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

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Article history:
Received 27 June 2014
Received in revised form 13 July 2014
Accepted 14 July 2014
Available online 19 July 2014

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.

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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).
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 et al., 2009). 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). 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, FAP 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.
2.4.2. Secondary outcome measures

Pain Reactivity Scale (PRS) is a self-rating scale designed for children and adolescents with chronic pain, and measures the concern and focus on the pain. The form consists of five questions that are graded on a scale from 1 (“not at all”) to 6 (“very much”). Three items are about feelings and thoughts about worry of pain. Two items relate to worry about not being able to do things because of pain. PRS has satisfactory psychometric properties (Wicksell et al., 2011).

Pain Interference Index (PII) is a self-rating scale that measures interference on function because of pain in children and adolescents. It consists of six questions concerning how much the respondent has difficulty in performing everyday activities, like climbing stairs, running 100 m and going shopping, and general activities such as helping out at home, eating meals and being in school all day. The scale ranges from 1 (no problem at all) to 5 (impossible). It is an often used instrument in pain studies in children and is validated for children and adolescents (no problem at all) to 5 (impossible). It is an often used instrument in pediatric pain studies to measure the impact on functional properties (Wicksell et al., 2011).

Functional Disability Inventory (FDI) measures the difficulty in performing everyday activities (Walker & Greene, 1991). It consists of 15 questions that concern specific activities like climbing stairs, running 100 m and going shopping, and general activities such as helping out at home, eating meals and being in school all day. The scale ranges from 1 (no problem at all) to 5 (impossible). It is an often used instrument in pain studies in children and is validated for children and adolescents with chronic abdominal pain with high internal consistency, α = .86 for boys and α = .91 for girls (Claar & Walker, 2006). FDI is the scale recommended in pediatric pain studies to measure the impact on function (McGrath et al., 2008).

Childhood Anxiety Sensitivity Index (CASI) consists of 18 items on a 3-point scale (1–3), which measures sensitivity to internal sensations that could be symptoms of anxiety. The 18 questions include items like “It scares me when I feel ‘shaky’” or “It scares me when my heart beats fast” as well as items directly relevant to a FGID population as “When my stomach hurts, I worry that I might be really sick”. The scale has high reliability, Cronbach’s α = .84 (Silverman et al., 1991).

Child Depression Inventory (CDI) is a depression scale for children (Kovacs, 1992) and consists of 27 items (0–2 p), each with three statements of varying severity, such as: “I am sad once in a while,” “I’m often sad” or “I am sad all the time.” Respondents are instructed to select the option that best matches their experience over the last two weeks. A clinical cut-off for mild depression at 19 points has been suggested (Friedberg & Sinderman, 2011). Psychometric properties have been validated with acceptable internal consistency, Cronbach’s alpha between α = .83 and α = .89 (Smucker et al., 1986).

Perceived Stress Scale (PSS) is a widely used self-assessment questionnaire (1983) to measure perceived stress. PSS-4 is a short version of the original, and consists of four statements about the feeling of being able to handle problems that need to be addressed. Responses are rated from 0 (“never”) to 4 (“very often”). Reliability for the version with 4 items is α = .72 (Cohen et al., 1983).

2.5. Intervention

2.5.1. Target and content

The treatment was a revised version of a protocol used in previous studies on adults with IBS (Ljótsson et al., 2010a,b, 2011a,b, 2014). The main changes included shorter texts, a simplified language, a colorful design, illustrations and photographs, audio files, examples based on young people’s experiences of having a FGID and an addition of a
parent-training protocol. Before the pilot study was launched, we treated five adolescents and their parents in traditional face-to-face format to evaluate and develop different elements of the protocol. The treatment was separated into six modules that the participants were granted gradual access to during 8 weeks.

Module one included education about FGID and a presentation of 3 fictive adolescents with IBS, FD and FAP respectively. These fictive adolescents were included in all modules to serve as examples for the three diagnoses and how exposure exercises could be performed. A treatment model was introduced explaining how FGID-specific behaviors, aimed at controlling and reducing FGID symptoms, in the long run could maintain and even reinforce symptoms. In this module the participants identified their individual symptoms and how they limited everyday activities in terms of control and avoidance behaviors. An exercise in mindfulness that facilitated interoceptive exposure for abdominal symptoms was assigned as homework.

