe comorbid OCD when individualized for the presenting youth. Fourth, medication and doses were not stable across the study period, so the relative contributions of CBT and pharmacotherapy cannot be disentangled. Fifth, we did not use a structured diagnostic measure at baseline. On balance, consensus procedure involving three experienced clinicians has advantages in terms of analyzing the complexity of psychiatric symptoms in youth with ASD [60]. Finally, the use of unblinded raters and open trial nature of this report (without follow-up) does not rule out varied confounding factors (e.g., passage of time, clinician bias, etc.). Thus, these findings require further examination in controlled settings, including a larger sample size. As in previous studies, participants in this study did not present marked intellectual or language impairments, and were judged to be in the low average or above level of intellectual functioning. Given the frequency of these deficits in ASD, future studies including participants with intellectual disability and/or language problems are necessary to test the effectiveness of CBT for OCD in these populations.

In sum, these preliminary results suggest that intensive CBT approach may be efficacious for adolescents with ASD who have severe OCD symptoms and/or have been incomplete responders to past OCD treatment. Further controlled examination of modified intensive CBT as an alternative intervention for youth with ASD and comorbid OCD is warranted, given the dearth of available evidence-based treatment options for youth who did not obtain complete benefits from weekly or medication approaches. Given this, one important implication of these findings is the need for dissemination of intensive modalities of CBT tailored for youth with OCD and ASD (and OCD alone). Yet, there are still important access barriers for this population in clinical practice such as expertise in both ASD and OCD, scarcity of trained mental health providers, and cost.

Summary

The current study examined the preliminary effectiveness of intensive CBT for OCD modified to be implemented in nine adolescents with ASD. Modifications included increased attention to affective education, reduced focus on cognitive techniques, incorporation of restricted interests of individual participants as rewards, and use of developmentally appropriate language, manipulatives, and visual cues. An intensive approach which is recommended for children with severe symptoms and impairments was associated with significant decreases in obsessive-compulsive symptoms and overall severity. Seventy-eight and 33% of participants were classified as responders and remitters respectively. Secondary outcomes demonstrated significant improvements in functional impairment, quality of life, anxiety and depressive symptoms. This report suggests that intensive CBT may serve as an augmentation approach for those that have not responded well to standard care.

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References

- 1. American Psychiatric Association (2013) Diagnostic and statistical manual of mental disorders, 5th edn. American Psychiatric Publishing, Washington DC
- Centers for Disease Control and Prevention. (2014) Prevalence of autism spectrum disorder among children aged 8 years: autism and developmental disabilities monitoring network. MMWR Surveill Summ 63:1–21
- Dean M, Kasari C, Shih W, Frankel F, Whitney R, Landa R et al (2014) The peer relationships of girls with ASD at school: comparison to boys and girls with and without ASD. J Child Psychol Psychiatry 55:1218–1225
- Smith LE, Greenberg JS, Mailick MR (2014) The family context of autism spectrum disorders: influence on the behavioral phenotype and quality of life. Child Adolesc Psychiatr Clin N Am 23:143–155
- Joshi G, Petty C, Wozniak J, Henin A, Fried R, Galdo M et al (2010) The heavy burden of psychiatric comorbidity in youth with autism spectrum disorders: a large comparative study of a psychiatrically referred population. J Autism Dev Disord 40:1361–1370
- Posserud M, Hysing M, Helland W, Gillberg C, Lundervold AJ (2016) Autism traits: the importance of "co-morbid" problems for impairment and contact with services. Data from the Bergen Child Study. Res Dev Disabil. doi:10.1016/j.ridd.2016.01.002
- Leyfer OT, Folstein SE, Bacalman S, Davis NO, Dinh E, Morgan J et al (2006) Comorbid psychiatric disorders in children with autism: interview development and rates of disorders. J Autism Dev Disord 36:849–861

