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Evaluating the effectiveness of exposure and acceptance strategies to improve functioning and quality of life in longstanding pediatric pain – A randomized controlled trial

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ABSTRACT

Although several studies have illustrated the effectiveness of cognitive behavior therapy (CBT) on adult pain patients, there are few randomized controlled trials on children and adolescents. There is particularly a need for studies on pediatric patients who are severely disabled by longstanding pain syndromes. Acceptance and Commitment Therapy, as an extension of traditional CBT, focuses on improving functioning and quality of life by increasing the patient's ability to act effectively in concordance with personal values also in the presence of pain and distress. Following a pilot study, we sought to evaluate the effectiveness of an ACT-oriented intervention based on exposure and acceptance strategies and to compare this with a multidisciplinary treatment approach including amitriptyline ($n = 32$). The ACT condition underwent a relatively brief treatment protocol of approximately 10 weekly sessions. Assessments were made before and immediately after treatment, as well as at 3.5 and 6.5 months follow-up. Prolonged treatment in the MDT group complicated comparisons between groups at follow-up assessments. Results showed substantial and sustained improvements for the ACT group. When follow-up assessments were included, ACT performed significantly better than MDT on perceived functional ability in relation to pain, pain intensity and to pain-related discomfort (intent-to-treat analyses). At post-treatment, significant differences in favor of the ACT condition were also seen in fear of re/injury or kinesiophobia, pain interference and in quality of life. Thus, results from the present study support previous findings and suggest the effectiveness of this ACT-oriented intervention for pediatric longstanding pain syndromes.

1. Introduction

Longstanding pediatric pain has gained increased attention in recent years. Epidemiological studies have shown frequent occurrence of this problem among youths [13], and several studies have illustrated the debilitating effects of longstanding pain syndromes for a subgroup of these children and adolescents [25,27,40]. Furthermore, a number of these adolescents enter adulthood with severely debilitating pain syndromes, entailing a substantial risk for chronicity [1,57,59]. Intervention studies have typically focused on reductions in pain and distress, while pain-related disability appears to be somewhat neglected in outcome studies on these patients [12,40,58]. Cognitive behavior therapy (CBT) for longstanding pain represents a wide variety of interventions aimed at decreasing pain and distress, and at restoring normal functions [51]. Although the empirical support for CBT on adult pain patients is strong [23,38], randomized controlled trials (RCTs) on pediatric patients disabled by longstanding pain are still scarce [12].

1.1. Acceptance and Commitment Therapy

CBT's continuing development includes Acceptance and Commitment Therapy (ACT) [22], which considers the avoidance of pain and distress a core problem leading to disability and reduced quality of life. According to ACT theory, avoidance occurs primarily when negative thoughts and emotions have excessive or inappropriate impact on behavior (denoted as cognitive fusion). In treatment, exposure to previously avoided situations is considered the core intervention. In contrast to most treatments which emphasize reduction or control of symptoms, ACT promotes acceptance of negative reactions that cannot be directly changed (thoughts, emo-

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tions, bodily sensations) in favor of engaging in activities that are meaningful although possibly painful or fear provoking. In this process, the patient learns to distance himself/herself from pain and distress in order to decrease the impact of these experiences on behavior (cognitive *de*-fusion). The treatment objective is to improve functioning by increasing psychological flexibility, defined as the ability to act effectively in accordance with personal values in the presence of interfering thoughts, emotions, and bodily sensations [20]. Repeatedly, correlational studies have shown that among people suffering from chronic pain, greater acceptance of pain is associated with, e.g., better functioning, work status, emotional well-being, and less health care and medication use [30,31]. Several studies have successfully incorporated acceptance into CBT with adult pain patients [32,34,65], as well as with people reporting pain and stress-related sick leave [6]. Furthermore, the intervention evaluated in the present study has been developed and tested on pediatric pain patients in a pilot study, indicating its usefulness for improving functioning and quality of life [66,67].

The aim of the present study was to evaluate the effectiveness of an intervention based on exposure and acceptance strategies (ACT) for children and adolescents with longstanding debilitating pain syndromes, and to compare this with a multidisciplinary treatment approach including amitriptyline (MDT). It was hypothesized that the ACT group would show statistically significant improvements on each dependent variable immediately following treatment and at follow-up. Also, it was hypothesized that ACT would generate greater improvements than MDT, particularly in pain-related functioning and quality of life.

2. Method

2.1. Setting and participants

Participants consisted of consecutive patients with longstanding idiopathic pain referred to the Pain Treatment Service (PTS) at Astrid Lindgren Children's Hospital, Karolinska University Hospital. Participants were recruited over the course of 26 months. Eligible participants were screened based on the inclusion/exclusion criteria. When asked to participate, all patients were offered the alternative treatment condition after the follow-up assessments. Two patients declined participation. A total of 32 participants were included in the study and randomized to one of the two treatment conditions. Two participants (one in each group) discontinued treatment after pre-treatment assessments were performed, and 30 participants completed treatment (Fig. 1). The study was approved by the Ethical Review Board in Stockholm, Sweden.

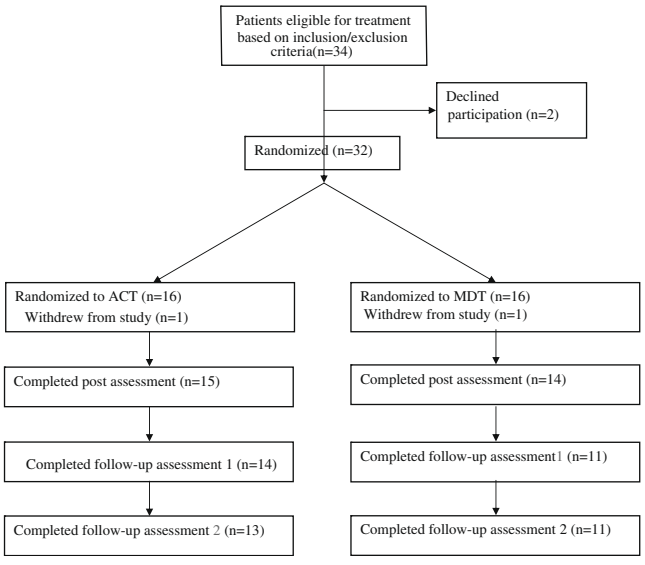

Fig. 1. A flow chart diagram of the trial.

