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Randomized controlled trial of an Internet delivered family cognitive behavioral therapy intervention for children and adolescents with chronic pain

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Abstract

Cognitive-behavioral therapy (CBT) interventions show promise for decreasing chronic pain in youth. However, the availability of CBT is limited by many factors including distance to major treatment centers and expense. This study evaluates a more accessible treatment approach for chronic pediatric pain using an Internet-delivered family CBT intervention. Participants included 48 children, ages 11–17 years, with chronic headache, abdominal, or musculoskeletal pain and associated functional disability, and their parents. Children were randomly assigned to a wait-list control group or an Internet treatment group. Primary treatment outcomes were pain intensity ratings (0–10 NRS) and activity limitations on the Child Activity Limitations Interview, both completed via an online daily diary. In addition to their medical care, the Internet treatment group completed 8 weeks of online modules including relaxation training, cognitive strategies, parent operant techniques, communication strategies, and sleep and activity interventions. Youth randomized to the wait-list control group continued with current medical care only. Findings demonstrated significantly greater reduction in activity limitations and pain intensity at post-treatment for the Internet treatment group and these effects were maintained at three-month follow-up. Rate of clinically significant improvement in pain was also greater for the Internet treatment group in comparison to the wait-list control group. There were no significant group differences in parental protectiveness or child depressive symptoms post-treatment. Internet treatment was rated as acceptable by all children and parents. Findings support the efficacy and acceptability of Internet delivery of family CBT for reducing pain and improving function among children and adolescents with chronic pain.

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Conflict of interests

The present manuscript is submitted exclusively to *Pain* and is not under consideration in any other journal. There are no financial relationships that might lead to a conflict of interest.

Keywords

pain; chronic pain; children; adolescents; randomized controlled trial; Internet; cognitive behavioral therapy

1. Introduction

Chronic and recurrent pain among children and adolescents is now recognized as an important problem because of its prevalence[25;32], negative impact on function and quality of life [10], and economic and social burden[31]. Systematic reviews and meta-analyses have demonstrated that cognitive-behavioral therapy (CBT) is a promising modality that significantly reduces the intensity of pediatric chronic pain[5;6].

CBT has been developed and evaluated primarily in children with headache and abdominal pain[7]. Although there are variations in CBT content, common strategies include relaxation training, activity pacing, problem solving, and distraction techniques[7]. Family CBT refers to the incorporation of parents into treatment, primarily through parental instruction in operant strategies. For example, Sanders, et al.[30] conducted contingency management training for parents and self-management training for children with recurrent abdominal pain, finding significant reductions in disability and pain in children receiving family CBT compared to those receiving standard care.

To improve access to treatment and address other barriers (such as stigma)[16] to receiving psychological treatment, several eHealth technologies have been evaluated to deliver treatment such as handheld wireless devices[17], videoconferencing[35], CD-ROMs[3], and the Internet [9]. Major advantages of the Internet over other forms of technology are the flexibility, ability to update information and to communicate with the patient in real time, and ease in dissemination. The Internet is also widely accessible; in the United States, the majority of youth (93%) use the Internet almost daily, and 75% of American households now have broadband Internet access[26]. Moreover, health information is cited as the primary information accessed online[27].

There is emerging support for the efficacy of Internet-delivered psychological treatments for children with acute and chronic health conditions[33]. One prior investigation used the Internet combined with therapist telephone contact to deliver distance treatment to youth with headache and recurrent abdominal pain[9], finding significant reduction in pain levels for children receiving the distance treatment versus a standard medical care control group. A significant limitation of this Internet intervention is that it did not utilize the interactivity, multimedia formats, personalization, and communication possible with the Internet, and instead relied heavily on telephone interaction with a therapist, thus limiting the potential dissemination of the intervention.

We developed and evaluated an interactive, self-guided Internet intervention to deliver family CBT. In a randomized controlled trial, we evaluated the Internet treatment among a clinically referred population of youth with mixed chronic pain conditions. Following recommendations of the PedIMMPACT group[18], we sought to assess multiple, relevant outcome domains that have been evaluated in face-to-face CBT studies including pain intensity, physical functioning (activity limitations), and emotional functioning (depressive symptoms). In addition, because the Internet intervention incorporated significant content directed toward parental behaviors, we examined effects on parental protective responses. We hypothesized that children receiving Internet treatment would have improvements in these domains in comparison to children in a wait-list control condition. Treatment improvements were hypothesized to be maintained in

children receiving Internet intervention at 3 months follow up. In addition, we sought in exploratory analyses to examine acceptability, satisfaction, and compliance with Internet treatment.