In the second module, after a detailed review of individual symptom-related behaviors, the participants were introduced to behavior analysis in a three-step-model, the Antecedent–Behavior–Consequence model (A–B–C), with long-term consequences of FGID-specific behaviors explained through an example of an adolescent with IBS. After completing an individual analysis on their own common FGID behaviors, the participants chose an alternative behavior to test during the coming week. This behavior experiment aimed at challenging symptom-catastrophizing as a precursor for later exposure modules.

In module three participants were taught an exercise in acceptance of abdominal symptoms. Participants who had high-frequency use of the toilet were introduced to an exercise to reduce their number of toilet visits, by postponing urgent toilet visits by a few minutes and by following a schedule to avoid numerous visits triggered by discomfort. This exercise aimed at reducing the reinforcing effect of immediate toilet visits following signs of FGID symptoms, and to establish a routine for normal toilet habits. Participants who had no unusual toilet behavior but used other problematic high-frequency behavior, like swallowing, drinking water or spitting to control nausea, or rest to reduce abdominal pain, were encouraged to reduce this instead.

Module four explained in detail how to conduct exposure to gastrointestinal symptoms. In preparing for exposure exercises, i.e. exercises to voluntarily perform behaviors that provoked symptoms, the participants made an individual hierarchy of difficult situations that normally elicited symptoms and planned alternative behavior in response to the symptoms that these situations could elicit. This meant that the treatment encouraged participants to fully engage in activities that could produce gastrointestinal symptoms without trying to control the symptoms.

In the fifth module the exposure continued with the addition to deliberately increase the symptoms, for example by eating large amounts of food that previously were avoided to control gastrointestinal symptoms, and enter situations in which the elicited symptoms would be perceived as difficult to accept. This systematic exposure aimed to reduce fear of having symptoms, which in turn would lead to reduced symptoms and improved functioning. It was explained that by this intensive exposure, the fear of symptoms would no longer be maintained by “threshold thinking” where symptoms would be tolerable only as long as they did not exceed a certain level. Instead participants would get to experience that they could have a lot of symptoms and still function reasonably. Participants worked with these exercises until the last week of treatment when module six was introduced, which was dedicated to relapse prevention.

Parents received four modules over 8 weeks. The first module consisted of psycho-education about FGID and the treatment model and increased positive time with the child in order to reduce attention to pain. Module two targeted how parental response to the child’s symptoms might function to maintain symptom severity and focused on individualized behavior modification in order to counteract this. Module three included the rational for their child’s work with exposure to symptoms and suggestions on how the parent could support their child during the exposure. Module four was a relapse prevention plan. See Fig. 2 for outline of content and timing of both the adolescent and parent treatment protocols.

2.5.2. Therapist contact

During treatment, participants had contact with one of three therapists, of which one was an experienced licensed psychologist (MB) and two were CRT-trained final year psychology students under supervision. The treatment contact was conducted over a secure internet platform developed specifically to deliver ICBT. Participants completed worksheets to report their work with the treatment, and their therapist provided weekly written feedback. Participants could also initiate

**Adolescent protocol**

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**Parent protocol**

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<tr>
<td>Week 1</td>
<td>w. 2</td>
<td>w. 3</td>
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FGID = Functional gastrointestinal disorder

*Fig. 2. Outline of treatment.*
contact by sending messages to the therapist via the platform, primarily for specific questions about the ongoing treatment, and the therapists responded to messages within two working days. The messages that therapists sent to participants, for example with feedback on homework exercises or responses to inquiries, were not written in accordance with a specific manual. However, the content published on the web platform was fixed, which ensured that the participants were exposed to the same main treatment content. All participants also received one telephone call from their therapist during the fifth and sixth weeks of treatment to provide encouragement and discuss how to conduct the exposure exercises. These telephone calls were not logged and the treatment platform did not allow for timing of therapist time spent per participant.

2.6. Analysis

To make use of all available data in an intent-to-treat analysis, we used the multiple imputation procedure in SPSS 20 to impute missing sum scores for study dropouts at the post-treatment and follow-up assessments. All estimates (i.e., means, standard deviations, effect sizes, standard errors, and confidence intervals) were calculated for each of five imputations and then combined into pooled estimates according to “Rubin’s rules” (Rubin & Schenker, 1991) and the small sample correction for pooled degrees of freedom (Barnard, 1999). Two-tailed dependent Student’s t-tests were performed to detect significant within-group differences between baseline and post-treatment, and between baseline and 6-month follow-up. Effect sizes and 95% confidence intervals of changes between assessments were calculated as within-groups d (Borenstein et al., 2011) i.e., the standardized mean difference. Effect sizes were categorized according to Cohen’s suggestion where small, medium, and large effect sizes are $d \geq 0.20, 0.50,$ and 0.80, respectively (Cohen, 1992).