2.1.1. Demographics and pre-treatment assessments

The sample consisted of 25 girls and seven boys, aged between 10.8 and 18.1 years (mean 14.8, sd 2.4). Time since pain onset varied between 6 and 96 months (mean 32.4 months). Among the patients included in the study, eight suffered primarily from headache. Seven participants were mainly bothered by back and/or neck pains. Six presented with widespread musculoskeletal pain, and six with complex regional pain syndrome. Visceral pain was reported by two participants, and in two other cases pain was primarily located in the lower extremities. One of the participants presented with a postherpetic type cheek pain. In 11 participants, pain onset was gradual and not associated with any significant event. Eight participants reported a minor trauma preceding the pain, and four related pain to previous surgery. For a few participants, the pain onset appeared to be associated with infections or psychological trauma. Continuous, spontaneous pain was seen in 23 of the participants. Nine reported recurrent pain. Allodynia or hyperalgesia was present in 15 participants.

As seen in Table 1, the participants in this study reported a substantial amount of pain (pain intensity: mean 5.2, sd 2.1) and distress (pain-related discomfort: mean 5.7, sd 2.2), well comparable with pediatric chronic pain samples in previous studies [7,28,41,42]. Difficulties with pain adjustment are clearly indicated by, for example, the Functional Disability Inventory (FDI) (mean 18.8, sd 12.7) and the Tampa Scale of Kinesiophobia (mean 40.2, sd 7.8), with similar ratings of disability as seen in previous studies [8,67]. Although different cut-off scores for the Center for Epidemiological Studies Depression Scale for Children (CES) have been suggested [14,39], depression scores were clearly elevated (mean 25.0, sd 10.1) as compared to a large sample of healthy adolescents [39]. However, no significant differences between the groups were seen in any of the outcome measures prior to treatment.

2.1.2. Previous investigations and treatments

When referred to the PTS, 21 patients had been seen by either a general practitioner or a pediatrician. Eleven participants had visited an emergency department at some point following pain onset. When presenting at PTS, seven had been seen by a pediatric neurologist, 14 by an orthopedic surgeon, 12 by a rheumatologist, and five by a dentist. Furthermore, nine of the participants had been seen by a psychiatrist and/or a psychologist. In general, the participants had undergone a large number of medical investigations, and MRI or CT had been conducted on 20 participants. Laboratory analyses, including rheumatic disease and titres for borrelia infection, were performed on 21 participants. Two of the participants were HLAB27 positive but otherwise tests were normal. Pharmacological treatment before referral to PTS included for all 32 participants paracetamol and/or NSAIDs. Codeine had been prescribed to six participants, of which two experienced some decrease in pain. Four participants had received tramadol, one dextropropoxiphen, and one morphine. (Ongoing pharmacological treatment at inclusion was not actively changed by the pain physician, except for the introduction of amitriptyline in the MDT.) Previous medical treatment included intraarticular steroid injections for five participants and oral steroids for another three participants. Occasionally, minor beneficial effects were obtained although most participants did not experience any improvements from these analgesics. Twenty-one participants had seen a physiotherapist. Five reported having tried acupuncture, TENS had been used by 11, and relaxation training had been performed on four participants. Eight patients had previously undergone massage, ultrasound, occlusal splints, chiropracy, naprapathy (manual manipulations with a focus on soft and

Table 1
Results for both groups on all measures. ANOVA, repeated measure, was used to evaluate each groups' improvement across time.

Dependent variables	Group	Means and standard deviations[a]				Effects of each treatment across time (ANOVA:pre,post,f-u1,f-u2)	
		Pre	Post	F-u 1	F-u 2	*F*-value (df),[b] *p*-value	Effect size[c]: η_p^2
Primary outcome variables							
FDI-child (0–60)	ACT	17.6 (13.0)	12.3 (13.9)	9.7 (13.4)	8.8 (12.9)	9.25 (1.63,45), $p = .002$	.38
	MDT	20.1 (12.7)	14.6 (11.3)	12.7 (9.7)	14.7 (12.1)	4.02 (1.32,45), $p = .049$	.21
FDI-parents (0–60)	ACT	16.5 (12.0)	8.1 (10.3)	7.9 (11.4)	8.3 (10.5)	8.72 (1.60,45), $p = .003$	.37
	MDT	23.2 (11.2)	13.9 (8.0)	12.9 (8.0)	11.8 (6.7)	16.18 (1.58,45), $p < .001$	.52
PAIRS (0–90)	ACT	50.5 (14.8)	34.5 (14.9)	31.3 (18.1)	28.5 (19.3)	13.33 (1.55,45), $p < .001$	.47
	MDT	54.6 (12.2)	51.6 (12.3)	46.7 (15.6)	44.3 (14.3)	7.32 (3,45), $p < .001$	.33
Pain interfer. (0–10)	ACT	5.7 (2.3)	3.9 (3.3)	3.4 (3.6)	3.1 (3.5)	5.82 (1.41,45), $p = .016$	.28
	MDT	6.3 (2.7)	6.0 (2.6)	4.7 (2.8)	4.9 (3.3)	6.05 (3,45), $p = .001$	.29
SF-36: phys. (0–100)	ACT	36.2 (9.4)	44.0 (11.1)	46.1 (11.2)	46.2 (11.0)	6.76 (1.43,45), $p = .010$	.31
	MDT	30.3 (9.5)	36.5 (11.9)	39.5 (9.9)	39.0 (12.4)	7.00 (1.84,45), $p = .004$	.32
SF-36: mental (0–100)	ACT	34.6 (10.8)	42.9 (12.2)	40.9 (10.1)	42.7 (9.6)	4.32 (1.79,45), $p = .027$	.22
	MDT	37.3 (14.7)	37.5 (15.0)	37.7 (17.1)	38.1 (15.3)	.03 (3,45), $p = .993$	.00
Secondary outcome variables							
TSK (17–68)	ACT	39.7 (8.4)	31.2 (6.4)	29.9 (8.7)	29.3 (8.0)	19.37 (1.43,45), $p < .001$	.56
	MDT	40.6 (7.3)	37.8 (8.9)	34.7 (8.4)	33.6 (9.0)	10.69 (1.76,45), $p = .001$	.42
CES (0–60)	ACT	24.5 (10.6)	18.4 (10.0)	16.5 (9.1)	18.1 (9.8)	3.22 (1.71,45), $p = .063$	.18
	MDT	25.6 (9.9)	25.0 (10.5)	23.2 (14.1)	25.5 (16.9)	.45 (1.35,45), $p = .568$	.03
Pain intensity (0–10)	ACT	5.3 (1.8)	3.6 (2.3)	3.1 (2.5)	3.1 (2.7)	8.17 (1.49,45), $p = .004$	.35
	MDT	5.2 (2.5)	5.0 (2.9)	4.3 (2.3)	4.5 (2.4)	1.46 (1.63,45), $p = .251$	.09
Int./cat. (5–25)	ACT	14.6 (5.8)	13.4 (3.9)	11.5 (3.7)	12.2 (4.6)	3.52 (1.71,45), $p = .051$	.19
	MDT	14.6 (4.4)	12.8 (5.5)	13.1 (6.4)	11.7 (5.8)	2.47 (1.89,45), $p = .106$	.14
Pain rel. dis. (0–10)	ACT	5.3 (2.2)	2.6 (1.8)	2.6 (2.7)	2.3 (2.5)	10.85 (1.14,45), $p = .003$	.42
	MDT	6.2 (2.2)	5.6 (2.3)	4.7 (3.1)	4.2 (3.1)	6.97 (1.66,45), $p = .006$	.32