2. Methods

2.1. Participants and setting

Participants included 48 children and adolescents, ages 11–17, with chronic pain and their parents. Children presenting as new patients were recruited over a 2-year period through specialty care physician referral from a multidisciplinary pediatric pain clinic, pediatric neurology clinic, and pediatric gastroenterology clinic at an academic health center in the northwestern United States. The study was approved by our Institutional Review Board and parents gave informed consent and children gave assent prior to any research procedures.

2.2. Inclusion/exclusion criteria

Inclusion criteria consisted of: (a) ages 11 to 17 years, (b) chronic idiopathic pain present over the previous 3 months, (c) pain occurs at least once per week, (d) pain interferes with at least one area of daily functioning, and (e) the child was a new patient being evaluated in the specialty clinic. If families met criteria but did not have access to a computer, a laptop was provided (6% of families). Participants were excluded if (a) the child had a serious comorbid chronic condition (e.g., diabetes, cancer), (b) was non-English speaking, or (c) was already receiving CBT for chronic pain.

2.3. Study flow

Figure 1 developed from the CONSORT statement[19] illustrates the enrollment progression of participants throughout the treatment study. As shown, 88 children were assessed for eligibility and 40 were excluded. Of the 40 excluded, 23 did not meet inclusion criteria ($n=18$ due to pain not occurring frequently enough, $n=1$ due to non-English speaking, $n=2$ due to receiving CBT, and $n=2$ due to presence of chronic health condition); 5 families refused to participate due to lack of time or interest, and 12 families were unable to be reached to complete pre-treatment assessments (passive refusals). Therefore, our participation rate was 74%. The final sample consisted of 48 participants randomly assigned to a treatment condition, Internet treatment ($n = 26$) or wait list control ($n = 22$). Both treatment groups continued to receive medical care for their pain condition through a specialty medical clinic. The majority of children in both treatment groups received 0 to 2 visits with the specialty clinic during their study participation.

One participant in the Internet treatment condition actively withdrew during the treatment phase due to computer problems. Three participants ($n = 2$ Internet treatment group, $n = 1$ waitlist control group) did not complete post-treatment assessments (Fig 1). Thus, our attrition rate was 8.4%. Intent-to-treat analysis was used, and therefore the effective number of cases analyzed for the primary and secondary outcome analyses was $n=26$ for the Internet treatment group and $n= 22$ for the wait- list control group.

2.4. Randomization procedure

A fixed allocation randomization scheme was used. Specifically, we used blocked randomization with blocks of 10 to assign participants to the two treatment conditions during the course of randomization. An online random number generator was used to produce the blocked randomization. Group assignments were identified by ID number in sealed envelopes during the 24 month recruiting period. Following completion of all pre-treatment assessments, a research coordinator opened the sealed envelope to reveal the group assignment. This

information was then programmed into the Web-MAP system, which generated a message on the web site to each study participant revealing their instructions for the treatment phase.

2.5. Assessment procedures

2.5.1. Pre- and immediate post-treatment assessment—Prior to randomization, participants completed a pre-treatment assessment at home, including an online daily diary for 7 days to record pain and activity limitations and questionnaire measures that were returned via postal mailing. Immediate post-treatment assessments were conducted at 8 to 10 weeks. Daily diaries monitoring pain and activity limitations were completed online and other measures were administered via questionnaires sent in postal mailings. Participants in the wait-list control condition were offered access to the Internet treatment following receipt of their completed post-treatment assessment at 8–10 weeks.

2.5.2. 3-month follow-up assessment—Participants in the Internet treatment group only completed a 3-month follow-up assessment to evaluate maintenance of treatment effects. These participants completed retrospective questionnaire measures at home and returned via postal mailing.

2.6. Primary outcome variables

2.6.1. Activity limitations—Activity limitations were assessed using the Child Activity Limitations Interview (CALI)[21], which includes both retrospective and prospective versions. On the retrospective version, children respond to an item-selection list of 21 activities and choose the eight activities that are the most difficult or bothersome to them due to recurring pain. Importance of each activity to the individual child is rated on a 5-point scale from (0) *not very important* to (4) *extremely important* to ensure that children identify activities that are considered relevant in their daily lives. The primary score for the retrospective version of the CALI is derived from the difficulty ratings of the eight most difficult activities, which are obtained on a 5-point scale from (0) *not very difficult* to (4) *extremely difficult*, with total scores ranging from 0 to 32. The CALI was completed at each assessment period.

The prospective version of the CALI was used in daily diaries at the pre- and post-treatment assessment periods. The same eight activities that were identified by the child were asked about daily at these assessment periods for 7 days each time. Responses were rated on a 5-point scale (0–4) with higher scores indicating greater perceived difficulty with activities. Mean scores across each 7-day period were used in analyses.

