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Children and Adolescents with Pain in Primary care

*Biopsychosocial determinants and behavioral
medicine treatment in a physical therapy framework*

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Abstract

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Pain during childhood and adolescents is prevalent and longstanding pain can have severe consequences for children, their families and in the long run for the society. Persisting pain influences many aspects of life and pain-related disability is often associated with impairment, decreased health-related quality of life, school functioning, participation in social life, emotional well-being, and with increased healthcare utilization. The overall aim was to explore, with cross-sectional design, pain conditions, to identify biopsychosocial determinants and their association with pain-related disability, and to study the feasibility of applying a behavioral medicine treatment for adolescents experiencing musculoskeletal pain using randomized controlled design and multimethod approach. Samples of children and adolescents and their parents seeking primary care physical therapy for a pain condition, and a sample of treating physical therapists were included.

The results showed that some children had profiles of biopsychosocial determinants that could increase the risk for long-term pain-related disability. Many had long pain duration and multiple pain locations. Girls reported higher levels of catastrophizing compared to boys, who in turn used more behavioral distraction generally regarded as a positive coping strategy.

Behavioral medicine treatment, based on a biopsychosocial approach, targeting adolescents with pain was shown to be feasible for use in primary care, with promising outcomes. Tailoring of the treatment was suboptimal but the effect of behavioral medicine treatment in pain-related disability exceeded the effect of the control treatment. The satisfaction with treatment content and results were high for both the control- and experimental condition, significantly higher for the experimental condition as rated by participants. Learning and delivering the behavioral medicine intervention was perceived challenging but rewarding by the treating physical therapists. The biopsychosocial approach in tailoring the treatment, and dialogs with parents were identified as key aspects in the behavioral medicine treatment program.

In conclusion, in children seeking primary care for pain, the factors associated with pain-related disability were complex and interrelated. The findings highlight the importance for primary care health care providers to apply in assessment and treatment the biopsychosocial approach for improved activities and participation, thereby helping children and adolescents regain health.

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To Hanna, Max and Ellen

List of Papers

This thesis is based on the following papers, referred to in the text by their Roman numerals:

- I Holm S, Ljungman G, Söderlund A. Pain in children and adolescents in primary care; chronic and recurrent pain is common. *Acta Paediatrica*. 2012; 101:1246-5.
- II Holm S, Ljungman G, Åsenlöf P, Söderlund A. How children and adolescents in primary care cope with pain and the biopsychosocial factors that correlate with pain-related disability. *Acta Paediatrica*. 2013; 102:1021-6.
- III Holm S, Ljungman G, Åsenlöf P, Linton S, Söderlund A. Treating youth in pain: A randomized controlled feasibility trial of a behavioral medicine approach in primary care. *In manuscript*
- IV Holm S, Åsenlöf P, Ljungman G, Söderlund A. Feasibility of a tailored behavioral medicine intervention for adolescents with persistent pain – the views of adolescents, parents and physical therapists. *In manuscript*

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Abbreviations

AUC	Area under the receiver operating characteristics curve
BMT	Behavioral medicine treatment
CDI	The Children's Depression Inventory
CT	Control treatment
FBA	Functional behavior analysis
FDI	Functional Disability Inventory
HRQoL	Health-related quality of life
IQR	Interquartile range
NRS	Numerical rating scale
PCQ	Pain Coping Questionnaire
PCS-C	The Pain Catastrophizing Scale-Child version
PRD	Pain-related disability
PT	Physical therapy
PTs	Physical therapists
RCT	Randomized controlled trial
TSK-C	Tampa Scale of Kinesiophobia for Children
SCT	Social cognitive theory
SEDA	Self-Efficacy for Daily Activities
SD	Standard deviation

Introduction

The clinical work in pediatric physical therapy provided me with an important insight: There were many children and adolescents with pain demanding assistance for their pain condition. I was not satisfied with the treatment outcomes, and quite a few patients returned for more physical therapy after some time had elapsed. I felt strongly that there was more to address than a child's physical problem and studying the pain literature helped me only partly. The literature on PT interventions targeting children and adolescents with pain in primary care was sparse, and could not guide me in my clinical work. My frustration and urge for more profound knowledge was the start-off point for this thesis.