[a] Original means and standard deviations, i.e. without adjustment for covariate (i.e. pre-treatment data).
[b] When assumption of sphericity is violated, degrees of freedom are corrected using Greenhouse–Geisser estimate. *F*- and *p*-values adjusted accordingly.
[c] $\eta_p^2 = 0.01$ (small effect), $\eta_p^2 = 0.09$ (medium effect), $\eta_p^2 = 0.25$ (large effect) (Cohen, 1988).

connective tissues [46]). In addition, four participants with CRPS had undergone sympathetic blocks without any sustainable decreases in pain. In 18 cases, the participants' mothers reported longstanding pain with four of them on disablement pension. In addition, 10 of the participants' fathers experienced longstanding pain. In 40% of the cases, the parents were divorced.

2.2. Inclusion/exclusion criteria

Patients between 10 and 18 years, referred to the PTS with pain duration of more than 3 months, were considered eligible for inclusion in the study. Patients were excluded if (a) pain was explained by an identified pathological process (e.g. arthritis, cancer, inflammatory bowel disease), (b) co-existing psychiatric or psychosocial issues were considered more relevant than pain to functioning, including risk for suicide (assessed in the psychological screening interview), (c) having a reduced proficiency in speaking Swedish, (d) suffering from major cognitive dysfunctions results in difficulties following a conversation and/or understanding the description of the study, (e) currently participating in another rehabilitation program based on cognitive behavior therapy, and (f) previously treated with amitriptyline.

2.3. Randomization procedure

A simple randomization technique was used on the 32 participants as a single block. During the 26-month recruiting period, participants meeting the criteria for inclusion were continuously randomized. A sealed envelope (prepared by a secretary blind to the objective of the study) containing a code for "exposure and acceptance" or "MDT" was opened, assigning the participant to one of the treatment conditions.

2.4. Assessment

Following randomization, all participants completed questionnaires and daily ratings during 2 weeks prior to treatment, immediately following the treatment phase (mean 5.3, sd 1.6 months after pre-treatment assessments), at follow-up 1 (mean 3.5, sd 0.8 months after post-treatment assessments), and at follow-up 2 (mean 6.8, sd 1.1 months after post-treatment assessments). All assessments were conducted by a nurse who was not involved in delivering the treatment protocol. Due to a lack of validated Swedish instrument for pediatric chronic pain, different strategies were used to create a battery of relevant measures. Some measures were included that were only validated on adult populations (e.g. TSK). Also questionnaires translated to Swedish but not psychometrically evaluated were used (e.g. FDI). Furthermore, instruments were invented by adjusting existing instruments (e.g. pain interference). Pain-related functioning (i.e. the functional disability inventory (FDI), the pain impairment relationship scale (PAIRS), and pain interference) and quality of life (i.e. SF-36, physical and mental subscales) were considered primary outcome variables. Assessments also included secondary outcome variables, e.g. kinesiophobia, and pain intensity.

2.4.1. Primary outcome variables

2.4.1.1. Functional disability inventory. The Functional Disability Inventory-child form (FDI) was designed to be applicable to a broad range of illnesses and to varying levels of severity [2,58]. Both the child and the parent forms of FDI were administered. The forms are similar, with fifteen items regarding different functional abilities (e.g. "walking upstairs", "being at school all day", "going shopping") to be rated by the participants on a 0-4 scale from "No trouble" to "Impossible". Results are expressed as total scores. The psychometric properties of the instrument have been found satisfactory. The FDI has been translated to Swedish and used to assess psychosocial impact in adolescents with headache [15]. Previous studies have shown a significant correlation between child and parent ratings, with slightly lower scores for the adults [2]. However, pre-treatment scores indicate a potential risk for floor effects [67] and the instrument does not specifically address pain-related impairments [2]. Therefore, to assess pain-related interference and disability, the FDI was supplemented by a

pain interference measure and by the PAIRS (see below). In this sample, the internal consistency (with missing values replaced with EM-method as described in 2.6), measured as Cronbach's alpha, was .93 for both the child and the parent versions of the questionnaire.

