Psicothema
ISSN: 0214-9915
psicothema@cop.es
Colegio Oficial de Psicólogos del Principado de Asturias
España

Iniesta Sepúlveda, Marina; Nadeau, Joshua M; Whelan, Megan K; Oiler, Christina M.; Ramos, Amaya; Riemann, Bradley C.; Storch, Eric A.

Intensive family exposure-based cognitive-behavioral treatment for adolescents with anorexia nervosa

Psicothema, vol. 29, núm. 4, 2017, pp. 433-439
Colegio Oficial de Psicólogos del Principado de Asturias
Oviedo, España

Psicothema

Psicothema 2017, Vol. 29, No. 4, 433-439
doi: 10.7334/psicothema2016.372

ISSN 0214 - 9915 CODEN PSOTEG

www.psicothema.com

Intensive family exposure-based cognitive-behavioral treatment for adolescents with anorexia nervosa

Marina Iniesta Sepúlveda[1], Joshua M Nadeau[2,3], Megan K Whelan[3], Christina M. Oiler[3], Amaya Ramos[3], Bradley C. Riemann[3] and Eric A. Storch[2,3,4]

[1] Catholic University of Murcia, [2] University of South Florida, [3] Rogers Memorial Hospital and [4] All Children's Hospital-Johns Hopkins Medicina

Abstract

Background: Anorexia nervosa exhibits high comorbidity rates and shared features with anxiety disorders and obsessive-compulsive disorder. Anxiety-based etiological models have proposed that fear of eating-related stimuli is the central mechanism around which avoidance of food and food-related rituals are performed. Building on this approach, exposure-based interventions have demonstrated promising results. Limited evidence in adolescents encourages the evaluation of exposure approaches in this population. **Method:** The current study presents a preliminary evaluation, in eight adolescents with anorexia nervosa, of an exposure-based CBT featuring an intensive format and parental involvement. **Results:** significant improvements in physical and psychological outcomes were observed. **Conclusions:** this case series provides preliminary support for the efficacy of intensive family exposure-based CBT for treating adolescents with severe anorexia nervosa.

Keywords: Anorexia nervosa, adolescents, cognitive behavioral therapy, exposure.

Resumen

Tratamiento cognitivo-conductual intensivo familiar con exposición para adolescentes con anorexia nerviosa. **Antecedentes:** la anorexia nerviosa presenta una alta comorbilidad y características compartidas con los trastornos de ansiedad y el trastorno obsesivo-compulsivo. Los modelos etiológicos centrados en la ansiedad han propuesto que el miedo a los estímulos relacionados con la alimentación es el mecanismo principal por el cual se llevan a cabo la evitación de la comida y los rituales relacionados con la alimentación. Basándose en este enfoque las intervenciones centradas en las técnicas de exposición han demostrado resultados prometedores. La escasa evidencia en adolescentes motiva la evaluación de los enfoques basados en la exposición en esta población. **Método:** en el presente estudio se llevó a cabo una evaluación preliminar de la TCC con exposición en formato intensivo e inclusión familiar, en ocho adolescentes con anorexia nerviosa. **Resultados:** se observaron mejoras significativas en las medidas de resultado físicas y psicológicas. **Conclusiones:** el presente estudio de serie de casos proporciona evidencia preliminar de la eficacia de la TCC intensiva familiar con exposición para adolescentes con anorexia nerviosa severa.

Palabras clave: anorexia nerviosa, adolescentes, tratamiento cognitivo-conductual, exposición.

Recent evidence suggests that anorexia nervosa (AN) is a relatively common eating disorder, with lifetime prevalence reaching rates up to 5.4% in European samples (Nagl et al., 2016). Incidence rates are increasing in adolescence, particularly in the 15 to 19 year-old age group (Gandarillas, Zorrilla, Sepúlveda, & Muñoz, 2004) such that, adolescence is now considered a high-risk period for AN onset (Nagl et al., 2016). Adolescents with AN exhibit significant deterioration in overall functioning; for example, Swanson, Crow, Le Grange, Swendsen, and Merikangas (2011) reported that 97.1% of young people with AN in a large community study reported impairment in home, occupational, family, and social domains. Further, Swanson et al. found that 55.2% met criteria for at least one comorbid disorder, and 31.4% experienced suicidal ideation. The observed annual mortality rate for women with AN is dramatically elevated (3.87 deaths per 1,000 person-years), and greater for those (6.6 deaths per 1,000 person-years), who typically experience a longer duration of disorder (Franko et al., 2013). These findings highlight the importance of early detection and treatment of AN. The guidelines of the American Academy of Child and Adolescent Psychiatry Committee on Quality Issues (Lock, La Via, & AACAP, 2015) recommends outpatient psychosocial treatments such as Family –Based Treatment (FBT; Lock & Le Grange, 2013), Adolescent-Focused Therapy (AFT, Fitzpatrick, Moye, Hostee, Le Grange, & Lock, 2010) and Enhanced Cognitive Behavioral Therapy (CBT-E, Fairburn, 2008) as the first choice for adolescents with AN.