3. Results

3.1. Description of sample

Participants in the pilot study consisted of 29 adolescents living in Stockholm, the majority were girls ($n = 22; 76\%$). Nineteen adolescents ($66\%$) were diagnosed with IBS while $5$ ($17\%$) adolescents had FD and $5$ ($17\%$) had FAP diagnoses. The majority of parents that participated in the parent-training program were female ($n = 26; 90\%$).

3.2. Completion rates

The fifth module did not introduce new treatment content but encouraged continued exposure and the last module only covered relapse prevention. Thus, we considered participants who had completed at least four modules to have been exposed to the full therapeutic content of the treatment and thus as treatment completers. Twenty-two adolescents ($76\%$) finished four or all six modules of the treatment, four ($14\%$) adolescents lagged behind and completed less than two modules, and three ($10\%$) adolescents actively withdrew from treatment (two due to lack of time and one due to computer problems). On average, parents participated in the treatment to the same extent as their child. Attrition rates were low, 27 adolescents ($96\%$) completed the assessments at post-treatment and 26 ($93\%$) completed the 6-month follow-up.

3.3. Results on outcome measures

Means, standard deviations, p-values of dependent t-tests, and effect sizes ($d$) with 95% confidence intervals are reported in Table 1. All effect sizes reported in text are statistically significant at the 95% level. Participants showed significant improvement in gastrointestinal symptoms, as measured by the primary outcome measure GSRS-IBS with moderate effect sizes at post-treatment and 6-month follow-up compared to baseline ($d = 0.50$ and 0.63, respectively). Worry about pain symptoms, as measured by PRS, was significantly improved immediately after treatment with a moderate to large effect size ($d = 0.74$). The follow-up suggested further improvement with a large effect size ($d = 1.05$). Pain interfering with function was not much improved directly after treatment, as shown by FDI with a small effect size ($d = 0.36$), however the pre-to 6-month follow-up effect size was moderate to large ($d = 0.76$). The same pattern was noted for increased function as measured by FDI, with a non-significant change at post-treatment and a moderate effect at follow-up ($d = 0.57$). This was also the case for sensitivity to inner symptoms of anxiety as measured by CASI, a non-significant change at post-treatment and a moderate effect at follow-up ($d = 0.44$). Effects on the levels of perceived stress (PSS-4) and depressive symptoms (CDI) were small or non-significant at both post-treatment and 6-month follow-up assessments.

4. Discussion

The objective of the present pilot study was to investigate the feasibility and potential efficacy of an internet-delivered cognitive behavioral treatment, primarily based on exposure exercises, for adolescents diagnosed with IBS, FAP or FD. The high adherence and low attrition rates indicate that the treatment is feasible for its target population. Results showed significant and moderate treatment effects on the primary outcome measure, GSRS-IBS, that were stable 6 months after treatment. To our knowledge, this is the first study of its kind for this population, both in terms of delivery format and treatment content.

The pronounced effects observed on PRS, are in line with the proposed treatment mechanisms (i.e. reduced avoidance leads to decreased fear of symptoms, which in turn leads to symptom improvement). The design of our study does not allow for any investigation of this as a proposed mechanism, but a mediational study of the exposure-based ICBT