2.4.1.2. Pain interference. The Multidimensional Pain Inventory, Interference scale (MPI) [29] and the Brief Pain Inventory, pain interference items (BPI) [3] have been suggested as measures of pain-related functioning [9]. Both of these instruments are short measures with certain advantages, e.g. the inclusion of items assessing sleep. Based on these two measures, a brief inventory was assembled to assess pain interference in adolescents. The six questions closely resembled the items in MPI and BPI although age-appropriately formulated. A composite score of pain interference was calculated by averaging the six items addressing interference with schoolwork, activities outside school (leisure activities), seeing friends, mood, physical ability, sleep. The items were rated on a 100-mm VAS-scale from "not at all" to "completely". The internal consistency in this dataset was .84.

2.4.1.3. Pain and Impairment Relationship Scale. The Pain and Impairment Relationship Scale (PAIRS) was developed to assess patients' beliefs and attitudes regarding pain, or ability to function despite discomfort [44]. Psychometric evaluations of the instrument have shown adequate internal consistency [47]. Also, PAIRS reliably discriminated between pain and non-pain groups, and the instrument was significantly related to impairment even after pain intensity, duration, and severity of spine dysfunction were controlled [47]. PAIRS scores have been shown to change significantly following a CBT-oriented treatment, indicating the instrument's sensitivity to change in this type of treatment [19]. The PAIRS consists of 15 statements reflecting thoughts, attitudes and opinions about pain, such as "As long as I am in pain, I'll never be able to live as well as I did before." The degrees to which the participant agreed or disagreed with each statement was rated on a seven-point Likert scale (higher scores indicating greater tendency to associate pain with impairment and to restrict functioning in the presence of pain). In one item, an age appropriate adaptation was made by changing the wording from "work" to "school". In the present set of data, Cronbach's alpha was .81.

2.4.1.4. SF-36. The Short Form-36 Health Survey (SF-36) is a well-developed 36-item measure assessing health-related quality of life [61]. The instrument is extensively evaluated and has shown good psychometric properties [35]. The SF-36 provides summary scores for two overarching subscales: the physical component scale (PCS) and the mental component scale (MCS), with higher scores indicating better functioning. The instrument was developed for the use on subjects from age 14, and a Swedish version of SF-36 has been validated showing adequate psychometric properties [49].

2.4.2. Secondary outcome variables

2.4.2.1. Center for Epidemiological Studies Depression Scale for Children. To assess symptoms of depression, the Center for Epidemiological Studies Depression Scale for Children (CES-DC) [63] was administered. The reliability and validity of the measure has been established, especially with adolescents between 12 and 18 years [14]. The CES-DC has been translated to Swedish and shown adequate reliability [39]. In the present study, the internal consistency was .87.

2.4.2.2. Tampa Scale of Kinesiophobia. The Tampa Scale of Kinesiophobia (TSK) assesses the participants' fear of (re)injury by physical movement or activity, or kinesiophobia [50,71]. The scale consists of 17 items that are rated on a four-point scale from "strongly disagree" to "strongly agree", higher scores indicating stronger fear of (re)injury. The TSK has shown to be a reliable assessment tool for longstanding pain in several studies, especially low-back pain [5,70]. The internal consistency in this sample was .76.

2.4.2.3. Pain intensity. Pain intensity was rated once a day ("How much pain have you had today?") by the participants on a visual analogue scale (VAS) from 0 ("Not at all") to 10 ("As bad as you can imagine") during a period of 2 weeks. The mean number of ratings for both groups across all assessment periods was 14.3 (sd 1.8). The daily ratings were used to calculate each individual's mean for the assessment period, and subsequently the group mean for that period.

2.4.2.4. Internalizing/catastrophizing. The Pain Coping Questionnaire (PCQ) [43] is a self-report instrument for children and adolescents used from age eight to measure how often a particular coping strategy is used, on a scale from 1 (never) to 5 (very often). Internalizing/catastrophizing is a five-item subscale that assesses one aspect of coping with negative emotions that likely impair the use of more adaptive strategies [43]. This particular subscale has been considered relevant to this population [11]. In the present sample, Cronbach's alpha for the subscale internalizing/catastrophizing was .87.

2.4.2.5. Pain-related discomfort. Previous studies have shown that worrying about longstanding pain is more distressing, difficult to dismiss, and distracting as compared with non-pain-related worrying [10]. However, there were no measurements readily available to assess this in pediatric pain patients. Thus, to assess the extent to which the participants were thinking of, or worrying about pain and disability, the authors generated five questions: (1) How often do you worry about pain or related symptoms? (2) How often do you think about having pain or other symptoms? (3) How often are you angry or sad because of pain or related symptoms? (4) How often do you worry about not being able to do things because of pain or related symptoms? (5) How often do you worry about not being able to do things in the future because of pain or related symptoms? The questions were rated using a VAS scale from "never" to "always". A composite score (i.e. mean) was calculated based on the five questions, with higher scores indicating more discomfort. Cronbach's alpha in this dataset was .81.

2.5. Intervention

2.5.1. Exposure and acceptance (ACT)

The intervention was conducted individually and the protocol consisted of 10 weekly sessions (60 min) with the participant and 1–2 sessions with the parents (90 min). Participants in the ACT group received an average of 10.3 (sd 3.6) sessions and the parents were seen 1.7 times (sd .6) from pre- to post-treatment assessment, during a period of 4.0 months (sd 1.4). In total, i.e. including parental and follow-up sessions, the ACT group received between 7 and 20 sessions (mean 13, sd 3.5). The two psychologists involved in the intervention were trained in CBT, and both the psychologists and the physician had experience as well as formal training in ACT. To maintain treatment fidelity, treatment content and progress were discussed continuously within the clinical research group (e.g. in supervision). The protocol for the exposure and acceptance intervention resembled in all important aspects the treatment content described in detail in previous papers [65–67]. In short, the treatment protocol was as follows. Exposure to previously avoided situations and private experiences was considered the core intervention, emphasizing acceptance as an alternative to avoidance in coping with negative reactions (such as pain