The behavioral medicine perspective

Over the past decades, the understanding of the complexity of pain and the acceptance of pain as a multidimensional experience has grown significantly. A broad framework is required to understand the interplay among the wide range of factors that influence pain. A biopsychosocial model aids in understanding the ways in which pain influences an individual's life (1-5), and the interrelations among physical, emotional, cognitive and social factors are central. Several existing pain management approaches are based on the biopsychosocial model and the application of the model provides guidance in research and clinical practice.

Behavioral medicine has its underpinnings in theories and knowledge from the biomedical, psychological, behavioral, epidemiological and social sciences. Behavioral medicine represents a broad interdisciplinary approach to health and provides concrete guidance for analyzing, treating and evaluating biopsychosocial factors in pain conditions. This thesis is founded in the most relevant theories in behavioral medicine for understanding and managing pain: theories of pain and exercise physiology (6-9), learning theories (10-13) and theories in health psychology (14). All of these theories are comprehensive; therefore, only the concepts with the greatest relevance to the current research are presented.

Theories of pain physiology (6, 9, 15) provide information on nociceptive processes and on how pain is maintained or altered by peripheral inputs. The perception of pain is often not proportionate to the underlying nociceptive stimulus (9, 16), and many factors in addition to the intensity of a nociceptive stimulus influence the experience of pain. In 1965, Melzack and Wall proposed in the gate control theory, which posited that descending signals from the brain could modulate nociceptive signals at the spinal level. This provided a plausible explanation for which other signals could influence the pain process (17), such as competing signals from other peripheral receptors or psychological processes.

Theories of exercise physiology (7, 8) offer guidance and descriptions of the bodily reactions that occur, for example, when muscular strength and endurance increases. Exercise refers to structured, repetitive body movements with the purpose of maintaining and/or improving components of movement and, in turn, activity (7).

Learning theories, including respondent learning, operant learning and social cognitive theory (SCT), all concern how behaviors are learned and how they can be changed.

Respondent learning is described as learning by association, and it concerns behaviors that elicit automatic biological responses. When a neutral stimulus is followed closely in time by an unconditioned stimulus that elicits an unconditioned response, the previously neutral stimulus will elicit the same or similar unconditioned response (10). Through respondent conditioning, a previously neutral stimulus can elicit an aversive conditioned response.

According to operant learning principles, a behavior is governed by its consequences (reinforcement or punishment). Operant conditioning concerns the modification of voluntary behaviors. A behavior can be reinforced (which increases the likelihood of the behavior occurring again) or punished (which decreases the likelihood of the behavior occurring again), depending on the consequences of the behavior (11).

Another learning theory that is relevant to the behavioral medicine approach is SCT (13, 18). According to SCT, behaviors exist within a constant, ongoing, dynamic interaction between the individual and the environment, referred to as reciprocal determinism. A change in behavior is regarded as a consequence of the interplay between these factors. Self-efficacy, a key feature of SCT, is the conviction that one can successfully execute the behavior required to produce the desired outcome in a specif-

ic situation (19). Self-regulation is another important concept in SCT. This concept involves the ability of individuals to control themselves through goal setting, self-monitoring, the evaluation and adaptation of behavior performance, and the enlistment of social support. According to SCT, self-regulating skills are important for the ability to initiate and withhold behavior change, and such processes are regarded as important for long-term positive outcomes (20). SCT also describes modeling or observational learning, which refers to the capacity to learn by observing others and thereafter to enact the observed behavior (21). Modeling is believed to play a part in the phenomenon of “pain families,” in which children experience the same types of pain syndromes as their parents (22).