High comorbidity between AN, and other psychiatric disorders has been observed, with obsessive-compulsive disorder (OCD) being a common co-occurring condition (3.4%) in female adolescents (Bühren et al., 2014). Both disorders share features in cognitive (perfectionism, obsessional concerns), behavioral

Received: December 1, 2016 • Accepted: June 21, 2017
Corresponding author: Marina Iniesta Sepúlveda
Catholic University of Murcia
Campus de los Jerónimos nº 135
30107 Murcia (Spain)
e-mail: miniesta@ucam.edu

(avoidance, compulsive and ritualistic behaviors) and emotional (disgust, fear) dimensions (Altman & Shankman, 2009). In similar fashion to OCD, etiological models for AN suggest fear conditioning to previously neutral stimuli – including food, eating behaviors, weight gain, etc. – that results in avoidance and compensatory behaviors (Strober, 2004). Intrusive thoughts related with their respective feared stimuli are presented in AN and OCD. It was observed that in both disorders, intrusive thoughts are experienced with similar frequency and cause similar level of emotional distress, although in OCD, intrusions are appraised as more disruptive and difficult to control (García-Soriano, Roncero, Perpiñá, & Belloch, 2014). Avoidance of caloric intake is a core feature in AN, and is generally accompanied by a range of compulsive and/or ritualized eating behaviors – analogous to compulsions in OCD, such as breaking food into small pieces, excessive physical activity, and frequent checking of body image – that are used to decrease anxiety around eating (Steinglass et al., 2012). In addition, the presence of strong associations between meal-related anxiety (especially anxiety prior to meals) and caloric intake have been observed (Steinglass et al., 2010; 2012).

Given phenomenological similarities between AN and OCD (Altman & Shankman, 2009; Davis & Kaptein, 2006), and following an anxiety-based etiological model (Guarda, Schreyer, Boersma, Tamashiro, & Moran, 2015), exposure with or without response prevention has been postulated as a potential intervention strategy in AN (Koskina, Campbell, & Schmidt, 2013). According to Steinglass et al. (2011) 'fear of fat' is central in AN, manifesting as anticipatory anxiety about eating, as well as in concerns about food content, shape, size, and weight gain. These fears result in maladaptive avoidance and eating-related rituals, which act to maintain symptoms through negative reinforcement. During exposures to anxiogenic food-related situations (anticipation of eating, ingesting food, and feeling full) patients recognize that feared consequences (loss of control, immediate obesity, etc.) typically do not occur when compensatory or ritualized behaviors are impeded (Guarda et al., 2015; Steinglass et al., 2011), reducing associations between feared situations and anxiety, rituals and relief, and disconfirming irrational beliefs (Steinglass et al., 2011). In the model proposed by Hildebrandt, Bacow, Markella, and Loeb (2012) anxiety triggers in AN are categorized in five domains: a) food (e.g., texture, smell, caloric content), b) eating (e.g., speed of eating, time of eating), c) interoceptive cues (e.g., feeling full, stomach sensations), d) shape and weight (e.g., changes, symmetry, size) and e) social evaluation (e.g., "dresses up" situations, shopping for clothes). These stimuli trigger anxiety- fear-, worry- and disgust-related experiences, resulting in food avoidance. According to these authors, exposures should be focused on these three types of emotional experiences.

Exposure-based approaches to psychotherapy have been used in adult samples, with the main goal of increasing caloric intake in patients with AN during the weight restoration phase of intervention. In an initial case-series, Steinglass et al. (2012) administered a 12-session ERP (exposure response prevention) intervention, over a 4-week time period, among nine women (ages 17 to 38 years) with AN after achieving minimally normal weight. Exposures were focused on feared eating situations, preventing avoidance and ritualized behaviors. Results showed that 4-day mean caloric intake at post-treatment was 2,568±425 kilocalories (kcal) per day. In the laboratory test meals, mean caloric intake increased after ERP, but this change was not statistically significant. At post-treatment, anxiety before and during the laboratory meal, general anxiety, and food-related distress outcomes decreased, but changes were not statistically significant. In the post-treatment laboratory meal, caloric intake was strongly associated with change in anxiety and food-related distress. The authors concluded that a modest dose of treatment did not result in significant changes in caloric intake. In another study, Steinglass et al. (2014) evaluated the same intervention in a small RCT (randomized controlled trial), where 30 weight-restored patients with AN (ages 16 to 45 years) were randomly assigned to a 12-session regimen of ERP or Cognitive Remediation Therapy (CRT) over a 4-week time period. Patients in the ERP group increased mean caloric intake in test meal from baseline to posttreatment, while mean caloric intake among patients from the CRT group decreased; this difference was statistically significant. State Anxiety subscale in STAI decreased in both groups after intervention; however, there was no statistically significant difference between the two treatment conditions.

Overall, research findings provide promising support for the use of ERP to increase caloric intake through exposure to feared eating-related stimuli (Steinglass et al., 2012; 2014). Samples from the aforementioned studies consisted predominantly of adults in the weight-restoration phase of treatment. Thus, the efficacy of ERP-based interventions in severe adolescent AN cases needs to be assessed.

We hypothesized that an intensive exposure-based CBT including family involvement would be effective in restoring weight, improving disordered eating behaviors, and reducing associated anxiety and depressive symptoms in adolescents with AN. This hypothesis relies upon two principles that have empirical research support in anxiety disorders and OCD: high parental involvement and intensive format. High parental involvement is considered critical when treating children and adolescents, as interventions with limited parental involvement show little efficacy in addressing issues such as comorbidity, compliance, and family or parental support (Storch, 2014). Between-session exposures are enhanced when parents utilize the home setting to multiply opportunities for corrective experiences, and to facilitate learning generalization (Taboas et al., 2015), which enhanced ERP outcomes (Rosa-Alcázar et al., 2015). As it was mentioned, promising findings on the efficacy of exposure intervention in the framework of family-based treatment (FBT) were reported by Hildebrandt, Bacow, Greif, and Flores (2014) in a group of 10 female adolescents (ages 12 - 17) with AN. Preliminary results showed that nine of the ten participants reached >85% of ideal body weight, with significant improvements in global eating symptoms (d=1.64) and eating concerns (1.46) in EDE-Q, depressive (d=1.63) and anxiety (d=2.58) symptoms.