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and distress) that cannot be directly changed. Initially, the dysfunctional character of longstanding pain syndromes was discussed with the participant, clarifying that pain was not caused by a potentially harmful disease or injury. Although not an ACT-intervention per se, the information was aimed at altering the context in which pain was experienced, and served to facilitate a shift in perspective from symptom reduction to valued living. It is neither particularly meaningful nor necessary to accept, or defuse from, wrong information or misunderstandings. In other words, helping the patient to understand the nature of pain syndrome sets the stage for the exposure and acceptance strategies. A thorough assessment of individual values in important life domains was performed, initiating a shift in perspective from symptom alleviation to valued living in the presence of pain and distress. This was followed by an exercise in which the workability of previous strategies to reduce pain and improve functioning was thoroughly evaluated. Since previous strategies (avoidance) had generally not reduced pain over time and still brought the patient farther from important activities, most patients experienced this exercise as emotionally challenging. However, this collaborative evaluation of previous strategies also revealed the possibility of increasing functionality and vital activities by instead accepting a certain amount of pain and distress. Based on identified values, behavioral goals were defined, followed by a discussion of gradual increase in previously avoided activities. Throughout the treatment, the participant was encouraged to notice and accept unpleasant private experiences, thus facilitating a de-fusion process (being aware of a thought without acting on its content). Discussing values, i.e. making up plans for the future, and negative thoughts about pain commonly resulted in discomfort and efforts to avoid the topic, which was addressed in therapy using exposure and acceptance strategies. Behavioral activation involving exposure to possible pain-eliciting situations other than emotional reactions was not performed in session but by the patients between sessions. Working with the parents, the shift in perspective from symptom alleviation to valued life was emphasized, as well as the principles of exposure and operant mechanisms. The parents' difficulties were addressed using the same techniques as described above (i.e. exposure, values orientation, acceptance, and de-fusion). Illustrations and metaphors were sometimes used with both patients and their parents to clarify concepts such as acceptance and de-fusion.

2.5.2. Multidisciplinary treatment and amitriptyline (MDT)

The MDT was performed by a psychiatrist, a child psychologist, a physiotherapist and by a pain physician, all experienced in working with longstanding pediatric pain. The clinical model followed the routines developed during 15 years of clinical work with this population, thus representing the usual treatment in this tertiary care setting. Within this approach, participants were seen by the different health care providers based on individual needs. A biobehavioral model of longstanding pain provided a general theoretical framework for this clinical approach, emphasizing perceived stress in everyday life as an important factor predicting the severity of longstanding pain and disability. This approach is supported in several articles and summarized in the biobehavioral model of pediatric pain [56,62]. Patients and parents were seen both individually and together by the psychiatrist and the psychologist, emphasizing the family context in discussing pain and disability. The length of sessions was 60 min. The secondary effects of pain were frequently discussed, including the relationships between longstanding pain, a lack of a medical explanation, fear of pain, muscle tension, physical and social inactivity, and school absence. Commonly, the school was contacted as part of treatment when, for example, learning difficulties were suspected. Interventions also included discussions of physical activation, relaxation, and imagery techniques. The physiotherapist sessions ranged between 40 and 60 min, and were mainly oriented towards increases in physical activities. Goal setting, graded training and pacing were frequently discussed. Physical exercises were performed at the hospital, and training programs were used as homework assignments. In some cases, TENS and warm water pool training were included. Parents were normally present during sessions, observing the training and participating in discussions regarding how to continue physical training between sessions. Patients and parents were seen by the pain physician during the 45- to 60-min sessions, which included monitoring the effects of amitriptyline (see below). Also, information about the dysfunctional character of longstanding pain was provided, clarifying that it is not caused by any harmful disease or injury, and that an increase in physical activities is an essential part of treatment. Amitriptyline doses were increased by 10 mg every week up to 50 mg, and then by 25 mg up to a maximum of 100 mg, with median max doses = 50 mg (mean 64.3, sd 27.5). The increase in doses was stopped when severe side effects appeared (e.g. sedation, dry mouth). Amitriptyline was administered during a period of 1.2 months to 19.6 months (mean 10.3, sd 5.9). Average time between pre and post assessments was 5.5 months (sd 1.9). During this period the participants in the MDT were seen for an average of 10.6 sessions (sd 4.7), equally divided between the physician, physiotherapist and the psychiatrist/psychologist. Importantly, following post assessments, participants received a substantial number of sessions (mean 11.7, sd 11.9). In addition, given that participants received amitriptyline for approximately 10 months, the pharmacological treatment also continued well beyond post assessments. At follow-up 2, the MDT group had received between 7 and 59 sessions (mean 22.8, sd 15.4) divided between the physician (mean 11.1, sd 9.1), physiotherapist (mean 3.6, sd 4.0), psychologist/psychiatrist (mean 6.7, sd 6.7), and others (mean 1.4, sd 2.1).

2.6. Statistical analyses

Two main research questions were addressed in the statistical analyses. First, did participants in the ACT-based intervention improve over time? Second, how did the effects seen in the ACT condition compare with the results from the multidisciplinary treatment including amitriptyline (MDT)? However, because the participants in the MDT condition received a substantially greater amount of treatment after post assessments, the groups were not fully comparable at follow-up. Thus, a comparison across conditions required additional analyses based on pre- and post-treatment assessments only. The results presented are based on intent-to-treat (ITT) analyses with two dropouts in the analyses. For the dropouts, pre-treatment scores were moved forward to subsequent assessments, and missing pre-treatment data were replaced with the group mean (at pre-treatment). Although one of the dropouts provided post-treatment data on one variable, the carrying forward procedure was consistently used across all variables. To ascertain that data were randomly absent, missing values were analyzed with Little's MCAR test in the Missing Values Analysis module in SPSS 15. Subsequently, empty cells were replaced by values estimated by the expectation-maximization-likelihood method (EM).