Within the field of health psychology, the Transactional Model of Stress and Coping (14) is important when working with patients with pain. Coping has been defined as “the individual’s response to internal or external stressors appraised as taxing or exceeding his or her resources and endangering his or her well-being” (23). The term “stressor” is used to describe an internal or external stimulus that imposes such high demands that it is appraised as exceeding an individual’s resources and affecting physical and psychological well-being (24). What is perceived as a stressor varies between situations and between individuals’ appraisals of potential harms or threats. “Primary appraisal” refers to an individual’s evaluation of the significance of a stressor. An individual’s evaluation of his or her present ability or resources to control the stressor is referred to as “secondary appraisal” (24). Appraisals are influenced by individual and situational factors and may be conscious or unconscious. Secondary appraisal is influenced by an individual’s self-efficacy beliefs concerning how to manage a stressor (13). “Coping efforts” refer to the actual strategies, thoughts or actions undertaken to manage a stressor.

A combination of these theories provides useful guidance for constructing modern treatments for childhood pain.

Pain in childhood and adolescence

According to international comparisons, Swedish children are healthy (25). There are, however, indications of a deterioration in both somatic and psychological health among Swedish school children in recent decades. Self-reported somatic and psychological problems are currently twice as common among Swedish schoolchildren as in the mid-1980s according to data from the Swedish The Swedish National Institute of

Public Health. However, the increase seems to have plateaued and there is possible a decrease for some problems (25). According to epidemiological data, pain is a common complaint, and many children and adolescents experience pain in their daily lives; approximately every third or fourth schoolchild experiences recurrent musculoskeletal pain (26, 27). The prevalence rates of pain during childhood and adolescence vary greatly and are difficult to estimate because of the lack of congruence among pain definitions and pain reporting periods in different studies. According to epidemiological studies, the prevalence of headache is reported to be 8–83%, back pain 14–24%, musculoskeletal pain 4–40%, abdominal pain 4–53% and multiple pains 4–49% (28). According to Swedish epidemiological data, 18–23% of schoolchildren report having headaches, 7–9% report back pain and 14–19% report stomachache approximately once per week (25, 29). Furthermore, approximately half of youth presenting with pain problems report pain originating from multiple locations (30, 31). The occurrence of most types of pain is reported to increase with age (27, 32). Headache, back pain and musculoskeletal pain appear to occur more frequently in older children and adolescents, whereas the relationship between age and abdominal pain and multiple pains is unclear (28).

Epidemiological research indicates that pain of most types is more prevalent in girls than in boys, although the reasons for this difference remain unclear (28, 33). Girls are more likely than boys to experience chronic pain (27, 28) and pain-related disability (34–36), and girls report higher pain intensity (27, 37). Girls tend to ascribe their pain to emotional distress to a greater extent than boys, who are more likely to attribute their pain to a physical cause (34). However, the significance of these gender differences and their possible effects on treatment outcomes remain unclear.

In the international literature, longstanding pain is referred to as chronic pain. This wording is not appropriate in Swedish, since the underlining meaning in chronic is that it is something that is difficult to cure or change. However, chronic pain is used in this paper, when referring longstanding pain. In children, as in adults, pain is commonly divided as acute or chronic (pain lasting more than three months) and as either continuous or recurrent (i.e., pain with pain-free intervals) (26, 27, 32). Recurrent pain in children can be defined as pain that occurs at least three times over a period of three months and that interferes with daily activities (5, 38). By this definition, Huguet et al found in 2009 that 37% in a sample of schoolchildren had chronic pain (39). In a recent study of a community sample, 27% of children and adolescents reported chronic

pain (40), which corresponds well to estimates of chronic pain in adults (41). In a study by Vervoort et al from 2014 a grading system for the pain severity (based on the work of Von Korff (42)) was used. Their findings indicate that a sizeable proportion of schoolchildren report moderate to severe pain, 14% of a Flemish schoolchildren reported pain in grade III or IV, moderate to severe pain (43). A high incidence of persistent pain at follow-ups after 1 to 6 years has been reported (26, 44-48), and pain during childhood appears to increase the risk of developing a chronic pain condition that continues into adulthood (49, 50).