The second principle is that of intensive treatment. Given the severe and resistant nature of AN (Abbate-Daga, Amianto, Delsedime, De-Bacco, & Fassino, 2013), intensive treatment is typically warranted and is recommended for children with severe and resistant symptomatology and acute functional impairments, including daily sessions of massed ERP practice (Storch et al., 2007). Despite the frequent use of hospitalization programs to treat AN, such programs involve separation of youth from family and social environment, and they are recommended when outpatient programs failed, symptomology is severe, and/or are unavailable (Lock, La Via, & American Academy of Child and Adolescent Psychiatry [AACAP] Committee on Quality Issues, 2015). An intensive approach avoids the complete withdrawal of youth, and

offers the potential capability to produce faster improvements could permit youth to rapidly resume normal activities (Whiteside et al., 2014). In light of these principles, we present a preliminary evaluation of a family-based intensive CBT utilizing ERP as its primary treatment modality in eight adolescents with AN.

Method

Participants

Participants included 8 female adolescent Caucasian patients aged between 11 and 15 years (*M*=14±1.41 years) seeking intensive outpatient treatment to address symptoms of AN. All participants had a primary diagnosis of AN according to the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; American Psychiatric Association [APA], 2013) criteria in which amenorrhea is not required. Six (75%) of the participants presented with restrictive subtype and two (25%) with purgative subtype. Diagnoses were derived through consensus procedure (Leckman, Sholomskas, Thompson, Belanger, & Weissman, 1982), which combined all available clinical information made during clinical interview with a board certified child/adolescent psychiatrist, interactions with two licensed psychologists, and review of clinical records (e.g., results on standardized measures such as Eating Disorder Examination Questionnaire –EDE-Q–, Fairburn & Beglin, 200; Yale-Brown-Cornell Eating Disorders Scale – YBC-EDS–, Mazure, Halmi, Sunday, Romano, & Einhorn, 1994; etc.). Each of the eight participants was reviewed by a caseness panel consisting of these three clinicians; there was 100% agreement related to diagnoses of AN as the primary psychiatric disorder according DSM-5 criteria. Beyond diagnosis, other inclusion criteria included were: (1) clinically significant AN symptom as evidenced by a score on EDE-Q above 75% percentile according normative scores by Carter, Stewart, and Fairburn (2001); and (2) had at least one parent available to participate actively in the intervention process. Exclusion criteria were to present with lifetime history of psychosis, current suicidal ideation, or substance abuse.

Time from AN symptoms onset ranged between 2 and 60 months (*M*=21.2 months). All participants presented with at least one comorbid disorder. Social anxiety and depression were diagnosed in five (62.5%) participants, OCD in two (25.0%) and generalized anxiety disorder in one (12.5%). Seven (87.5%) participants received previous impatient or outpatient psychosocial interventions. Five (62.5%) participants were taking psychotropic medications, three (37.5%) SSRI (one escitalopram and two fluoxetine), one (12.5%) tricyclic antidepressant (clomipramine), and one (12.5%) antipsychotics (quetiapine).

Mothers primarily attended treatment sessions in six cases (75%) and both parents in the two cases (25%). Parents were married in four cases (50%) and were divorced in the other four instances (50%). Four (50%) participants had two siblings, two (25%) had one sibling, one (12.5%) had one sibling, and one (12.5%) had four siblings.

Instruments

Primary Outcome Measures

Eating Disorder Examination Questionnaire (EDE-Q; Fairburn & Beglin, 2008). The EDE-Q is a 28-item self-report measure assessing eating disorder (ED) psychopathology. Frequency (from 0 = *no days* to 6 = *every day*) and severity (from 0 = *not at all* to 6 = *markedly*) of symptoms, and beliefs are rated over the past 28 days. The questionnaire includes four subscales, Restraint, Eating Concern, Shape Concern, and Weight Concern; and 6 items rating frequency of binge eating, self-induced vomiting, laxative use or excessive exercise, among others. Adequate psychometric properties have been observed across studies (Berg, Peterson, Frazier, & Crow, 2012).

Yale-Brown-Cornell Eating Disorders Scale (YBC-EDS; Mazure et al., 1994). The YBC-EDS is a semi-structured clinician-administered interview to assess eating-related rituals and/or preoccupations frequently experienced by patients with eating disorders. Concerns and rituals related to food, eating, weight, and shape appearance are endorsed in the Symptoms Checklist. Symptoms are then rated on a 16-item Severity Scale assessing time occupied, interference, distress, resistance, control, association with self, reasonableness, and desire for change. Good reliability, validity (Mazure et al., 1994) and sensitivity to change after treatment (Jordan et al., 2009) have been observed.

Clinical Global Impression–Severity (*CGI-S*; NIMH, 1985). The CGI-S is one of the most used measures in treatment studies of mental disorders in children and adolescents. It is a single-item clinician-rated scale assessing anxiety disorder severity based on associated impairment from 0 (*no illness*) to 6 (*extremely severe*).

Secondary Outcome Measures

Body Checking Questionnaire (BCQ; Reas, Whisenhunt, Netemeyer and Williamson, 2002). The BCQ is a 23-item self-report assessing body checking. Items describe control behaviors linked to three domains (Overall Appearance, Specific Body Parts, and Idiosyncratic Checking) and are rated from 1 (*never*) to 5 (*very often*). Good psychometric properties (internal consistency, test-retest reliability and concurrent and discriminating validity) were found for total score and subscales (Reas et al., 2002).

Quick Inventory of Depressive Symptomatology 16-item Self-Report (QIDS-16-SR, Rush et al., 2003). The QIDS-16-SR is a brief self-report assessing several domains of depressive symptomatology, including sad mood, difficulty with concentration, self-criticism, suicidal ideation, decrease in general interest, energy/fatigue and sleep disturbance, among others. Sixteen items are rated in a 4-point scale where 0 usually describes a typical situation with no disorder (e.g., *I do not wake up at night*) and 3 a typical situation when depressive disorder is present (e.g., *I awaken more than once a night and stay awake for 20 minutes or more*). Increasing scores associated with depressive symptom severity in the previous seven days. The QIDS-16-SR has shown acceptable internal consistency, and convergent and discriminant validity (Reilly, MacGillivray, Reid, & Cameron, 2015).