2.6.2. Pain—A daily online diary was used to assess self-reported pain at the pre-treatment and post-treatment assessment periods. Children reported on presence and intensity of pain. Pain intensity was assessed using an 11-point numerical rating scale (NRS) with anchors of 0= no pain to 10= worst pain. For the purpose of analyses, the mean pain intensity reported (range 0 to 10), and a dichotomous rating of clinically significant improvement using a criterion of 50% reduction in pain intensity were computed from the 7 days of diary data. This criterion is a standard metric previously used in pain clinical trials[7;20].

Children also completed a questionnaire at each assessment period asking for their retrospective report of usual pain over the past month. Usual pain intensity was assessed using an 11-point numerical rating scale (NRS) with anchors of 0= no pain to 10= worst pain.

2.7. Secondary outcome variables

2.7.1. Depressive symptoms—As a measure of emotional functioning, children completed the major depressive disorder (MDD) subscale of the Revised Child Anxiety and Depression Scale[2] (RCADS). T-scores are calculated based upon the child's gender and

grade in school. The RCADS has previously demonstrated good internal consistency, adequate test-retest reliability, and validity[2]. Coefficient alpha for the MDD subscale in the current sample was $\alpha=0.85$.

2.7.2. Parental response to pain behavior—Children and their parents completed the Protect subscale from the Adult Responses to Children's Symptoms (ARCS), a measure assessing parental responses to child pain behavior[34]. The Protect subscale assesses solicitous parent responses (e.g., providing extra attention). Respondents rate on a five-point Likert scale the frequency with which parents engage in specific behaviors in response to their child's pain. Coefficient alpha for the Protect subscale in the current sample was $\alpha=0.83$ for parent report and $\alpha=0.84$ for child report.

2.7.3. Treatment acceptability and satisfaction—Children and their parents in the Internet treatment group completed an adapted version of the Treatment Evaluation Inventory-Short Form[14]. For the purposes of analysis, responses to two items measuring acceptability ("I find this treatment to be an acceptable way of dealing with adolescent's pain") and global satisfaction ("Overall, I have a positive reaction to this treatment") were examined. Response options range from (1) *Strongly Disagree* to (5) *Strongly Agree*.

2.8. Treatment Conditions

2.8.1. Wait-list control group—Participants in the wait-list control group continued with their medical care recommended by their subspecialty physician, which for most patients involved 0 to 2 visits at the specialty clinic over the treatment period. Children received access to the web site to complete diary assessments at pre- and post-treatment assessments. Participants were asked not to initiate psychotherapy during the 8-week wait period. Following completion and receipt of their post-treatment (8–10 week) assessment, families were offered the opportunity to receive the Internet treatment using the same procedures outlined below. Of the 22 children in the wait-list control group, 68.2% chose to access the Internet treatment following the post-treatment assessment, and 50% completed at least half of the intervention modules.

2.8.2. Internet treatment group—Participants in the Internet treatment group also continued with their medical care recommended by their subspecialty physician (involving on average 0–2 specialty care visits), and were asked not to initiate psychotherapy during the 8-week treatment period. Following completion of pre-treatment assessments and randomization, children and parents were given access to the treatment content of the web program (Web-MAP: Web-based Management of Adolescent Pain) consisting of two separate web sites, one for child access and one for parent access. Web-MAP has three main sections: passport (home) page (see Figure 2), treatment modules, and daily diary. Children and parents were each asked to log on once per week to the web site to complete a module and assignment, designed to take about 30 minutes to complete. Total treatment duration was approximately 9 hours per family (4 hours child modules, 4 hours parent modules, 1 hour therapist time).

The program was travel-themed, and children and parents visited eight modules in destinations (e.g., China, Costa Rica; 1 per week), each focused on a different skill/topic. Children and parents interacted with the web program through completing fillable responses to queries (e.g., listing current stressors), which then tailored and personalized instructions and assignments. There were also several tasks that children completed using animations on the program (e.g., packing a suitcase to learn tips on relaxation). Web-MAP contains over 200 page views and makes use of multimedia elements to enhance delivery of information such as video files of teens that have experienced chronic pain and their parents, and audio files of deep breathing and muscle relaxation instructions.

Each treatment module served as an online analog for the weekly sessions typically used when delivering CBT in a face-to-face format. The same general structure was used in each module: a summary of what will be learned and why this information is important, fun facts about the destination being visited, main content, a question-and-answer game to test retention of information, and an assignment screen that presented instructions for carrying out a specific skill over the coming week. At some destinations, children received online postcards from previous places they had visited reminding them to continue to practice core treatment skills.

Assignment completion: In six child modules and in seven parent modules, there were assignments corresponding with skills being taught in the modules. Assignments were designed to be completed approximately once per week. These were similar to weekly homework assignments used in face-to-face CBT and focused on skills acquisition during the treatment program. For example, during the week that children completed the relaxation module, they were instructed to participate in daily relaxation practice using audio files provided on the website. For their assignment, they reported on level of ease and helpfulness of relaxation, and were asked to comment on their practice. Assignment completion was requisite before children could proceed to the subsequent module.