The transition from acute pain to chronic pain is a complex process involving biological, psychological and social factors (51). Today, the most widely used definition of pain is that of the International Association for the Study of Pain: "Pain is an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (52). This definition embraces a psychological component and includes the emotional component as an inseparable part of the pain experience. Hence, pain is much more than nociception, and biological factors cannot fully explain the pain experienced by an individual. Thus, the biopsychosocial assessment and management of pain conditions in children and adolescents are important to reduce the risk that a chronic pain condition will affect an individual's adult life and quality of life.

Coping with pain

Pain coping strategies are the cognitive and behavioral responses used by patients to manage painful episodes (53). The use of coping strategies may affect pain outcomes and influence how well a child adjusts to pain-related situations (36, 54-56). Most of the knowledge derived from coping research is based on adult studies. The ways in which children cope with pain-related stressors appear to differ from the coping strategies of adults more than was previously believed. Children emotionally appraise threats and its consequences different from adults, and in turn, choose different coping strategies (57). Although research on pain coping in children has increased in recent years, extensive systematic research is still needed. In addition, the existing studies are often based on secondary and tertiary care samples. Studies involving primary care samples are lacking.

Theoretically, coping strategies can be classified in several ways and according to several aspects. One way of labeling coping strategies is to differentiate cognitive coping strategies from behavioral coping strategies. Cognitive coping strategies refer to internal strategies to handle a stressor, whereas behavioral coping strategies are more activity related (58). Coping strategies have also been viewed as either emotion-focused or problem-focused strategies (14). Emotion-focused coping is defined as efforts aiming to change an individual's attention or emotional reactions to a stressor, whereas problem-focused coping is defined as efforts aiming to change an actual behavior (23). Certain coping strategies are not necessarily better than others; the efficacy of a particular coping strategy depends on the circumstances in which it is used.

The frequent use of emotion-focused strategies, such as externalizing and catastrophizing, has been characterized as maladaptive coping and has been reported to be strongly associated with depression and disability (55, 59). Pain-related catastrophizing has been defined as “an exaggerated negative ‘mental set’ brought to bear during actual or anticipated pain experience” (60). In addition, rumination and fearful appraisals of pain are common. Catastrophizing has been reported to be a predictor of pain-related disability (PRD) in children and adolescents (54, 56, 61, 62). Studies report that pain catastrophizing contributes to PRD beyond the effects of gender, age, pain intensity, negative affectivity (63), higher levels of anxiety and depression (54). By contrast, a greater use of adaptive coping strategies, such as behavioral distraction and positive self-statements, may assist a child in managing pain and may increase the feeling of control (64, 65). Recent experimental research has revealed associations between pain-related cognitions and increased central sensitization. When subjects reduced pain catastrophizing by altering pain-related thoughts, the signs of central sensitization were reduced (66). This finding suggests that teaching patients alternative ways of thinking can have an effect on the central nervous system and can positively affect pain.

There are no studies of pain coping by children in primary care samples. However, it is possible to conclude from the above studies that coping strategies, especially catastrophizing, influence children's pain conditions. In addition to traditional PT treatment, coping skills training may be an important target for intervention in primary health care.

Pain-related fear

Fear is a distressing emotion and a response that motivates escape behavior (67), and the relationship between pain and fear elicits an inherent automatic response. The fear-avoidance model incorporates both respondent and operant learning mechanisms, and pain-related fear is likely an integral factor influencing the transition from acute to chronic pain (67, 68). A child's coping responses can influence escape/avoidance behaviors and can in turn affect the development and maintenance of chronic pain. Parental management and responses to pain (solicitous/protective) can also provide inputs to a vicious cycle of pain deterioration.