Table 1

<table>
<thead>
<tr>
<th></th>
<th>Pre-treatment</th>
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<th>6 mo follow-up</th>
<th>Pre- to 6 mo follow-up comparison</th>
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<tr>
<td></td>
<td>m (SD)</td>
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<td>t df d [95% CI]</td>
<td>m (SD)</td>
<td>t df d [95% CI]</td>
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<tr>
<td>GSRS-IBS</td>
<td>33.72 (13.62)</td>
<td>27.25 (12.00)</td>
<td>3.24* 26.1 0.50 [0.16, 0.84]</td>
<td>25.90 (10.47)</td>
<td>3.67* 25.6 0.63 [0.24, 1.02]</td>
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<tr>
<td>PRS</td>
<td>16.21 (7.26)</td>
<td>10.99 (6.83)</td>
<td>4.91* 25.9 0.74 [0.39, 1.09]</td>
<td>9.35 (5.48)</td>
<td>5.00* 25.5 1.05 [0.59, 1.59]</td>
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<tr>
<td>PII</td>
<td>13.97 (8.91)</td>
<td>10.77 (8.90)</td>
<td>3.04* 25.4 0.36 [0.11, 0.61]</td>
<td>7.48 (7.92)</td>
<td>4.98* 25.9 0.76 [0.41, 1.12]</td>
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<tr>
<td>FDI</td>
<td>23.59 (7.30)</td>
<td>21.32 (5.69)</td>
<td>1.81 26.0 0.34 [0.06, 0.74]</td>
<td>19.99 (4.90)</td>
<td>2.67 25.9 0.57 [0.10, 1.04]</td>
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<tr>
<td>CASI</td>
<td>32.24 (7.23)</td>
<td>30.88 (6.34)</td>
<td>1.51 26.1 0.20 [0.07, 0.47]</td>
<td>29.18 (6.50)</td>
<td>2.61 25.2 0.44 [0.08, 0.81]</td>
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<tr>
<td>CDI</td>
<td>9.00 (5.57)</td>
<td>8.44 (6.28)</td>
<td>0.60 25.4 0.09 [0.15, 0.33]</td>
<td>8.00 (6.08)</td>
<td>1.12 24.6 0.16 [-0.14, 0.46]</td>
</tr>
<tr>
<td>PSS 4</td>
<td>6.28 (3.14)</td>
<td>5.18 (3.19)</td>
<td>2.24* 23.7 0.35 [0.02, 0.69]</td>
<td>5.25 (3.42)</td>
<td>1.60 21.0 0.31 [-0.10, 0.73]</td>
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Note: All estimates and test-statistics are based on pooling of multiple ($n = 5$) imputations. Abbreviations: GSRS-IBS = Gastrointestinal Symptom Rating Scale-IBS version (range 13–91); PRS = Pain Reactivity Scale (range 0–30); PII = Pain Interference Index (range 0–36); FDI = Functional Disability Inventory (range 15–75); CASI = Childhood Anxiety Sensitivity Index (range 18–54); CDI = Child Depression Inventory (range 0–54); PSS 4 = Perceived Stress Scale 4 item version (range 0–16); $d$ = within-group standardized mean difference (Cohen's $d$); $t = t$-value of dependent t-test, df = degrees of freedom.  
* $p \leq .05$ of dependent t-test.
for adults with IBS showed that reduction in fear and avoidance of symptoms were associated with and preceded improvement in IBS symptoms (Ljótsson et al., 2013), indicating that the same mechanisms of change may have been active in this trial. The low and non-significant effects on function, as measured by PII and FDI, seemed to increase at follow-up. In general, low or non-significant effects were observed on measures of anxiety sensitivity, stress and depression. The average pre-treatment scores on the measures of stress and depression, PSS 4 and CDI, were below clinical ranges (Javorsso et al., 2006; Warttig et al., 2013), but the pre-treatment anxiety scores, CASI, were comparable to clinical populations (Chorpita & Lilienfeld, 1999). This indicates that while the ICBT might lead to symptom improvement and reduced worry about symptoms, it does not seem to address clinical levels of anxiety sensitivity.

The moderate within-group effect size on the GSRS-IBS was notably lower than the large effects that internet-delivered exposure treatment has shown on GI symptoms in adults with IBS (Ljótsson et al., 2010b, 2011a,b, 2014). However, there were important differences between this study and the previously conducted studies on adult patient samples. In one of the studies (Ljótsson et al., 2010b), the adults with IBS in the treatment condition reported a mean of 48.5 (SD = 8.8) on the GSRS-IBS at baseline which was reduced to 32.4 (SD = 12.1) after treatment. In the present study, the mean baseline GSRS-IBS score was 33.7 (SD = 13.6), which is quite close to the average post-treatment symptom burden in the adult trial. A lower initial symptom burden allows for less improvement, which may have affected the observed effect sizes. Reporting fewer or less bothersome gastrointestinal symptoms could partly be an effect of age; young people may not have developed as severe symptoms as adults.