Users submitted their completed assignments for review by the Web-MAP online therapist. The online therapist was a Ph.D. level psychology postdoctoral fellow who had one year of specific experience in delivery of face-to-face CBT to children with chronic pain. In addition, a manual was developed prior to the RCT with examples of therapist responses to assignments. The therapist responded via a Message Center to each assignment to review progress, encourage continued skills practice, and to help problem solve barriers to implementing skills. These brief messages specifically included a summary of the child or parent's reported progress, positive reinforcement for carrying out assignments, and encouragement to continue with the program.

Therapist time devoted to these responses was approximately 5 minutes per assignment for a total of 30 minutes per participant (60 minutes per family). Development and usability testing of the Web-MAP program is described in a previous manuscript[15].

2.8.2.1. Theoretical framework and specific content: The primary theoretical frameworks used to guide the intervention were cognitive-behavioral and social learning frameworks. Core components of CBT were incorporated into the modules. The eight child modules were: 1) education about chronic pain, 2) recognizing stress and negative emotions, 3) deep breathing and relaxation, 4) distraction, 5) cognitive skills, 6) sleep hygiene and lifestyle, 7) staying active, and 8) relapse prevention. The eight parent modules included: 1) education about chronic pain, 2) recognizing stress and negative emotions, 3) operant strategies I, 4) operant strategies II, 5) modeling, 6) sleep hygiene and lifestyle, 7) communication, and 8) relapse prevention.

The child modules included instruction in identifying stress, applying deep breathing and progressive muscle relaxation, and modifying cognitions about pain and functional ability. In addition, one lesson in the child program focused on enhancing children's sleep habits (instruction in adequate sleep duration and sleep habits) and increasing their physical activity participation through goal setting and activity pacing. Parent modules sought to provide skills in adaptive communication and interaction patterns. Operant procedures, similar to previous research[30] were taught including use of reinforcement for the child's maintenance of normal activity despite pain. Parents were instructed in creating privilege and point-based reward systems to target specific functional activities for reinforcement (e.g., school attendance, exercise). In addition, specialized content in the parent modules focused on the importance of modeling, supporting independence, and enhancing communication with their child.

3.0. Statistical Analyses

Sample size calculations were performed on the outcome pain intensity based on the results of previous studies of CBT for treatment of chronic pain in children[5] and included an adjustment to compensate for attrition, estimated at 20% from similar studies. Our study was powered to enroll 60 subjects and with 20% attrition, we expected to retain 48 subjects. There was minimal missing data on either diaries (all children had a minimum of 5 days of completed entries) or questionnaires. One participant in the wait-list control group did not complete a post-treatment diary but did complete post-treatment questionnaire data. All analyses are based on the intent-to-treat sample using the last observation carried forward method for imputation of values for participants dropping out of conditions.

To verify that randomization produced equivalent groups, t-test and chi-square analyses were conducted. Primary hypothesis testing was conducted with an analysis of covariance (ANCOVA) using the GLM procedure in SPSS v17.0. Group was specified as a fixed between-subject factor, and pre-treatment values were entered as covariates in these models. Separate ANCOVAs were computed for the primary outcomes (pain and activity limitations) and for the secondary outcomes (depression and parental protectiveness). Effect sizes for ANCOVAs are expressed as η^2 which are interpreted using univariate guidelines as a small effect size at 0.01, a medium effect size at 0.06, and as a large effect size when greater than or equal to 0.13 [8]. Means (SDs) are reported for the unadjusted values on all outcomes.

Clinically significant improvement in pain intensity was evaluated with a dichotomous responder analysis. Participants attaining at least a 50% reduction in pain intensity scores from pre- to post-treatment were considered to be responders or clinically significantly improved on pain intensity. Chi-square analysis was used to evaluate the association between clinically significant improvement and group assignment.

To examine maintenance of treatment effects in the Internet treatment group at the 3-month follow-up, one-way ANOVA with repeated measures was conducted for pain, activity limitations, depression, and parental protectiveness outcomes. The Wilks' Lambda multivariate test statistic was used for these analyses. In exploratory analyses, we examined treatment acceptability, satisfaction, and compliance using descriptive statistics, and examined the relationship between compliance and child treatment outcomes using Pearson correlations.