Pediatric Quality of Life Enjoyment and Satisfaction Questionnaire (*PQ-LES-Q*; Endicott, Nee, Yang, & Wohlberg, 2006). The PQ-LES-Q is a 15-item self-report measuring quality of life in children and adolescents, aged 6 to 17 years, across several domains. Items are rated on a 6-point scale, ranging from 1 (*very poor*) to 5 (*very good*). Total score is the sum of all items, with higher scores indicating greater quality of life. Good psychometric properties such as excellent internal consistency, reliability, and convergent validity have been reported for the PQ-LES-Q (Endicott et al., 2006).

State-Trait Anxiety Inventory (STAI; Spielberger, 1983). The STAI is one of the most used and investigated measures of anxiety. It consists of two 20-item scales measuring state and trait anxiety, respectively, with all items rated on a 4-point Likert scale from 1 (*not at all*) to 4 (*very much*). Adequate internal consistency and test-retest reliability have been reported for the STAI (Barnes, Harp, & Jung 2002).

Procedure

The institutional review boards at the University of South Florida and Rogers Memorial Hospital approved this study, which involved archival records review. Participants were families who had completed a regimen (treatment days range = 15-69) of intensive exposure-based CBT at a multidisciplinary behavioral health center specializing in the treatment of youth with AN. Therefore, convenience non-random sampling was used, including all accessible participants that fulfilled inclusion criteria. Pre-treatment measures were administered within 3 days of admission to the treatment program, and post-treatment measures on the final day of treatment. Clinician-rated measures were administered by unblinded trained graduate-level evaluators familiar with the patient and family being treated.

Treatment. All participants completed a regimen of intensive partial hospitalization program of 6.5 hours per day, 5 days per week CBT incorporating exposure-based behavioral tasks. 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 15 to 69 days (*Mean* = 40±17.2) across participants.

Intensive treatment was based on the CBT-E by Fairburn (2008) that included four stages. Stage one is focused on psychoeducation, self-monitoring, and regularizing eating patterns. In second stage, progress, potential barriers and disorder formulation are reviewed. In the third stage, mechanisms that are maintaining the disorder (e.g., over-evaluation and checking of shape, "feeling fat" perception, etc.) are addressed. Finally, the fourth stage is focused on supporting changes and preventing future relapses.

Relevant modifications to treatment were included. First, a focus on cognitive components was displaced in favor of ERP practice. Second, modifications to allow for delivery of treatment in an intensive outpatient setting included shortened check-in/-out procedures, increased use of group-based activities (e.g., nutrition education, nursing education), and altered activity and homework schedules to reflect daily treatment sessions.

ERP was the primary component of the treatment. Across ERP trials, feared situations (e.g., smelling food when cooking, eating caloric food, seeing a specific body part in the mirror, etc.) were gradually confronted and escape responses (e.g., repeated body checking, cutting food in little pieces, doing exercise, etc.) resisted. Exposure tasks were initiated at during the third day of admission, and practiced daily in sessions and as homework. In each trial, exposure was prolonged until reaching at least an anxiety reduction of 50%. In all cases, treatment also included psychoeducation about the disorder and treatment rationale, hierarchy development of exposure situations, establishing socially appropriate coping strategies (e.g., problem-solving, respiratory control, assertiveness), weight restoration (as appropriate), psychiatric medication management, and relapse prevention. Care was given to ensure that treatment materials and language were developmentally appropriate for adolescents.

At least one parent attended all treatment sessions with the adolescent. In sessions, parents received psychoeducation, and learned how to guide ERP, monitor anxiety, and support the youth. From this approach, every family meal is an exposure task for adolescents, parents guided and supported youth when coping with anxiety during meal-related situations at home. Also, as necessary parents were instructed to facilitate treatment engagement using contingency management techniques.

Treatment was delivered by Masters-level therapists with at least 1 year of experience working with youth. Regular clinical supervision with licensed clinical psychologists was provided.

Data analysis

Data on primary outcome measures are presented separately for each child. Average indices were calculated for both primary and secondary outcomes. Nonparametric Wilcoxon signed rank tests were used to test treatment effects. To be considered in remission, participants had to reach a BMI score in the "Healthy" range according to growth charts promulgated by the National Center for Chronic Disease Prevention and Health Promotion (Ogden et al., 2002). Following Couturier and Lock (2006) a second remission criteria involves combination of Ideal Body Weight (IBW) ≥ 90% and EDE-Q Restrain subscale scores within 1 SD of normative means. IBW was computed following the BMI method described in Phillips, Edlbeck, Kirby, and Goday (2007). We used EDE-Q norms developed by Carter et al. (2001) for young adolescent girls.

Results

Characteristics of participants at pre-treatment

Participant demographics, diagnoses, and scores on primary outcome variables are presented in Table 1. At pre-treatment, according to percentile ranks for adolescent girls (Carter et al., 2001) for EDE-Q total scores, three (37.5%) participants presented with symptoms at the 80th percentile, two (25.0%) at the 90th percentile, two (25.0%) at the 95th percentile, and one (12.5%) at the 99th percentile. Associated impairment as measured by the CGI-Severity included four (50%) participants with extremely severe symptoms, and four (50%) with severe symptoms. Table 3 shows pre-treatment mean scores for primary and secondary outcome measures.