In addition to causing a child to miss potentially enjoyable social events, avoidance may lead to reductions in muscle condition and aerobic fitness, which may in turn increase the pain condition and PRD.

The fear-avoidance model predicts that the continuation of valued activities should reduce disability (67). Graded exposures to previously avoided behaviors and activities reduce pain and fear-avoidance behaviors and beliefs (69, 70).

Self-efficacy

According to SCT, self-efficacy emphasizes the likelihood of engaging in and continuing to perform a specific behavior despite aversive experiences and difficulties. In studies of adults, self-efficacy has demonstrated a solid effect on self-care activities (71). Self-efficacy beliefs in the ability to function despite pain have been reported to negatively correspond to avoidance and pain behaviors (72). There are few studies available of self-efficacy in pediatric pain, but there is no reason to believe that the processes of self-efficacy in children and adults differ. In a study by Piira et al., pain self-efficacy (i.e., the belief in one's ability to master or cope with painful experiences) in children was found to be positively correlated with age (73).

In a sample of children and adolescents with juvenile idiopathic arthritis, higher levels of self-efficacy in the ability to cope with pain was related to lower levels of pain, somatic complaints and depression. Burch et al. studied self-efficacy for functioning while in pain in a sample of children and adolescents at a tertiary pain clinic. The authors found that high lev-

els of self-efficacy for functioning was correlated with higher self-esteem and fewer somatic complaints (74).

Self-efficacy has been found to be a predictor for PRD (75) and is likely an important target for the successful treatment of pain. High levels of self-efficacy for important daily activities could enhance a child's sense of control over activities. In treatment, self-efficacy could be reinforced both by the mastery of new activity or behavior and by verbal feedback from a PT, a parent or oneself (76).

Parental behavior and children's pain

Directing attention to pain conditions in childhood is important for multiple reasons: the suffering of the individual, the risk of developing various pathological/neurological symptoms, the social and psychological effects of pain and the effects on the family. These factors are important to consider with respect to children and adolescents with pain. Chronic pain has been reported to be more common among children who have a caregiver with a chronic pain condition in some studies (77-79), but not in others (37, 80, 81). The effect of pain problems may become an economic burden for families as a result of healthcare costs and time away from work for caregivers (82). Thus, the assessment and management of pain problems in children and adolescents must also consider parents' situations.

Social learning theory (21) has often been used to describe the relationship between family factors and children's pain experience. Learning is thought to occur through parental modeling and through the parental reinforcement of pain behaviors. The way in which a child learns to cope with pain is thought to be reinforced by social and environmental consequences (83). Modeling, operant learning theories and the social reinforcement of maladaptive pain behaviors are suggested ways of explaining why pain runs in families, but the relationships are complex (84, 85), and include bidirectional interactions among child, parent and the social context (86). Parents may unconsciously reinforce behaviors that maintain or increase a child's somatic symptoms (87). Protective parental behavior and social consequences such as positive attention and activity restriction have been shown to predict increases in somatic symptoms (87), disability (86, 88), school absences (89) and child healthcare utilization (90). Minimization, which is defined as discounting and criticizing a

child's pain as excessive, has also been associated with increased somatic symptoms (91).

Parents' views, thoughts and feelings about their child's pain are important to acknowledge in the treatment of a child's pain. Adolescents suffering from chronic pain are reported to experience delays in the development of relationships, confidence and independence (92). Limitations in autonomy from parents are proposed to reduce a child's self-efficacy for daily activities (93). Helping parents to reinforce age-appropriate activities and positive health behaviors and to act as appropriate role models could assist in improving the situation (94). Changing parents' thoughts and giving them increased knowledge of how to sufficiently support their children may help improve children's functioning (95, 96).

Pain-related disability

A complex relationship among biological, psychological and environmental factors influences pain and contributes to PRD (74, 97, 98). "Disability" can be considered an umbrella term that encompasses impairments, activity limitations and participation restrictions in an individual's life (99). PRD is a complex phenomenon that reflects the interaction between an individual's body functions and features of the context in which the individual lives. In this thesis, PRD is the term used to describe the limitations and restrictions on performing age-appropriate activities of daily life that are perceived by children and adolescents as being caused by a pain condition.