4. Results

4.1. Sample characteristics

Table 1 summarizes the demographic and pain-related characteristics of the sample. Participants were 48 children and adolescents (13 male, 35 female) between the ages of 11 and 17 years ($M=14.8$, $SD=2.0$). Children were primarily Caucasian (89.6%) and middle class as indicated by annual household income between \$50,000 and \$100,000 reported by the majority of parents (53.3%). Highest parental education attained included high school diploma or less (12.8%), vocational school or some college (40.4%), college degree (34.0%), and graduate or professional school (12.8%). The majority of parents reported being married (79.2%). Child participants were referred to the treatment study for abdominal pain (50%), followed by headache (25%), and musculoskeletal pain (25%) (e.g., back pain, limb pain). Many children in the sample reported pain in multiple locations (1 location = 41.7%; 2–3 locations = 35.5%; 4 or more locations = 23.0%; see Table 2). Most children were experiencing daily pain (72.9%) that was rated as moderate to severe intensity. Participants had pain present on average for 30 months with a range of 5 months to 13 years. Families lived on average 67 miles from the treatment center with a range from 6.7 to 273 miles.

4.2. Group equivalence on demographic and pre-treatment variables

Treatment groups were equivalent on gender, race, primary pain problem, and distance from treatment center. There was a slight difference in child age, with the Internet treatment group being younger than the wait-list control group but this did not reach significance ($p = .07$). We examined the relationship between age and the primary and secondary treatment outcome variables in Pearson bivariate correlations and because there were no significant associations we chose to not include age as a covariate in any subsequent analyses. Treatment groups were also equivalent on pre-treatment measures of pain intensity, activity limitations, depression, and parental protectiveness.

4.3. Primary treatment outcome analyses: Pre to post-treatment results

4.3.1. Activity Limitations—Baseline-adjusted ANCOVAs to evaluate group differences on child-reported activity limitations on the online diary at post-treatment were significant, $F(1, 45) = 9.25$, $p = .004$ with a large effect size of partial $\eta^2 = .17$. As shown in Table 2, mean post-treatment values were as expected, with the Internet treatment group having a smaller post-treatment mean ($M = 3.87$) relative to the wait-list control group ($M = 6.61$) indicating greater reduction in diary-reported activity limitations for the Internet treatment group. Similarly, retrospective report of activity limitations as measured by the questionnaire version of the CALI was significantly lower post-treatment in the Internet treatment group relative to the wait-list control group, $F(1, 45) = 4.20$, $p = .05$ with a medium effect size of partial $\eta^2 = .09$, and means of 12.69 (Internet treatment group) compared to 16.00 (wait-list control group).

4.3.2. Pain intensity—Baseline-adjusted ANCOVAs to evaluate group differences on child-reported pain intensity on the online diary at post-treatment was also significant, $F(1, 45) = 5.28$, $p = .03$, with a medium effect size of partial $\eta^2 = .11$. As shown in Table 2, mean post-treatment values were as hypothesized, with the Internet treatment group having a lower post-treatment mean ($M = 3.69$) relative to the wait-list control group ($M = 4.86$) indicating greater reduction in diary-reported pain intensity for the Internet treatment group. Retrospective ratings of pain in the past month decreased in the Internet treatment group ($M = 5.15$), but this was not significantly different than the wait-list control group ($M = 5.45$) immediately post-treatment, $F(1, 45) = .53$, $p = .47$, partial $\eta^2 = .01$.

4.3.3. Clinically significant improvement in pain—A dichotomous responder analysis for pain intensity was computed using clinically significant improvement in pain (defined as at least a 50% reduction in diary-reported pain intensity scores from pre- to post-treatment), a standard metric used in pain clinical trials. The mean percent pain reduction in the Internet treatment group was 33.2%. The rate of clinically significant improvement in pain intensity was greater in children in the Internet treatment group versus children in the wait-list control group for the intent to treat sample (38.5% vs. 13.6%, $X^2(1) = 3.72$, $p = .05$). Among treatment completers ($n=44$), 10 of 23 children in the Internet treatment group and 3 of 21 children in the wait-list control group achieved clinically significant improvement in pain intensity, resulting in a number needed to treat based on these results of 3.4.

4.4. Secondary outcome analyses: Pre to post-treatment results

Baseline-adjusted ANCOVAs were also computed to evaluate group differences on secondary outcomes of child depressive symptoms and parental protectiveness. Mean post-treatment values were similar between groups and ANCOVAs did not reveal a significant group effect on either depression or parental protectiveness (see Table 2).

4.5. Within subject maintenance of treatment effects: Three-month post-treatment follow-up results in the Internet treatment group