Given the novelty of the treatment content and delivery format, it is encouraging to note that our study shows similar results as the largest randomized study of face-to-face CBT so far conducted in children with functional abdominal pain (Levy et al., 2010). Levy and colleagues reported moderate between-group effects on pain and a moderate within-group increase in the level of function after a brief 3-session CBT-intervention focused on parental behavior, relaxation and improved coping. Our treatment, on the other hand, was based on exposure exercises to gradually decrease the fear of symptoms and avoidance behaviors. ICBT based on stress reduction, relaxation, and symptom management, has also been tried for adolescent abdominal pain with large effects on pain intensity (Hicks et al., 2006; Palermo et al., 2009). However, the studies by Hicks (Hicks et al., 2006) and Palermo (Palermo et al., 2009) did not exclusively include adolescents with an FGID and did not report effects on gastrointestinal symptoms other than pain. In contrast, we included only patients with pain-related FGID: IBS, FAP, and FD in this pilot trial and our main outcome measure, the GSRS-IBS, captures the total symptom burden of all these diagnoses, including abdominal pain, bloating, disturbed defecation, and dyspepsia. Thus, our results suggest that ICBT based on exposure exercises may produce global symptom relief in FGID, with effects similar to those obtained in pain-focused CBT based on relaxation and coping strategies.

A recent meta-analysis has questioned the involvement of parents in CBT for anxiety in children, showing no added effect when parents were involved in treatment (Thulin et al., 2014). In our treatment we chose to involve the parents with a specific parent training protocol, since the literature is rich on examples of how parent behavior might influence children's pain and illness behavior (Whitehead et al., 1982; Levy et al., 2004, 2007; Jellesma et al., 2008) and traditional CBT targeting parent behavior has been shown to relieve their children's symptoms (Levy et al., 2010). Because we were testing a new exposure-based intervention, it also seemed appropriate to provide the parents with a rationale for the often-times difficult exercises that their child would perform.

The most important limitation of this pilot study is the lack of randomization to a control group, limiting the internal validity of the study. We cannot therefore separate the treatment effects from other important factors that may affect symptom levels, for example, spontaneous improvement, season variation in symptoms, regression to the mean or non-specific therapeutic elements such as attention from a caregiver and hope of improvement. However, as the time period between baseline and post-treatment was short and the effect sizes on the primary outcome were fairly large we view the findings as encouraging and unlikely to be fully explained by other factors than the treatment. The choice of a primary outcome measure that has not been validated for adolescents is an important limitation. Other measures of adolescent GI symptoms that were available had other important drawbacks, for example, the questionnaire on pediatric gastrointestinal symptoms (QPGS) (Caplan et al., 2005) is quite extensive and the Child somatization Inventory (CSI) (Walker et al., 2009) has not been validated as a measure of change in gastrointestinal symptoms. In contrast, the GSRS-IBS is brief and has been responsive to treatment in our studies of ICBT for IBS in adults. A separate measure of pain in addition to the GSRS-IBS would also have been desirable to allow for comparison of the results with previous studies of CBT for adolescents with FGID. This study is also limited by the lack of relevant background data about the participants. More information should have been collected and reported about family structure, IBS subtype, and duration of illness. Furthermore, we did not measure if there were any adverse effects of the treatment. The potential adverse effects of psychological treatments are generally believed to be insufficiently studied (Nutt & Sharpe, 2008; Barlow, 2010). Exposure interventions are seldom part of psychological treatments for FGID in children and adolescents, which may be partly explained by the fact that some mental health professionals view exposure as unethical, unsafe and ineffective (Meyer et al., 2014). The ICBT format could also make the exposure exercises harder to perform properly for the patient, because the online therapist has less information about how the exposure exercises are conducted (Rozental et al., 2014). To learn more about how FGID patients react to exposure exercises in an ICBT context, it would have been preferable if adverse events had been monitored and reported.

5. Conclusions

Exposure-based ICBT may be a feasible and efficacious treatment for adolescents diagnosed with IBS, FAP or FD. The results obtained in this pilot study will be used in the planning of future randomized controlled trials, required to confirm these preliminary results. The internet-modality is particularly interesting as it allows any geographical distance between patient and therapist and could potentially be used to increase accessibility to effective psychological treatment for this patient group.

Acknowledgments

The authors wish to thank all adolescents and parents participating in the study, as well as Kristina Aspvall and Klara Hammarlund for the help in the initial development of the protocol.

References