To assess comparability of the groups, analyses of variance (ANOVA) were conducted on pre-treatment data. To detect possible therapist effects, the interaction between therapist and time was analyzed with ANOVA, 2×4 mixed design (all four assessments included). Prior to running parametric tests, the data set was analyzed to detect possible violations of assumptions (i.e. normal distribution or homogeneity of variance). In analyses where the sphericity assumption was violated, degrees of freedom were adjusted using the Greenhouse–Geisser correction [16].

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The effects for each individual group across time were analyzed using ANOVA repeated measures design (including pre, post, follow-up 1, and follow-up 2 assessments). In order to maximize power with this relatively small sample, the two groups were compared using ANCOVAs with pre-treatment data used as covariates [60,64]. The first set of analyses was based on all assessments (i.e. including follow-up assessments). Additional analyses include only pre- and post-treatment assessments, due to the previously described incomparability at follow-up assessments. As a measure of effect size, partial eta-squared (η_p^2) was used. Effect sizes were evaluated as follows: $\eta_p^2 = 0.01$ (small effect), $\eta_p^2 = 0.09$ (medium effect), and $\eta_p^2 = 0.25$ (large effect) [4]. Although the level of statistical significance was set at $p < 0.05$, exact p-values are presented for each relevant test to facilitate a critical interpretation of the data [18]. Statistical analyses were performed using SPSS 15.0.

3. Results

Analyses of pre-treatment data confirmed that the two groups were comparable on all outcome measures. Results on outcome measures were based on intent-to-treat analyses. One participant in each group dropped out of treatment and withdrew from the study. Furthermore, two participants discontinued medication due to adverse side effects of amitriptyline. Analyses of potential therapist effects indicated no differences in results for the two therapists involved in the study. Table 1 summarizes the results for both groups on all measures, using ANOVA repeated measures to evaluate each group's improvement across time. In Table 2, comparisons between the ACT and MDT groups are presented (both with and without follow-up assessments). Original means and standard deviations are presented without adjustments for covariates in Tables 1 and 2.

3.1. Primary outcome variables

Significant improvements over time were seen for the ACT group in all primary outcome measures. Table 1 illustrates increases in pain-related functioning, as measured with FDI-child ($F = 9.25$, $p = .002$), FDI-parent ($F = 8.72$, $p = .003$), PAIRS ($F = 13.33$, $p < .001$), and pain interference ($F = 5.82$, $p = .016$). Both the physical scale ($F = 6.76$, $p = .010$) and the mental scale ($F = 4.32$, $p = .027$) of the SF-36 indicated substantial improvements in health-related quality of life for the ACT group. Effect sizes were large in all primary outcome measures, ranging from .22 to .47.

When comparing the groups, it was noted that also the MDT group improved during the overall course of treatment (Table 1), although not in SF-36, mental scale. As illustrated in Table 2, a significantly larger improvement in the ACT group was seen in pain impairment beliefs (PAIRS) ($F = 8.46$, $p = .007$) when comparing the groups including follow-ups 1 and 2. A difference between the groups in favor of the ACT condition could also be seen in health-related quality of life, as measured with SF-36: mental scale, although the difference was not statistically significant ($F = 3.63$, $p = .067$). No differences between the groups were seen in FDI-child ($F = .53$, $p = .47$), or in FDI-parent ($F = .40$, $p = .53$), or in SF-36: physical scale ($F = 1.54$, $p = .22$).

When confining the analyses to changes from pre- to post-treatment assessments (before extent of treatment started to diverge between groups) the ACT group performed significantly better than the MDT in pain impairment beliefs (PAIRS) ($F = 11.79$, $p = .002$), and in pain interference ($F = 5.70$, $p = .024$) as well as in SF-36: mental scale ($F = 4.99$, $p = .033$).

3.2. Secondary outcome variables

Table 1 illustrates a large decrease in kinesiophobia (TSK) ($F = 19.37$, $p < .001$) for the ACT group following treatment. Significant improvements for the ACT group were also seen in pain intensity ($F = 8.17$, $p = .004$) and in pain-related discomfort ($F = 10.85$, $p = .003$). Decreases were also seen in catastrophizing ($F = 3.52$, $p = .051$) and in depression (CES) ($F = 3.22$, $p = .063$), although not statistically significant. In general, the effect sizes were large for the ACT group's improvements in the secondary outcome variables (.18 to .56).

The MDT group improved substantially in two of the secondary outcome variables (kinesiophobia and pain-related discomfort), as seen in Table 1. As shown in Table 2, significant or nearly significant differences in favor of the ACT group were seen in kinesiophobia, as measured with TSK ($F = 4.10$, $p = .052$), pain intensity ($F = 4.25$, $p = .048$), and with pain-related discomfort ($F = 5.12$, $p = .031$) when the groups were compared including follow-up assessments. Comparisons between the groups based on post-treatment assessments showed the same pattern, although with a stronger effect. The ACT group improved significantly more in kinesiophobia (TSK) ($F = 7.66$, $p = .010$), pain intensity ($F = 4.35$, $p = .046$), and in pain-related discomfort ($F = 14.96$, $p = .001$). In depression (CES), there was a difference between the groups over time in favor of the ACT group, although slightly above the criteria for statistical significance ($F = 4.00$, $p = .055$).

4. Discussion

Results from this study suggest that an ACT-oriented treatment based on exposure and acceptance strategies can contribute to the improvement of functioning and quality of life for pediatric patients with debilitating longstanding pain. Although data indicate room for further advances, the ACT group showed substantial and sustained improvements in all measures, with mostly large effect sizes, supporting our previous findings [65,67]. Furthermore, comparisons between the two conditions (including follow-up assessments) indicated that ACT performed significantly better on perceived functional ability in relation to pain (PAIRS), fear of re/injury or kinesiophobia (TSK), pain intensity, and pain-related discomfort. Analyses conducted on post-treatment assessments (before groups diverged in extent of treatment) illustrated significant differences between the groups (in favor of the ACT condition) also in pain interference and quality of life: mental scale, with moderate to large effect sizes.