One-way ANOVAs with repeated measures were conducted to evaluate the maintenance of treatment effects at three-month follow-up for the Internet treatment group on retrospective reports of pain, activity limitations, depressive symptoms, and parental protectiveness. These analyses were conducted with the intent to treat sample with last observation carried forward, and using Bonferroni contrasts. Child retrospective report of pain intensity declined in the treatment group Wilks' Lambda = .44, overall $F(2, 24) = 15.02$, $p < .001$. Contrasts showed that treatment effects were maintained at follow-up, with T1-T2 significant ($p = .02$) and T1-T3 significant ($p < .001$). This effect size was large, with partial $\eta^2 = .34$. Child retrospective report of activity limitations also declined in the treatment group, Wilks' Lambda = .46, overall $F(2, 24) = 14.03$, $p < .001$. Contrasts showed that treatment effects were maintained at follow up, with T1-T2 and T1-T3 contrasts significant (p values $< .001$). This effect size was large with partial $\eta^2 = .43$. Depressive symptoms were significantly lower at three-month follow-up than at post-treatment, Wilks' Lambda = .78, overall $F(2, 24) = 3.47$, $p = .05$, with contrasts significant for T2-T3 ($p = .04$). This effect size was medium with partial $\eta^2 = .07$. Parent report of protectiveness declined across the three time points, Wilks' Lambda = .38, overall $F(2, 24) = 19.71$, $p < .001$, with contrast showing T1-T2 and T1-T3 significant ($p < .001$), as well as T2-T3 ($p = .04$). This effect size was large, with partial $\eta^2 = .54$. Child report of parental protectiveness declined over time, Wilks' Lambda = .77, overall $F(2, 24) = 3.52$, $p = .05$, and this was also a large effect size of partial $\eta^2 = .19$. Contrasts were significant for T1-T2 ($p = .05$) and T1-T3 ($p = .04$).

4.6. Exploratory analyses: Treatment acceptability, satisfaction, and compliance

Children and parents in the Internet treatment group reported moderate to high ratings of treatment acceptability (child report $M = 3.55$, $SD = .80$, parent report $M = 3.82$, $SD = .50$). Global satisfaction was also moderate to high (child report $M = 3.68$, $SD = .84$, parent report $M = 4.09$, $SD = .61$). Parent and child report of acceptability and satisfaction were positively correlated ($r = .50$, $p = .02$; and $r = .53$, $p = .01$, respectively). The vast majority of children (91%) and all parents rated the treatment as acceptable and were satisfied with the treatment (ratings > 3). The majority of children and parents in the Internet treatment condition were compliant with completing treatment modules. Children completed a mean of 7.11 ($SD = 1.86$) modules, with 20 of 26 children (77%) completing all 8 modules. Parents completed a mean of 6.42 ($SD = 2.24$) modules, with 14 of 26 parents (54%) completing all modules. The mean number of modules completed by families (child and parent modules summed) was 13.54 ($SD = 3.8$). The number of modules completed by families was not significantly correlated with immediate post-treatment primary or secondary outcomes.

5. Discussion

Findings support the efficacy of Internet-delivered family CBT in significantly reducing pain intensity and improving function for children and adolescents with chronic pain. This study extends previous work on distance treatment in youth with chronic pain[3;9] by demonstrating the efficacy of a self-guided Internet intervention with minimal therapist time. Similar to recent findings of Internet delivered treatment in adults with health conditions including chronic pain and headache[4], this mode of treatment delivery can also be successfully used with children and their parents.

Advances in the mode of treatment delivery of CBT in pediatric chronic pain are critically needed as most children and adolescents do not have access to specialized treatment. In many parts of the world, when referred for specialty evaluation and care, children with chronic pain encounter significant difficulties accessing care due to a shortage of dedicated pain treatment

facilities for children. For example, in a recent survey of Canadian pediatric pain treatment facilities, only five centers were available with sufficient expertise to treat children and these were all located in major urban cities[24]. These authors concluded that a significant proportion of children with chronic pain in Canada, particularly those living in rural areas, did not have adequate access to care[24]. A similar scenario exists in the United States and other parts of the world. Use of the Internet to deliver psychological treatment to children with chronic pain may be critical to providing access to a large number of patients at a low cost.

Our findings also extend previous work on family CBT[29;30] demonstrating its efficacy among children with mixed chronic pain conditions. Parents received equal treatment time as their child with our Internet treatment. This dose of treatment directed toward the parent is more intensive than that reported in previous research in which interventions conducted with parents have primarily relied on brief educational sessions[13;29;30]. In addition to education and operant strategies, we incorporated content directed toward parent-child communication, modeling, and parental stress management. It is possible that these parental strategies contributed to the positive child outcomes. Future studies are needed to compare child focused treatment to parent focused treatment and their combination to better understand the contribution of each to treatment success.

We also extend knowledge of parent outcomes in the present study. Specific hypothesized effects of family CBT have not previously been evaluated in parents despite the inclusion of parents in treatment. We measured changes in parental behaviors (i.e., parental protectiveness) that were targeted in treatment. However, unexpectedly, we did not demonstrate any significant group differences on this specific parental behavior, although in within- subjects analyses, we found that parental protectiveness diminished over time in parents in the Internet treatment condition. Because outcomes related to parental behaviors have not previously been reported in family CBT studies, it is not possible to compare our findings to similar studies. Perhaps our multicomponent parent treatment was not specific to addressing parental protectiveness, and/or that the assessment tool used is not stable enough over time to adequately measure treatment-related changes. Test-retest reliability of the Adult Responses to Children's Symptoms measure has not yet been examined[34]. In addition, we found that parents were less compliant than children in completing treatment modules and this may have affected their exposure to intervention content. Future studies should evaluate strategies to encourage parental participation with treatment as well as measure additional relevant parental outcomes such as parenting stress, parental emotional functioning, and parent-child communication.