Treatment Effects on Primary Outcome Measures

Table 3 presents means and standard deviations for outcome measures, as well as Wilcoxon signed rank test. Physical outcomes showed significant mean increases from pre- to post-treatment in both weight (*M* = 5.51±3.44) and BMI (*M* = .91±.55). In addition, all symptom severity outcomes reflected significant improvements: EDE-Q total score (*M* = –1.33±1.02), EDE-Q Restraint (*M* = –1.97±1.58), EDE-Q Eating Concerns (*M* = –2.07±1.88), EDE-Q Shape Concerns (*M* = –2.44±1.56), EDE-Q Weight Concerns (*M* = –2.27±1.13), YBC-EDS total score (M = –13.14±11.74), YBC-EDS Concerns (M = –6.14±6.47), YBC-EDS Rituals (M = –7.00±5.77), and CGI-Severity (*M* = –.62±.52). The CGI-Severity score also reflected significant change (*M* = –.63±.51) from pretreatment to posttreatment.

Table 2 shows weight and BMI of participants at pre- and post-treatment. Regarding clinical significance, at post-treatment

Table 1
Participant demographics, diagnosis, and scores on primary outcome variables

Participant	Gender	Age	Diagnosis	EDE-Q										YBC-EDS						CGI-Severity	
				Total		Restraint		Eating		Shape		Weight		Total		Concerns		Rituals			
				Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
1	Female	15	AN purgative	4.4	0.1	4.0	0.0	4.2	0.0	5.0	0.3	4.4	0.0	33	0	19	0	14	0	4	3
2	Female	15	AN	5.4	3.3	4.8	4.2	6.0	1.2	5.8	4.3	5.2	3.6	42	36	21	20	21	16	4	4
3	Female	15	AN	3.1	1.3	4.4	0.6	1.8	1.8	4.1	2.8	2.0	0.0	24	10	12	6	12	4	5	4
4	Female	13	AN	2.8	0.6	2.0	0.0	1.2	1.2	4.6	0.9	3.2	0.2	24	13	13	9	11	4	5	4
5	Female	14	AN purgative	3.7	2.6	3.8	0.8	2.0	1.0	4.1	4.1	5.0	4.4	20	25	11	12	9	13	5	4
6	Female	11	AN	2.7	1.4	3.0	1.6	1.2	0.4	3.5	1.9	3.2	1.6	–	24	–	12	–	12	4	4
7	Female	14	AN	4.1	1.3	3.2	0.8	3.2	0.2	5.4	2.1	4.6	2.2	29	16	14	8	15	8	5	4
8	Female	15	AN	4.4	1.5	3.6	0.4	4.6	1.8	5.8	2.5	3.8	1.2	40	20	19	11	21	9	4	4

Note. AN: anorexia nervosa, EDNOS: eating disorder no specified, OCD: obsessive-compulsive disorder. EDE-Q: Eating Disorder Examination Questionnaire, YBC-EDS: Yale-Brown-Cornell Eating Disorders Scale, CGI-S: Clinical Global Impression–Severity

Table 2
Participant weight and BMI at pre- and post-treatment

Participant	Weight (lbs)		BMI	
	Pre-treatment	Post-treatment	Pre-treatment	Post-treatment
1	138.8	141.4	23.1	23.5
2	101.4	107.4	17.9	19.0
3	117.0	124.0	19.0	20.0
4	101.2	112.0	15.8	17.5
5	92.0	98.8	15.3	16.4
6	103.0	108.0	18.2	19.1
7	110.0	110.0	20.1	20.1
8	95.6	101.0	19.0	20.1

eight (100%) participants exhibited BMI scores in the "Normal" range (Ogden et al., 2002). With respect to the combination of IBW ≥90% and EDE Restrain subscale score criteria, six (75%) participants were classified as in remission.

Treatment Effects on Secondary Outcome Measures

Table 3 shows results from secondary measures. Body checking on BCQ decreased significantly for total score (M = –16.16±14.82) as well as for Appearance (M = –4.33±8.33), Body (M = –7.33±6.40), and Idiosyncratic Checking (M = –4.16±2.56) dimensions. For quality of life, the PQ-LES-Q score showed significant increase (M = 11.83±10.74). Depressive symptoms also exhibited significant improvement (M = –6.87±3.60). Finally, a significant decrease was observed for trait anxiety (M = –10.00±10.22) as measured on the STAI; however, only a non-significant decrease was seen for state anxiety (M = –8.40±14.06).

Discussion

Findings from this initial case series provide preliminary support for the efficacy of an intensive family-based exposure therapy program to improve the core pathological features in AN (i.e., weight/BMI, related-eating concerns, rituals, body checking), as well as quality of life, depressive symptoms and general anxiety. Findings of some previous studies where exposure therapy was associated with increased weight are confirmed (Hildebrandt et al, 2014). Improvements in weight and BMI imply an increased caloric intake following treatment,

Table 3
Means, standard deviations and results for Wilcoxon signed rank test on outcome measures

Outcome	n	Pre-treatment Mean (SD)	Post-treatment Mean (SD)	Z	p
Weight	8	107.37(14.90)	112.82(13.83)	2.366	.018
BMI	8	18.55(2.45)	19.46(2.10)	2.384	.017
EDE-Q Total	8	3.83(.95)	1.50(1.03)	–2.521	.012
EDE-Q Restraint		3.60 (.88)	1.05 (1.37)	–2.521	.012
EDE-Q Eating		3.03(1.77)	.95(.69)	–2.201	.028
EDE-Q Shape		4.78(.83)	2.35(1.40)	–2.371	.018
EDE-Q Weight		3.93(1.08)	1.65(1.67)	–2.521	.012
YBC-EDS Total	7	39.29(8.42)	17.14(11.47)	–2.197	.028
YBC-EDS Concerns		15.57(3.99)	9.43(6.11)	–2.120	.034
YBC-EDS Rituals		14.71(4.72)	7.71(5.56)	–2.197	.028
CGI-Severity	8	4.50(.53)	3.87(.35)	–2.236	.025
BCQ Total	6	59.67(20.96)	43.50(15.15)	–1.782	.075
BCQ Appearance		25.67(10.69)	21.33(7.69)	–1.367	.171
BCQ Body		23.33(9.16)	16.00(6.48)	–1.997	.046
BCQ Checking		11.67(3.08)	7.50(2.43)	–2.207	.027
PQ-LES-Q	6	46.00(7.01)	57.83(11.78)	1.997	.046
QIDS-16-SR	8	11.13(3.09)	4.25(2.71)	–2.524	.012
STAI State	5	36.80(13.29)	28.40(10.43)	–1.214	.225
STAI Trait	5	48.40(5.50)	38.40(11.71)	–2.023	.043