Significant improvements in the MDT condition imply that a generally useful treatment was used as a control condition. Notably, the prolonged treatment in the MDT group complicated comparisons between groups at follow-up assessments. Ideally, the number of sessions and treatment length should have been restricted to make the groups fully comparable. However, since the ordinary treatment approach at the hospital was used as the control condition, a change in clinical routines was not feasible.

Using the criteria outlined by Yates et al., the methodological quality of the present study was considered adequate [73]. However, some methodological limitations (in addition to the differences in treatment length) should be noted. First, the relatively small sample size increases the risk of judging genuine differences as non-significant (type-II error). At the same time, the number of variables being analyzed raises the concern of receiving a significant result by chance (type-I error). However, due to the lack of similar studies and uncertainty regarding expected changes following treatment, it was considered appropriate to use several different outcome measures. To facilitate a critical interpretation of the data, we have presented F-values and exact p-values for each relevant test, provided measures of effect size, and thoroughly described all statistical analyses, as suggested by Greenwald and colleagues [18]. However, it should be emphasized that more studies are needed to confirm the stability of these findings. The low n also limits the number of hypotheses that could be tested. In future

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Table 2
Comparisons between the two groups using ANCOVA with pre-treatment data as covariates. Due to discrepancies in treatment length between the groups, analyses were conducted both with and without follow-up assessments.

Dependent variables	Group	Means and standard deviations[a]				Comparison between groups including follow-up assessments[c] ANCOVA:post,f-u1,f-u2)		Comparison between groups based on post-treatment assessment[c] ANCOVA:post)	
		Pre	Post	F-u 1	F-u 2	*F*-value (df), *p*-value	Effect size[b]: η_p^2	*F*-value (df), *p*-value	Effect size[b]: η_p^2
Primary outcome variables									
FDI-child (0–60)	ACT	17.6 (13.0)	12.3 (13.9)	9.7 (13.4)	8.8 (12.9)	.53 (1,29), $p = .474$	.02	.01 (1,29), $p = .944$	.00
	MDT	20.1 (12.7)	14.6 (11.3)	12.7 (9.7)	14.7 (12.1)				
FDI-parents (0–60)	ACT	16.5 (12.0)	8.1 (10.3)	7.9 (11.4)	8.3 (10.5)	.40 (1,29), $p = .531$	.01	.96 (1,29), $p = .334$	.03
	MDT	23.2 (11.2)	13.9 (8.0)	12.9 (8.0)	11.8 (6.7)				
PAIRS (0–90)	ACT	50.5 (14.8)	34.5 (14.9)	31.3 (18.1)	28.5 (19.3)	8.46 (1,29), $p = .007$	.23	11.79 (1,29), $p = .002$	.29
	MDT	54.6 (12.2)	51.6 (12.3)	46.7 (15.6)	44.3 (14.3)				
Pain interference (0–10)	ACT	5.7 (2.3)	3.9 (3.3)	3.4 (3.6)	3.1 (3.5)	2.77 (1,29), $p = .107$	.09	5.70 (1,29), $p = .024$	.16
	MDT	6.3 (2.7)	6.0 (2.6)	4.7 (2.8)	4.9 (3.3)				
SF-36: phys. (0–100)	ACT	36.2 (9.4)	44.0 (11.1)	46.1 (11.2)	46.2 (11.0)	1.54 (1,29), $p = .224$	.05	.84 (1,29), $p = .367$	.03
	MDT	30.3 (9.5)	36.5 (11.9)	39.5 (9.9)	39.0 (12.4)				
SF-36: mental (0–100)	ACT	34.6 (10.8)	42.9 (12.2)	40.9 (10.1)	42.7 (9.6)	3.63 (1,29), $p = .067$	.11	4.99 (1,29), $p = .033$	.15
	MDT	37.3 (14.7)	37.5 (15.0)	37.7 (17.1)	38.1 (15.3)				
Secondary outcome variables									
TSK (17–68)	ACT	39.7 (8.4)	31.2 (6.4)	29.9 (8.7)	29.3 (8.0)	4.10 (1,29), $p = .052$	.12	7.66 (1,29), $p = .010$	.21
	MDT	40.6 (7.3)	37.8 (8.9)	34.7 (8.4)	33.6 (9.0)				
CES (0–60)	ACT	24.5 (10.6)	18.4 (10.0)	16.5 (9.1)	18.1 (9.8)	3.12 (1,29), $p = .088$	.10	4.00 (1,29), $p = .055$	.12
	MDT	25.6 (9.9)	25.0 (10.5)	23.2 (14.1)	25.5 (16.9)				
Pain intensity (0–10)	ACT	5.3 (1.8)	3.6 (2.3)	3.1 (2.5)	3.1 (2.7)	4.25 (1,29), $p = .048$	.13	4.35 (1,29), $p = .046$	.13
	MDT	5.2 (2.5)	5.0 (2.9)	4.3 (2.3)	4.5 (2.4)				
Int./cat. (5–25)	ACT	14.6 (5.8)	13.4 (3.9)	11.5 (3.7)	12.2 (4.6)	.01 (1,29), $p = .916$	.00	.25 (1,29), $p = .622$	.01
	MDT	14.6 (4.4)	12.8 (5.5)	13.1 (6.4)	11.7 (5.8)				
Pain rel. dis. (0–10)	ACT	5.3 (2.2)	2.6 (1.8)	2.6 (2.7)	2.3 (2.5)	5.12 (1,29), $p = .031$	.15	14.96 (1,29), $p = .001$	.34
	MDT	6.2 (2.2)	5.6 (2.3)	4.7 (3.1)	4.2 (3.1)				

[a] Original means and standard deviations, i.e. without adjustment for covariate (i.e. pre-treatment data).
[b] $\eta_p^2 = 0.01$ (small effect), $\eta_p^2 = 0.09$ (medium effect), $\eta_p^2 = 0.25$ (large effect) (Cohen, 1977).
[c] Pre-treatment data used as covariate.