Within the Internet treatment group, effects of treatment appear to be maintained over time and, for some outcomes, continued improvements were demonstrated at the three month follow-up. However, these results should be interpreted with caution as due to the wait-list control design, we did not have available comparison data from the control group at 3-month follow-up. Therefore, we cannot determine that continued improvements in children in the treatment condition were due specifically to the Internet intervention. In future studies, it will be important to use study designs that allow for examination of maintenance of treatment effects. In a recent systematic review of psychological therapies for chronic pain in children, in the few studies that have examined maintenance of treatment effects[6], CBT was found to produce enduring changes in pain intensity.

A strength of the present study is that the outcome assessment plan included relevant multidimensional outcomes. Although pain intensity has been the most frequently reported outcome from chronic pain trials, it is critical that research on pediatric chronic pain also incorporate measurement of functional outcomes related to pain[22]. Recently, recommendations have been made by the Pediatric Initiative on Methods, Measurement, and Pain Assessment in Clinical Trials (Ped-IMMPACT) concerning outcome domains that are

important to assess in clinical trials with children and adolescents who have chronic or recurrent pain[18]. Eight domains were recommended: pain intensity, physical functioning, symptoms and adverse events, global satisfaction with treatment, emotional functioning, role functioning, sleep, and economic factors. In this study, in addition to pain intensity, we evaluated treatment effects on children's physical and emotional functioning.

Our study is one of the first to demonstrate changes in children's physical functioning with psychological treatment. Treatment content directed toward both children and parents emphasized a return to usual physical activities. Children received instruction in activity pacing and goal-setting around physical activities. The involvement of parents in treatment may have also contributed to these improvements. Our findings are unique in that there is little evidence from RCTs that children's physical or role functioning is changed by psychological treatments [6;23] . One exception is a recent RCT of acceptance and exposure strategies in which a significant improvement on one measure of function, pain interference, was found for the treatment group in comparison to a usual care group[36].

Group differences in children's emotional functioning, specifically in their depressive symptoms, from pre to post treatment were not found. Overall, depressive symptoms were in the subclinical range both before and after treatment. However, at three month follow-up depressive symptoms were significantly decreased in the Internet treatment group. It is unclear whether a lengthier period of time was necessary for changes to occur in children's emotional functioning or whether measurement issues contributed to this finding. Somewhat surprisingly, depressive symptoms have also been unchanged in other RCTs of psychological treatments for chronic pain[6].

Because of the novel method of treatment delivery, we were interested in documenting treatment acceptability, satisfaction, and compliance with treatment. The participation rate in this study (74%) was similar to rates of participation in studies of face-to-face CBT[13;29], suggesting acceptability. In our exploratory analyses, we also found that almost all children and parents rated the Internet treatment as acceptable and satisfactory. Moreover, the attrition rate was very low with only 8% of children dropping out of the trial. Overall, child and parent compliance with the program was good, with the majority of participants completing the entire program. As noted, child compliance was higher than parental compliance. However, treatment compliance was not related to treatment outcomes. Although Internet interventions present unique opportunities to measure real-time usage of an intervention, tracking the amount of time spent on web sites is challenging due to factors such as variations in individual user habits (e.g., not logging out or closing browser windows) and internet connectivity issues. In other Internet intervention studies, user characteristics such as comfort with the Internet have predicted treatment success[1], and may be important to assess in future studies.

Our findings should be interpreted in light of several limitations. Because we did not include a measure of treatment expectancy or an attention comparison condition, we are unable to know the influence of expectancies nor are we able to separate treatment from placebo effects. Future studies with a placebo attention condition would help to answer this question. We were also unable to examine moderators of treatment outcome. This is a critical area of future investigation, as to date, there is virtually no available information on individual differences in treatment response in pediatric chronic pain trials[6]. In addition, generalizability is not addressed as our sample included a relatively homogenous group of patients (mostly Caucasian and middle class) recruited from one treatment center. Last, our design would have been strengthened by inclusion of all data collection online. It is unknown if there might be differences in how children responded to the written questionnaire measures versus to the online questionnaire measures; however data from other studies suggest high levels of

correspondence between written and computer-based administration of measures of domains such as pain and quality of life (e.g., [11;12]).