- Bellini S (2004) Social skill deficits and anxiety in high-functioning adolescents with autism spectrum disorders. Focus Autism Other Dev Disabl 19:78–86
- Lewin AB, Wood JJ, Gunderson S, Murphy TK, Storch EA (2011) Phenomenology of comorbid autism spectrum and obsessive-compulsive disorders among children. J Dev Phys Disabil 23:543–553
- McGuire JF, Piacentini J, Lewin AB, Brennan EA, Murphy TK, Storch EA (2015) A meta-analysis of cognitive behavior therapy and medication for child obsessive-compulsive disorder: moderators of treatment effectiveness, response, and remission. Depress Anxiety 32:580–593
- Sánchez-Meca J, Rosa-Alcázar AI, Iniesta-Sepúlveda M, Rosa-Alcázar Á (2014) Differential efficacy of cognitive-behavioral therapy and pharmacological treatments for pediatric obsessive-compulsive disorder: a meta-analysis. J Anxiety Disord 28:31-44
- 12. Geller DA, Biederman J, Stewart SE, Mullin B, Farrell C, Wagner KD (2003) Impact of comorbidity on treatment response to paroxetine in pediatric obsessive-compulsive disorder: is the use of exclusion criteria empirically supported in randomized clinical trials? J Child Adolesc Psychopharmacol 13:19–29
- Storch EA, Merlo LJ, Larson MJ, Geffken GR, Lehmkuhl HD, Jacob ML (2008) Impact of comorbidity on cognitive-behavioral therapy response in pediatric obsessive-compulsive disorder. J Am Acad Child Adolesc Psychiatry 47:583–592
- Storch EA, Björgvinsson T, Riemann B, Lewin AB, Morales MJ, Murphy TK (2010) Factors associated with poor response in cognitive–behavioral therapy for pediatric obsessive–compulsive disorder. Bull Menninger Clin 74:167
- Ung D, Selles R, Small BJ, Storch EA (2015) A systematic review and meta-analysis of cognitive-behavioral therapy for anxiety in youth with high-functioning autism spectrum disorders. Child Psychiatry Hum Dev 46:533–547
- Sofronoff K, Attwood T, Hinton S (2005) A randomized controlled trial of a CBT intervention for anxiety in children with Asperger syndrome. J Child Psychol Psychiatry 46:1152–1160
- Reaven JA, Blakeley-Smith A, Nichols S, Dasari M, Flanigan E, Hepburn S (2009) Cognitive–behavioral group treatment for anxiety symptoms in children with high-functioning autism spectrum disorders a pilot study. Focus Autism Other Dev Disabl 24:27–37
- White SW, Albano AM, Johnson CR, Kasari C, Ollendick T, Klin A (2010) Development of a cognitive-behavioral intervention program to treat anxiety and social deficits in teens with high-functioning autism. Clin Child Fam Psychol Rev 13:77–90
- Storch EA, Lewin AB, Collier AB, Arnold E, De Nadai AS, Dane BF (2014) A randomized controlled trial of cognitive– behavioral therapy versus treatment as usual for adolescents with autism spectrum disorders and comorbid anxiety. Depress Anxiety 32:174–181
- Sze KM, Wood JJ (2007) Cognitive behavioral treatment of comorbid anxiety disorders and social difficulties in children with high-functioning autism: a case report. J Contemp Psychother 37:133–143
- Wood JJ, Drahota A, Sze K, Har K, Chiu A, Langer DA (2009) Cognitive behavioral therapy for anxiety in children with autism spectrum disorders: a randomized, controlled trial. J Child Psychol Psychiatry 50:224–234
- 22. Wood JJ, Ehrenreich-May J, Alessandri M, Fujii C, Renno P, Laugeson E (2015) Cognitive behavioral therapy for early adolescents with autism spectrum disorders and clinical anxiety: a randomized, controlled trial. Behav Ther 46:7–19
- White SW, Ollendick T, Albano AM, Oswald D, Johnson C, Southam-Gerow MA (2013) Randomized controlled trial:

- El-Ghoroury NH, Krackow E (2011) A developmental–behavioral approach to outpatient psychotherapy with children with autism spectrum disorders. J Contemp Psychother 41:11–17
- Elliott SJ, Fitzsimons L (2014) Modified CBT for treatment of OCD in a 7-year-old boy with ASD—a case report. J Child Adolesc Psychiatr Nurs 27:156–159
- Lehmkuhl HD, Storch EA, Bodfish JW, Geffken GR (2008) Brief report: exposure and response prevention for obsessive compulsive disorder in a 12-year-old with autism. J Autism Dev Disord 38:977–981
- 27. Nadeau JM, Arnold EB, Storch EA, Lewin AB (2014) Family cognitive-behavioral treatment for a child with autism and comorbid obsessive compulsive disorder. [Clin Case Stud 13:22-36
- Reaven J, Hepburn S (2003) Cognitive–behavioral treatment of obsessive–compulsive disorder in a child with asperger syndrome:a case report. Autism 7:145–164
- Rooney M, Alfano CA, Walsh KS, Parr AF (2011) Differential diagnosis and treatment of obsessive–compulsive, inattentive, and sleep symptoms in a 7-year-old with PDD-NOS. Clin Case Stud 10:133–146
- 30. Murray K, Jassi A, Mataix-Cols D, Barrow F, Krebs G (2015) Outcomes of cognitive behaviour therapy for obsessive–compulsive disorder in young people with and without autism spectrum disorders: a case controlled study. Psychiatry Res 228:8–13
- Russell AJ, Mataix-Cols D, Anson MAW, Murphy DGM (2009) Psychological treatment for obsessive-compulsive disorder in people with autism spectrum disorders-a pilot study. Psychother Psychosom 78:59–61
- Russell AJ, Jassi A, Fullana MA, Mack H, Johnston K, Heyman I, Mataix-Cols D (2013) Cognitive behavior therapy for comorbid obsessive–compulsive disorder in high-functioning autism spectrum disorders: a randomized controlled trial. Depress Anxiety 30:697–708
- 33. Vause T, Neil N, Jaksic H, Jackiewicz G, Feldman M (2015) Preliminary randomized trial of function-based cognitive-behavioral therapy to treat obsessive compulsive behavior in children with autism spectrum disorder. Focus Autism Other Dev Disabl. doi:10.1177/1088357615588517
- Scahill L, Riddle MA, McSwiggin-Hardin M, Ort SI, King RA, Goodman WK (1997) Children's Yale-Brown obsessive compulsive scale: reliability and validity. J Am Acad Child Adolesc Psychiatry 36:844–852
- AACAP (2012) Practice parameter for the assessment and treatment of children and adolescents with obsessive–compulsive disorder. J Am Acad Child Adolesc Psychiatry 51:98–113
- 36. Storch EA, Bagner DM, Geffken GR, Adkins JW, Murphy TK, Goodman WK (2007) Sequential cognitive–behavioral therapy for children with obsessive–compulsive disorder with an inadequate medication response: a case series of five patients. Depress Anxiety 24:375–381
- Goodwin R, Koenen KC, Hellman F, Guardino M, Struening E (2002) Helpseeking and access to mental health treatment for obsessive-compulsive disorder. Acta Psychiatr Scand 106:143–149
- Kessler RC, Avenevoli S, Costello EJ, Georgiades K, Green JG, Gruber MJ et al (2012) Prevalence, persistence, and sociodemographic correlates of DSM-IV disorders in the National Comorbidity Survey Replication Adolescent Supplement. Arch Gen Psychiatry 69:372–380
- Kessler RC, Berglund P, Demler O, Jin R, Merikangas KR, Walters EE (2005) Lifetime prevalence and age-of-onset distributions of DSM-IV disorders in the National Comorbidity Survey Replication. Arch Gen Psychiatry 62:593–602