Note: EDE-Q: Eating Disorder Examination Questionnaire. YBC-EDS: Yale-Brown-Cornell Eating Disorders Scale. CGI-S: Clinical Global Impression–Severity. BCQ: Body Checking Questionnaire. PQ-LES-Q: Pediatric Quality of Life Enjoyment and Satisfaction Questionnaire. QIDS-16-SR: Quick Inventory of Depressive Symptomatology 16-item Self-Report. STAI: State–Trait Anxiety Inventory

as seen in previous studies (Steinglass et al., 2014; 2012). In terms of treatment response, we observed that seven participants were responders according to BMI criteria, and six according to IBW+EDE-score criteria. Our results are similar to Hildebrandt et al. (2014), who reported a 90% response rate in nine adolescents with AN after receiving an exposure-based program. As in the current study, the parental involvement was central in their intervention protocol, which may have supported generalization of skills in the natural environment. Although preliminary in nature, these findings may also reflect the intensive nature of the present exposure therapy protocol, which allowed for multiple feared triggers to be targeted including those related to AN as well as comorbid psychopathology.

Present findings suggest that massed practice of exposure enhances improvement in psychological variables, as suggested by AN symptom severity scores (EDE-Q and YBC-EDS) and in secondary outcomes (body checking, quality of life, depressive symptoms and trait anxiety). This agrees with Hildebrandt et al. (2014), who reported reductions in global severity, eating concerns, depressive and anxiety symptoms after a 20-session program. Differing from our findings, Hildebrandt et al. (2014) reported no significant changes in restraint, shape and weight concerns. It may be that an intensive treatment has a larger effect on these symptoms than weekly approaches, due to massed practice in combination with increased opportunities for corrective experiences demonstrating that feared consequences (e.g., instantaneous weight gain or shape changes) do not occur. Some studies failed to improve anxiety significantly with four sessions of exposure (Steinglass et al., 2012). In this sense we also observed no significant change in state anxiety. It is possible that measures that reflect overall anxiety levels are not sensitive to change to eating-related anxiety which is the targeted outcome of exposure interventions in AN, and tools assessing this specific phenomenon should be included in future studies (e.g., Fear of Food Measure – FOFM –; Levinson & Byrne, 2015).

It is considered that family involvement enhanced exposure therapy in the current study, providing additional support for the importance of family participation in treatment of youth with AN (Hildebrandt et al., 2014; Lock et al., 2015). In addition to assisting with exposures and reducing accommodating behaviors, family members need to be taught how to generalize skills outside the treatment setting, change maladaptive patterns of family functioning especially around food, eating behavior and appearance.

There are several limitations to the current study that should be noted. First, the small sample size limits generalizability of results. Second, as treatment duration was determined by individual presentation, length of stay and pharmacological regimen was different across participants. Examining whether length of intervention has an effect on treatment effectiveness would be interesting for future studies. Third, a laboratory test meal was not included as an outcome measure, such that we were unable to calculate changes in caloric intake. Forth, exposure techniques were included within a multicomponent treatment package. Future dismantling studies would be necessary to examine the specific contribution of ERP to improve symptoms in AN. Within these limitations, the current case series provides preliminary support for the efficacy of exposure-based CBT to treat adolescents with severe AN in, utilizing a family-based intensive approach. These results are of particular importance given the need for increased evidence-based treatment options for AN.

Acknowledgements

Funding for preparation this manuscript was provided to the first author by Ministry of Education, Culture and Sports of Spanish Government in the "José Castillejo" Program, Grant CAS15/00381. Ministry of Education, Culture and Sports had no role in the study, or the decision to submit the paper for publication. Preparation of this article was also supported in part by a Fulbright Scholar Award to the last author.

References

Abbate-Daga, G., Amianto, F., Delsedime, N., De-Bacco, C., & Fassino, S. (2013). Resistance to treatment and change in anorexia nervosa: A clinical overview. *BMC Psychiatry*, *13*, 294. doi:10.1186/1471-244X-13-294

Altman, S. E., & Shankman, S. A. (2009). What is the association between obsessive-compulsive disorder and eating disorders? *Clinical Psychology Review*, *29*, 638-646. doi:10.1016/j.cpr.2009.08.001

American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.). Washington, DC: American Psychiatric Publishing.

Barnes, L. L., Harp, D., & Jung, W. S. (2002). Reliability generalization of scores on the Spielberger state-trait anxiety inventory. *Educational and Psychological Measurement*, *62*, 603-618. doi: 10.1177/0013164402062004005

Berg, K. C., Peterson, C. B., Frazier, P., & Crow, S. J. (2012). Psychometric evaluation of the eating disorder examination and eating disorder examination-questionnaire: A systematic review of the literature. *International Journal of Eating Disorders*, *45*, 428-438. doi: 10.1002/eat.20931

Bühren, K., Schwarte, R., Fluck, F., Timmesfeld, N., Krei, M., Egberts, K., ..., & Herpertz-Dahlmann, B. (2014). Comorbid psychiatric disorders in female adolescents with first-onset anorexia nervosa. *European Eating Disorders Review*, *22*(1), 39-44. doi:10.1002/erv.2254

