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Internet-delivered cognitive behavior therapy for adolescents with functional gastrointestinal disorders — An open trial

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ABSTRACT

Functional gastrointestinal disorders (FGID), including irritable bowel syndrome, functional dyspepsia and functional abdominal pain, are common in adolescents and are associated with substantially decreased quality of life. Cognitive behavior therapy for children and adolescents with FGID is one of few treatments that have shown effect, but treatment access is limited. In adults with irritable bowel syndrome, exposure-based internet-delivered CBT (ICBT) leads to reduced symptoms and increased quality of life, but studies in children are lacking. This open pilot aimed to evaluate feasibility and the potential efficacy of an exposure-based ICBT-program for adolescents with pain-predominant FGID. Twenty-nine adolescents (age 13–17), with FGID were included. The ICBT-program lasted for 8 weeks with weekly online therapist support. The protocol for adolescents included exposure to abdominal symptoms, while the protocol for parents aimed at increasing parents' attention to adolescent healthy behaviors. Assessment points were baseline, post-treatment and 6-month follow-up. The primary outcome was the Gastrointestinal Symptoms Rating Scale-IBS (GSRS-IBS). Effect sizes were calculated using Cohen's *d* in an intent to treat analysis. GSRS-IBS improved significantly from baseline to post-treatment (mean difference 6.48; 95% CI [2.37–10.58]) and to follow-up (mean difference 7.82; 95% CI [3.43–12.21]), corresponding to moderate effect sizes (within-group Cohen's $d = 0.50$; 95% CI [0.16–0.84] and $d = 0.63$; 95% CI [0.24–1.02], respectively). Treatment adherence was high with 22 of 29 (76%) adolescents completing the entire treatment period. High adherence indicates acceptability of format and content, while symptomatic improvement suggests potential efficacy for this ICBT intervention in adolescents with FGID.

1. Introduction

Functional gastrointestinal disorders (FGID) are common in adolescents, with a prevalence of 14% reported in Japan (Sagawa et al., 2013) and as much as 24% reported in an often cited school-based study (Saps et al., 2009). FGID are associated with school absenteeism, high health care consumption, increased anxiety and depression (Saps et al., 2009) and have a large negative impact on the quality of life (Youssef et al., 2006). Three of the most common types of FGID are irritable bowel syndrome (IBS), functional abdominal pain (FAP), and functional dyspepsia (FD) (Sagawa et al., 2013). They are characterized by recurrent abdominal pain or discomfort and in IBS, also a change in defecation patterns and consistency (Rasquin et al., 2006). Other gastrointestinal symptoms like nausea, bloating, and flatulence are also common in FGID (Rasquin et al., 2006). FGID symptoms have been shown to be stable over time (Walker et al., 1998) and sustain into adulthood for many patients (Campo et al., 2001). There is currently no support that medical or dietary treatments have any meaningful beneficial effects (Huertas-Ceballos et al., 2008, 2009) but cognitive behavior therapy (CBT) has been evaluated for children and adolescents with FGID with promising results (Foisy et al., 2011; Sprenger et al., 2011).

Abbreviations: IBS, irritable bowel syndrome; FAP, functional abdominal pain; FD, functional dyspepsia; FGID, functional gastrointestinal disorders; CBT, cognitive behavior therapy; ICBT, internet-delivered cognitive behavior therapy; GSRS-IBS, Gastrointestinal Symptom Rating Scale-IBS version; PRS, Pain Reactivity Scale; PII, Pain Interference Index; FDI, Functional Disability Inventory; CASI, Childhood Anxiety Sensitivity Index; CDI, Child Depression Inventory; PSS-4, Perceived Stress Scale-4.

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Although CBT can be an effective treatment for FGID in children, there is a shortage of CBT therapists (Shafran et al., 2009). During the last two decades, internet-delivered CBT (ICBT) has been applied to more than 20 different clinical disorders in adults in more than 100 studies with results equivalent to traditional CBT (Hedman et al., 2012). While ICBT treatments are often based on the same therapeutic content as face-to-face CBT, the content is presented online and patients and therapists interact using written messages. Thus, ICBT can potentially increase the availability of effective psychological treatments through reduced therapist time per patient and greater geographical reach. Although the majority of ICBT studies have included adult participants, ICBT has been shown to lead to improvement for children and adolescents suffering from anxiety disorders (Richardson et al., 2010; Vigerland et al., 2013), OCD (Lenhard et al., 2014), chronic pain (Hicks et al., 2006; Palermo et al., 2009), and other health problems (Cushing & Steele, 2010). However, although studies of ICBT for children and adolescents with chronic pain have included participants with abdominal pain, among other pain conditions, these treatments have not been designed to specifically target the full range of symptoms in FGID, for example disturbed defecation in IBS, and dyspeptic symptoms in FD (Rasquin et al., 2006). Thus, it has not been investigated if ICBT tailored specifically for FGID could lead to global symptom relief.