- Oswald TM, Winter-Messiers MA, Gibson B, Schmidt AM, Herr CM, Solomon M (2016) Sex differences in internalizing problems during adolescence in autism spectrum disorder. J Autism Dev Disord 46:624–636
- Rosa-Alcázar AI, Sánchez-Meca J, Rosa-Alcázar Á, Iniesta-Sepúlveda M, Olivares-Rodríguez J, Parada-Navas JL (2015) Psychological treatment of obsessive-compulsive disorder in children and adolescents: a meta-analysis. Span J Psychol 18:e20
- 42. Leckman JF, Sholomskas D, Thompson D, Belanger A, Weissman MM (1982) Best estimate of lifetime psychiatric diagnosis: a methodological study. Arch Gen Psychiatry 39:879
- Iniesta-Sepúlveda M, Rosa-Alcázar AI, Rosa-Alcázar Á, Storch EA (2014) Evidence-based assessment in children and adolescents with obsessive-compulsive disorder. J Child Fam Stud 23:1455–1470
- 44. Storch EA, Murphy TK, Geffken GR, Soto O, Sajid M, Allen P et al (2004) Psychometric evaluation of the Children's Yale– Brown obsessive–Compulsive Scale. Psychiatry Res 129:91–98
- 45. Wu MS, McGuire J, Arnold E, Lewin A, Murphy T, Storch EA (2014) Psychometric properties of the Children's Yale-Brown Obsessive Compulsive Scale in Youth with Autism Spectrum Disorders and obsessive–Compulsive Symptoms. Child Psychiatry Hum Dev 45:201–211
- 46. Bird HR, Andrews H, Schwab-Stone M, Goodman S, Dulcan M, Richters J et al (1996) Global measures of impairment for epidemiologic and clinical use with children and adolescents. Int J Methods Psychiatr Res 6:295–307
- Endicott J, Nee J, Yang R, Wohlberg C (2006) Pediatric quality of life enjoyment and satisfaction questionnaire (PQ-LES-Q): reliability and validity. J Am Acad Child Adolesc Psychiatry 45:401–407
- Birmaher B, Khetarpal S, Brent D, Cully M, Balach L, Kaufman J, Neer SM (1997) The screen for child anxiety related emotional disorders (SCARED): scale construction and psychometric characteristics. J Am Acad Child Adolesc Psychiatry 36:545–553
- Stern JA, Gadgil MS, Blakeley-Smith A, Reaven JA, Hepburn SL (2014) Psychometric properties of the SCARED in youth with autism spectrum disorder. Res Autism Spectr Disord 8:1225–1234
- Irwin DE, Stucky B, Langer MM, Thissen D, DeWitt EM, Lai JS et al (2010) An item response analysis of the pediatric PROMIS anxiety and depressive symptoms scales. Qual Life Res 19:595–607
- NIMH (1985) Rating scales and assessment instruments for use in pediatric psychopharmacology research. Psychopharmacol Bull 21:839–843
- Lewin AB, Piacentini J (2010) Evidence-based assessment of child obsessive compulsive disorder: recommendations for clinical practice and treatment research. Child Youth Care Forum 39:73–89
- Pituch KA, Green VA, Didden R, Lang R, O'Reilly MF, Lancioni GE, Sigafoos J (2011). Parent reported treatment priorities for children with autism spectrum disorders. Res Autism Spectr Disord 5:135–143
- 54. Storch EA, Lewin AB, De Nadai AS, Murphy TK (2010) Defining treatment response and remission in obsessive–compulsive disorder: a signal detection analysis of the Children's Yale-Brown Obsessive Compulsive Scale. J Am Acad Child Adolesc Psychiatry 49:708–717
- 55. Storch EA, Arnold EB, Lewin AB, Nadeau JM, Jones AM, De Nadai AS et al (2013) The effect of cognitive-behavioral therapy versus treatment as usual for anxiety in children with autism spectrum disorders: a randomized, controlled trial. J Am Acad Child Adolesc Psychiatry 52:132–142
- 56. Conelea CA, Freeman JB (2015) What do therapists and clients do during exposures for OCD? Introduction to the special issue

on theory-based exposure process. J Obsessive Compuls Relat Disord $6{:}144{-}146$

- Taboas WR, McKay D, Whiteside SP, Storch EA (2015) Parental involvement in youth anxiety treatment: conceptual bases, controversies, and recommendations for intervention. J Anxiety Disord 30:16–18
- Whiteside SP, McKay D, De Nadai AS, Tiede MS, Ale CM, Storch EA (2014) A baseline controlled examination of a 5-day intensive treatment for pediatric obsessive–compulsive disorder. Psychiatry Res 220:441–446
- 59. Storch EA, Lehmkuhl HD, Ricketts E, Geffken GR, Marien W, Murphy TK (2010) An open trial of intensive family based cognitive-behavioral therapy in youth with obsessive-compulsive disorder who are medication partial responders or nonresponders. J Clin Child Adolesc Psychol 39:260–268
- Kerns CM, Kendall PC, Berry L, Souders MC, Franklin ME, Schultz RT et al (2014) Traditional and atypical presentations of anxiety in youth with autism spectrum disorder. J Autism Dev Disord 44:2851–2861