Carter, J. C., Stewart, D. A., & Fairburn, C. G. (2001). Eating disorder examination questionnaire: Norms for young adolescent girls. *Behaviour Research and Therapy*, *39*, 625-632. doi:10.1016/S0005-7967(00)00033-4

Couturier, J., & Lock, J. (2006). What is recovery in adolescent anorexia nervosa? *International Journal of Eating Disorders, 39*, 550-555. doi:10.1002/eat.20224

Davis, C., & Kaptein, S. (2006). Anorexia nervosa with excessive exercise: A phenotype with close links to obsessive-compulsive disorder. *Psychiatry Research*, *142*(2), 209-217. doi:10.1016/j.psychres.2005.11.006

Endicott, J., Nee, J., Yang, R., & Wohlberg, C. (2006). Pediatric quality of life enjoyment and satisfaction questionnaire (PQ-LES-Q): Reliability and validity. *Journal of the American Academy of Child & Adolescent Psychiatry, 45*, 401-407. doi:10.1097/01.chi.0000198590.38325.81

Fairburn, C. G. (2008). *Cognitive behavior therapy and eating disorders*. New York: Guildford Press.

Fairburn, C. G., & Beglin, S. J. (2008). Eating Disorder Examination Questionnaire (EDE-Q 6.0). In C. G. Fairburn (Ed.), *Cognitive Behavior Therapy and Eating Disorders*. New York: Guilford Press.

Fitzpatrick, K., Moye, A., Hostee, R., Le Grange, D., & Lock, J. (2010). Adolescent focused therapy for adolescent anorexia nervosa. *Journal of Contemporary Psychotherapy, 40*, 31-39. doi: 10.1007/s10879-009-9123-7

Franko, D. L., Keshaviah, A., Eddy, K. T., Krishna, M., Davis, M. C., Keel, P. K., & Herzog, D. B. (2013). A longitudinal investigation of mortality in anorexia nervosa and bulimia nervosa. *The American Journal of Psychiatry*, *170,* 917-925. doi:10.1176/appi.ajp.2013.12070868

Gandarillas, A., Zorrilla, B., Sepúlveda, A. R., & Muñoz, P. E. (2004). Trastornos del comportamiento alimentario: prevalencia de casos en mujeres adolescentes de la Comunidad de Madrid [Eating disorders: Prevalence of cases in adolescent women from Madrid]. *Psiquiatria.com*, *8*(2), 1-4.

García-Soriano, G., Roncero, M., Perpiñá, C., & Belloch, A. (2014). Intrusive thoughts in obsessive-compulsive disorder and eating disorder patients: A differential analysis. *European Eating Disorders Review*, *22*(3), 191-199. doi:10.1002/erv.2285

Guarda, A. S., Schreyer, C. C., Boersma, G. J., Tamashiro, K. L., & Moran, T. H. (2015). Anorexia nervosa as a motivated behavior: Relevance of anxiety, stress, fear and learning. *Physiology & Behavior*, *152*, 466-472. doi:10.1016/j.physbeh.2015.04.007

Hildebrandt, T., Bacow, T., Greif, R., & Flores, A. (2014). Exposure-Based Family Therapy (FBT-E): An open case series of a new treatment for anorexia nervosa. *Cognitive and Behavioral Practice*, *21*, 470-484. doi:10.1016/j.cbpra.2013.10.006

Hildebrandt, T., Bacow, T., Markella, M., & Loeb, K. L. (2012). Anxiety in anorexia nervosa and its management using family-based treatment. *European Eating Disorders Review, 20*, e1-e16. doi:10.1002/erv.1071

Jordan, J., Joyce, P. R., Carter, F. A., McIntosh, V. V., Luty, S. E., McKenzie, J. M., ..., & Bulik, C. M. (2009). The Yale–Brown-Cornell eating disorder scale in women with anorexia nervosa: What is it measuring? *International Journal of Eating Disorders*, *42*(3), 267-274. doi:10.1002/eat.20605

Koskina, A., Campbell, I. C., & Schmidt, U. (2013). Exposure therapy in eating disorders revisited. *Neuroscience & Biobehavioral Reviews*, *37*(2), 193-208. doi:10.1016/j.neubiorev.2012.11.010

Leckman, J. F., Sholomskas, D., Thompson, D., Belanger, A., & Weissman, M. M. (1982). Best estimate of lifetime psychiatric diagnosis: A methodological study. *Archives of General Psychiatry*, *39*, 879. doi:10.1001/archpsyc.1982.04290080001001

Levinson, C. A., & Byrne, M. (2015). The Fear of Food Measure: A novel measure for use in exposure therapy for eating disorders. *International Journal of Eating Disorders*, *48*(3), 271-283. doi:10.1002/eat.22344

Lewin, A. B., Wu, M. S., McGuire, J. F., & Storch, E. A. (2014). Cognitive behavior therapy for obsessive-compulsive and related disorders. *Psychiatric Clinics of North America*, *37*, 415-445. doi:10.1016/j.psc.2014.05.002

Lock, J., & Le Grange, D. (2013). *Treatment manual for anorexia nervosa: A family-based approach*. New York: Guildford Press.

Lock, J., La Via, M. C., & American Academy of Child and Adolescent Psychiatry –AACAP– Committee on Quality Issues –CQI– (2015). Practice parameter for the assessment and treatment of children and adolescents with eating disorders. *Journal of the American Academy of Child & Adolescent Psychiatry*, *54*(5), 412-425. doi:10.1016/j.jaac.2015.01.018

Mazure, C. M., Halmi, K. A., Sunday, S. R., Romano, S. J., & Einhorn, A. M. (1994). The Yale-Brown-Cornell Eating Disorder Scale: Development, use, reliability and validity. *Journal of Psychiatric Research*, *28*(5), 425-445. doi:10.1016/0022-3956(94)90002-7

Nagl, M., Jacobi, C., Paul, M., Beesdo-Baum, K., Höfler, M., Lieb, R., & Wittchen, H. U. (2016). Prevalence, incidence, and natural course of anorexia and bulimia nervosa among adolescents and young adults. *European Child & Adolescent Psychiatry*, *25*(8), 903-918. doi:10.1007/s00787-015-0808-z

National Institutes of Mental Health (1985). Rating scales and assessment instruments for use in pediatric psychopharmacology research. *Psychopharmacological Bulletin, 21,* 839-843.