Randomized controlled trials of psychological therapies for chronic pain in children remain limited, particularly for children with pain conditions other than headache[6]. This study contributes to this growing knowledge base of psychological therapies for pediatric chronic pain. As yet, the majority of studies of Internet interventions are limited to feasibility and efficacy trials[28], and have not involved large-scale dissemination. However, if future research continues to find results that support this treatment modality, Internet interventions such as ours may have the potential for broad dissemination.

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Figure 1.
Study Participant Flow.

Figure 2.
Internet treatment condition: Web-MAP home page (child site)

Table 1
Characteristics of the Sample, Combined and by Treatment Group

	Combined Sample (n=48)	Internet Treatment (n=26)	Wait-list Control (n=22)
Characteristic	N(%) / M(SD)	N(%) / M(SD)	N(%) / M(SD)
Age (years)	14.8 (2.0)	14.3 (2.1)	15.3 (1.8)
Gender			
Male	13 (27.1%)	6 (23.1%)	7 (31.8%)
Female	35 (72.9%)	20 (76.9%)	15 (68.2%)
Race			
White/Caucasian	43 (89.6%)	21 (80.8%)	22 (100%)
Hispanic	3 (6.2%)	3 (11.5%)	0
Other	2 (4.2%)	2 (7.7%)	0
Referral Pain Problem			
Headache	12 (25.0%)	4 (15.4%)	8 (36.4%)
Abdominal pain	24 (50.0%)	14 (53.8%)	10 (45.5%)
Musculoskeletal pain	12 (25.0%)	8 (30.8%)	4 (18.2%)
Pain Location ^a			
Face	5 (10.4%)	2 (7.7%)	3 (13.6%)
Head	20 (41.7%)	10 (38.5%)	10 (45.5%)
Abdomen	30 (62.5%)	18 (69.2%)	12 (54.5%)
Chest	14 (29.2%)	6 (23.1%)	8 (36.4%)
Low Back	17 (35.4%)	10 (38.5%)	7 (31.8%)
Spine	9 (18.8%)	5 (19.2%)	4 (18.2%)
Shoulder or neck	11 (22.9%)	6 (23.1%)	5 (22.7%)
Arm or hand	5 (10.4%)	4 (15.4%)	1 (4.5%)
Leg or foot	9 (18.8%)	7 (26.9%)	2 (9.1%)
Pain frequency			
1 – 2 ×/week	6 (12.5%)	2 (7.7%)	4 (18.2%)
3 – 6 ×/week	7 (14.6%)	4 (15.4%)	3 (13.6%)
Daily	35 (72.9%)	20 (76.9%)	15 (68.2%)
Distance from treatment center (miles)	66.2 (72.4)	62.9 (65.8)	70.1 (80.9)

^aNote: Percentages do not sum to 100% as pain was reported in multiple locations.

Table 2
Unadjusted descriptive statistics on primary and secondary treatment outcomes by treatment condition

Variable	Wait-list Control <i>n</i> = 22 Mean (SD)	Internet Treatment <i>n</i> = 26 Mean (SD)
Pain Intensity- online diary		
Pre-treatment T1	5.17 (1.65)	5.45 (2.25)
Post-treatment T2 ^a	4.76 (1.84)	3.54 (2.42)
Pain Intensity- retrospective report		
Pre-treatment T1	6.16 (1.84)	6.63 (1.87)
Post-treatment T2 ^b	5.45 (2.04)	4.96 (2.18)
3-month follow-up T3 ^c	n/a	4.27 (2.00)
Activity Limitations- online diary CALI		
Pre-treatment T1	6.30 (4.61)	5.89 (4.92)
Post-treatment T2 ^a	6.62 (4.76)	3.60 (2.86)
Activity Limitations- retrospective CALI		
Pre-treatment T1	18.82 (6.59)	20.54 (5.80)
Post-treatment T2 ^b	16.00 (6.34)	12.69 (6.29)
3-month follow-up T3 ^c	n/a	10.27 (8.04)
Depressive symptoms		
Pre-treatment T1	57.91 (15.04)	56.08 (13.65)
Post-treatment T2 ^b	61.59 (18.67)	58.96 (13.10)
3-month follow-up T3 ^c	n/a	53.45 (16.16)
Parental protectiveness- child report		
Pre-treatment T1	23.18 (8.97)	26.92 (9.78)
Post-treatment T2 ^b	21.45 (11.83)	22.91 (8.28)
3-month follow-up T3 ^c	n/a	19.86 (12.36)
Parental protectiveness- parent report		
Pre-treatment T1	24.55 (7.88)	28.77 (8.11)
Post-treatment T2 ^b	19.11 (10.15)	19.91 (9.76)
3-month follow-up T3 ^c	n/a	15.54 (8.57)

^a Wait-list control *n* = 21, Internet Treatment *n* = 23

^b Waitlist Control *n* = 22, Internet Treatment *n* = 23

^c Internet Treatment *n* = 23