studies with larger samples, mediator and moderator analyses should be conducted. The psychometric properties for some instruments used in the present study need to be further validated (e.g. for the appropriate age group, the Swedish version). For example, both the pain interference and the pain-related discomfort measures may be useful, and the validities of these questionnaires need to be further explored. A potential floor effect in FDI was noted that may suggest the need to further examine the usefulness of the instrument. The inclusion of other instruments to supplement self-report measures could also be encouraged. Also, audio/video recordings of the sessions would have facilitated formal assessment of therapist competence and protocol adherence. Differences in treatment format rather than content may in part explain the results obtained. For example, the importance of a common and clearly defined treatment objective should not be overlooked, and possibly this may have been more explicit in the ACT than in the MDT condition. Furthermore, in future trials the methodological quality can be further improved by including checks for equivalence of treatment expectations as well as methods to prevent allocation bias.

Few studies have investigated costs involved in caring for children with longstanding pain, but economic benefits from developing effective treatments for this group are clearly indicated [48]. The participants in the MDT group received, on average, twice as many sessions as the participants in the ACT group, which indicates that the results obtained should be evaluated in relation to the costs involved in delivering the intervention. However, the difference in treatment length was unintended and the data needed for such analysis could not be systematically obtained retrospectively. Tentatively, results imply that an ACT approach may be a cost-effective intervention for these patients, but it is suggested that future studies set up the data collection procedures to facilitate such analyses.

High prevalence rates and poor prognosis for many children and adolescents with longstanding pain syndromes [36,59] suggest behavioral treatment of pediatric pain as an important area of research. A relatively recent review of 18 RCTs suggested the effectiveness of psychological strategies in reducing severity of longstanding pain in children and in adolescents [12]. However, the authors noted that several studies were not carried out in clinical settings, were limited to headache, and with sometimes very short length of treatment. A number of clinical studies have recently evaluated treatments based on CBT with various types of longstanding pediatric pain conditions [7,11,28,45]. Although favorable outcomes are reported, methodological concerns (e.g. no control condition or lack of follow-up assessments) limit the validity of several of these findings.

As previously pointed out, the focus in outcome studies with pediatric pain patients has largely been on symptom reduction in the sense that pharmacological and/or non-pharmacological strategies are applied to either reduce pain directly or to assist the patient in controlling pain and distress [12]. Although such an approach displays a logical validity, considering the difficulties in achieving sustainable and clinically important reductions in pain for the most disabled patients, the workability of symptom reduction oriented approaches may be questioned. In addition, given the complex nature of longstanding pain, a potential decrease in (but not removal of) pain intensity may not in itself be enough to facilitate an increase in physical and social functioning [72]. There are to date enough studies to conclude that pain in itself does not explain disability [5,52]. The complex link between pain and disability is also illustrated in studies with pediatric patients [26]. Thus, based on the present and previous papers, it is argued that, in contrast to pain control per se, the patients' ability to act effectively in the presence of pain and distress constitutes a key factor in functioning and quality of life. This idea is supported by a growing empirical base for interventions based on exposure and acceptance [21]. Also, this perspective is reflected in conclusions drawn from a recent study that sought to establish consensus on factors contributing to longstanding pain and disability among pediatric patients [37], in which children's self-concept of being disabled, hesitation about exercise due to fear of injury, and catastrophizing were all considered predictors of pain-related disability.

Biopsychosocial models of longstanding pediatric pain have since long tried to conceptualize the relationship(s) between pain and disability [62]. The functional relationship between longstanding pain and disability can also be explained using a learning theory model [17]. Behaviors resulting in short-term relief tend to be reinforcing even if not stimulating. Although adaptive in the acute phase, such behavior pattern tends to gradually decrease functioning and life quality without a corresponding decrease in symptoms [54,72]. In an ACT-oriented treatment, following an initial behavior analysis, the target in treatment is to clarify and reduce avoidance behaviors that prevent the patient from living a vital life. Obviously, values-based exposure is difficult given the possible increase in pain and distress during this process. Based on the ACT assumption that every person is capable of living a vital life in the presence of pain and distress [20], it follows that strategies aimed at acceptance, rather than interventions to reduce pain, may facilitate exposure and behavior activation.

Although ACT has been described as a novel treatment, it has been argued that ACT is not distinct from CBT, and that it is fully compatible with the traditional CBT model of psychological functioning [24]. Similarities and differences between ACT and other CBT approaches should be further explored in component analyses. Furthermore, a recent meta-analysis indicates a need for more empirical evaluations of ACT and similar approaches, especially RCTs [74]. In addition, future studies with large enough samples and power are required to conduct mediator analyses to address issues concerning mechanisms of change in successful CBT/ACT treatments. In a recent study of this kind, it was concluded that patients' perceived pain control explained a large proportion of the treatment effect [53]. Given the recent findings on acceptance and pain [30], mediator analyses including acceptance-type variables should be performed on longstanding pediatric pain patients. A critical issue for such analyses is the availability of adequate process measures. Questionnaires to assess dimensions related to psychological flexibility, such as the Psychological Inflexibility in Pain Scale (PIPS) [69] and the Chronic Pain Acceptance Questionnaire (CPAQ) [33,68], have recently been developed and validated with adult pain patients. However, these measures are not yet validated for use with younger pain patients. Furthermore, the importance of family factors to the child's pain and disability is well known [40,55] and their roles as mediators and moderators on treatment effects need to be further investigated.

In summary, a relatively brief and low intense intervention was evaluated and compared with a multidisciplinary approach including amitriptyline. The improvements seen in the ACT condition compared to the effects from the MDT are promising. There are methodological concerns to consider when interpreting the results, and larger scale studies are warranted. However, the results from this study support previous findings indicating that interventions focusing on exposure and acceptance strategies can lead to important benefits beyond symptom control with pediatric patients suffering from debilitating longstanding pain.

Conflict of interest

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

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