Many CBT protocols for abdominal pain rely on interventions that target stress or teach symptom management techniques, such as applied relaxation, deep breathing and distraction (Sprenger et al., 2011). These interventions are often based on the common observation that abdominal pain is associated with stress (Song et al., 2012). However, studies of adults with IBS have indicated that fear and avoidance of symptoms rather than stress may be the most important factor associated with diminished quality of life and symptom severity (Hazlett-Stevens et al., 2003; Labus et al., 2004, 2007; Jerndal et al., 2010; Reme et al., 2010). Thus, instead of aiming at symptom improvement through symptom control and stress reduction, another target of treatment could be to use exposure treatment i.e., to practice having symptoms in difficult situations, to reduce the fear of symptoms and avoidance behaviors, and to ultimately reduce symptoms. Several studies have shown that exposure treatment is effective for reducing gastrointestinal symptoms in adults with IBS, both in face-to-face format (Ljótsson et al., 2010a; Craske et al., 2011) and delivered over the internet (Hunt et al., 2009; Ljótsson et al., 2010b, 2011a,b, 2014). In these studies, the exposure-based ICBT has targeted avoidance behaviors and fears that are related to both the abdominal pain and the disturbed defecation in IBS.

In summary, no previous study has investigated ICBT specifically tailored for adolescents with FGID and neither has any study investigated the effect of exposure-based CBT, regardless of delivery format, for FGID in adolescents. As preparation for a planned randomized controlled trial, we therefore conducted this open pilot trial to evaluate the feasibility and potential efficacy of a newly developed internet-delivered exposure-based CBT for adolescents diagnosed with pain-related FGID, specifically IBS, FAP or FD.

2. Methods

2.1. Design

This was an open pilot trial with no control group. All adolescents received the same exposure-based ICBT with therapist support during 8 weeks. One parent of each adolescent also participated in a parallel parent-training program during the 8 weeks. The adolescents were assessed at baseline (1 week before treatment), post-treatment (after 8 weeks), and at 6 months follow-up. The adolescents were asked to complete the post-treatment and follow-up assessments regardless of the number of finished modules. We aimed to recruit 25–30 adolescents to achieve a power of at least 80% to detect a within-group effect size of Cohen's $d = 0.6$ on the primary outcome measure, i.e., a moderate treatment effect (Cohen, 1992). This study is reported according to the TREND Statement Checklist for nonrandomized interventions (Jarlais et al., 2011). The study was approved by the Regional Ethical Review Board in Stockholm in December 2011 and is registered on clinicaltrials.gov (reg.no: NCT02033161).

2.2. Eligibility criteria

Eligibility criteria were age 13–17 years, residence in Stockholm County and referral to the study by a treating physician. Moreover, the adolescent and at least one parent had to have easy access to the internet, sufficient computer experience, and be able to read and write in Swedish. Adolescents were not included if there were any concurrent serious medical condition or gastrointestinal organic disorder, any psychiatric disorder that required immediate treatment or psychiatric examination, any current psychological treatment, school attendance less than 80% (because a high absence from school was judged to require a more intensive intervention), or on-going maltreatment, violence or severe parental psychiatric illness in the family.

2.3. Procedure and referral

Fifty-five adolescents and their parents were referred to the study from specialized pediatric clinics in the greater Stockholm urban area. Within this group 46 families declared interest to participate in the study and underwent a screening interview performed by a psychologist (MB). The treating physician referred the adolescents to the study by consulting a pediatric gastroenterologist (OO) ensuring that the patient had been clinically diagnosed with FGID and that he/she had abdominal pain at least every week for at least the two last months (i.e. the requirements for pain frequency and duration according to the Rome III criteria for IBS, FAP or FD were fulfilled). The treating physician also had to ensure that all the following investigations had been normal: growth during childhood, IgA-tissue tranglutaminase, complete blood count, erythrocyte sedimentation rate or C-reactive protein analysis, liver enzymes, and fecal calprotectin (Rasquin et al., 2006). At the screening interview, the phenotype of FGID at study entry (i.e. IBS, FD or FAP) was established using the Rome III criteria (Rasquin et al., 2006). Of the interviewed adolescents, 29 met all eligibility criteria and entered the study. The participants were recruited and treated in two cohorts, 12 families were assessed at baseline in April 2012 with post-treatment assessments in June and 6 months follow-up in December, and 17 families were assessed at baseline in September 2012 with post-treatment assessments in November and 6 months follow-up in May 2013. All outcome measures were completed by the adolescents and were conducted online. See Fig. 1 for participants flow through the study. Parents and adolescents gave written informed consent for participation in the study at the inclusion interview. No compensation was paid for participation.

2.4. Measures

2.4.1. Primary outcome measure

The primary outcome measure was the Gastrointestinal Symptom Rating Scale-IBS version (GSRS-IBS), that includes 13 items about how bothersome gastrointestinal symptoms have been during the past week, e.g., bloating, diarrhea, constipation, early satiety/dyspepsia and abdominal pain (Wiklund et al., 2003). Although developed primarily for IBS, the GSRS-IBS also includes questions about abdominal pain (the main symptom of FAP) as well as early satiety and prolonged fullness (symptoms of FD). The GSRS-IBS has excellent psychometric properties with internal consistency between $\alpha = .74$ (for abdominal pain) and $\alpha = .85$ (for satiety) in adults (Wiklund et al., 2003). The GSRS-IBS has not been validated for adolescents, but there are few alternative measures for adolescents that include the full range of abdominal symptoms present in pain-related FGID.