Ogden, C. L., Kuczmarski, R. J., Flegal, K. M., Mei, Z., Guo, S., Wei, R., ..., & Johnson, C. L. (2002). Centers for Disease Control and Prevention 2000 growth charts for the United States: Improvements to the 1977 National Center for Health Statistics version. *Pediatrics, 109*, 45-60. doi: 10.1542/peds.109.1.45

Phillips, S., Edlbeck, A., Kirby, M., & Goday, P. (2007). Ideal body weight in children. *Nutrition in Clinical Practice*, *22*(2), 240-245. doi:10.1177/0115426507022002240

Reas, D. L., Whisenhunt, B. L., Netemeyer, R., & Williamson, D. A. (2002). Development of the Body Checking Questionnaire: A self-report measure of body checking behaviors. *International Journal of Eating Disorders*, *31*, 324-333. doi:10.1002/eat.10012

Reilly, T. J., MacGillivray, S. A., Reid, I. C., & Cameron, I. M. (2015). Psychometric properties of the 16-item Quick Inventory of Depressive Symptomatology: A systematic review and meta-analysis. *Journal of Psychiatric Research, 60*, 132-140. doi:10.1016/j.jpsychires.2014.09.008

Rosa-Alcázar, A. I., Sánchez-Meca, J., Rosa-Alcázar, Á., Iniesta-Sepúlveda, M., Olivares-Rodríguez, J., & Parada-Navas, J. L. (2015). Psychological treatment of obsessive-compulsive disorder in children and adolescents: A meta-analysis. *The Spanish Journal of Psychology, 18*, e20. doi:10.1017/sjp.2015.22

Rush, A. J., Trivedi, M. H., Ibrahim, H. M., Carmody, T. J., Arnow, B., Klein, D. N., ..., & Thase, M. E. (2003). The 16-Item Quick Inventory of Depressive Symptomatology (QIDS), clinician rating (QIDS-C), and self-report (QIDS-SR): A psychometric evaluation in patients with chronic major depression. *Biological Psychiatry*, *54*, 573-583. doi:10.1016/S0006-3223(02)01866-8

Spielberger, C. D. (1983). *Manual for the State-Trait Anxiety Inventory*. Palo Alto, CA: Consulting Psychologists Press

Steinglass, J. E., Albano, A. M., Simpson, H. B., Wang, Y., Zou, J., Attia, E., & Walsh, B. T. (2014). Confronting fear using exposure and response prevention for anorexia nervosa: A randomized controlled pilot study. *International Journal of Eating Disorders, 47*, 174-180. doi:10.1002/eat.22214

Steinglass, J. E., Sysko, R., Glasofer, D., Albano, A. M., Simpson, H. B., & Walsh, B. T. (2011). Rationale for the application of exposure and response prevention to the treatment of anorexia nervosa. *International Journal of Eating Disorders*, *44*(2), 134-141. doi:10.1002/eat.20784

Steinglass, J. E., Sysko, R., Mayer, L., Berner, L. A., Schebendach, J., Wang, Y., ..., & Walsh, B. T. (2010). Pre-meal anxiety and food intake in anorexia nervosa. *Appetite*, *55*, 214-218. doi:10.1016/j.appet.2010.05.090

Steinglass, J., Albano, A. M., Simpson, H. B., Carpenter, K., Schebendach, J., & Attia, E. (2012). Fear of food as a treatment target: Exposure and response prevention for anorexia nervosa in an open series. *International Journal of Eating Disorders*, *45*, 615-621. doi:10.1002/eat.20936

Storch, E. A. (2014). Can we improve psychosocial treatments for child anxiety? *Depression and Anxiety*, *31*, 539-541. doi:10.1002/da.22283

Storch, E. A., Bagner, D. M., Geffken, G. R., Adkins, J. W., Murphy, T. K., & Goodman, W. K. (2007). Sequential cognitive-behavioral therapy for children with obsessive-compulsive disorder with an inadequate medication response: A case series of five patients. *Depression and Anxiety*, *24*, 375-381. doi:10.1002/da.20260

Strober, M. (2004). Pathologic fear conditioning and anorexia nervosa: On the search for novel paradigms. *International Journal of Eating Disorders*, *35*, 504-508. doi:10.1002/eat.20029

Swanson, S. A., Crow, S. J., Le Grange, D., Swendsen, J., & Merikangas, K. R. (2011). Prevalence and correlates of eating disorders in adolescents: Results from the national comorbidity survey replication adolescent supplement. *Archives of General Psychiatry*, *68*, 714-723. doi:10.1001/archgenpsychiatry.2011.22

Taboas, W. R., McKay, D., Whiteside, S. P., & Storch, E. A. (2015). Parental involvement in youth anxiety treatment: Conceptual bases, controversies, and recommendations for intervention. *Journal of Anxiety Disorders*, *30*, 16-18. doi:10.1016/j.janxdis.2014.12.005

Whiteside, S. P., McKay, D., De Nadai, A. S., Tiede, M. S., Ale, C. M., & Storch, E. A. (2014). A baseline controlled examination of a 5-day intensive treatment for pediatric obsessive-compulsive disorder. *Psychiatry Research*, *220*(1), 441-446. doi:10.1016/j.psychres.2014.07.006