It is common to experience pain during childhood, but not all pain has a substantial influence on daily functioning (32, 100). The reason that some children and adolescents are greatly affected by pain and experience substantial PRD while others (even with similar pain intensity levels) experience little or no interference in their daily lives is most likely rooted in psychosocial factors. Studies have found that psychosocial and contextual factors may contribute to PRD to an even greater extent than pain intensity (101, 102). Nonetheless, numerous studies have reported an association between high pain intensity and poor functioning in children (97, 100, 103). Depression is another factor reported to be a predictor of PRD (55, 100, 103, 104). Children's coping strategies also appear to play a role in PRD. Maladaptive coping has been reported to be a predictor of PRD

in children and adolescents (54, 56, 61, 62). There is a gap in the knowledge base regarding the factors that influence PRD in the pediatric pain primary care population because only studies from secondary and tertiary care are available.

School attendance

In adults, work performance is often used as a dependent variable in pain management studies. School attendance is often closely related to PRD and among children and adolescents with persistent pain elevated levels of school absence is common (27, 105, 106). Many children and adolescents miss school and perceive pain to interfere with academic success (106). School attendance rates are reported to be associated with parental protective response and catastrophizing and with high levels of school absence over long periods of time children and adolescents are placed at heightened risk for poor school performance and in the long run for poor occupational achievement (107).

Health-related quality of life

Health-related quality of life (HRQoL) refers to an individual's subjective perception of his or her functioning across multiple domains, including physical and psychological well-being, as well as social and school functioning. Chronic pain has a strong negative effect on the quality of life of children and adolescents (108, 109). Several studies of population-based secondary and tertiary care samples have reported that high pain intensity and frequent pain are related to lower HRQoL in children and adolescents with pain (108, 110, 111). This association has not been studied extensively in the primary care pediatric population.

Pain duration, psychosocial variables and psychological vulnerability may be additional factors affecting HRQoL in children and young people, with possibly greater effects than demographic factors (111). The associations between HRQoL and recurrent pain were examined in a large, population-based sample of schoolchildren, and the authors showed that all aspects of HRQoL were considerably impaired in children with recurrent pain, especially in children experiencing frequent pain at multiple locations (109). A recent Swedish study concluded that girls and adolescents experienced poorer HRQoL than boys and younger children. How-

ever, experiencing psychosomatic symptoms explained a substantial part of the variation (112). The gender difference is also apparent in healthcare utilization, which exhibits an uneven gender distribution according to most studies of pediatric pain. Improvements in functioning and HRQoL are often the goal of healthcare services, but no studies of HRQoL of children and adolescents seeking primary care because of pain have been published.

Physical therapy treatment

A 2006 report by the SBU (the Swedish Council on Health Technology Assessment) concluded that there was insufficient scientific evidence to determine whether PT is effective in relieving chronic pain in children (113). The report was based on only three studies: two on complex regional pain syndrome type 1 (CRPS-1) (114, 115) and one on growing pains (116), all with positive outcomes from PT treatment. Since then, only a handful of studies on the effectiveness of PT intervention have been added to the literature. For example, a recent meta-analysis of 8 studies with a total sample of 334 subjects concluded that PT treatments (with exercises and manual therapy showing the best results) appeared to be short term effective for the treatment of low back pain in children and adolescents (117). Moreover, two published studies investigated the clinical role of PT in the area of in pediatric pain, identifying PT as a core component in multidisciplinary treatment (118, 119). However, there remains an urgent need for research on PT interventions targeting pain in childhood.

Behavioral medicine treatment within a physical therapy framework

A purely biomedical approach is often insufficient to reduce or eliminate pain (2, 16), which is not surprising since pain is a multifactorial condition. There is growing research in adults with musculoskeletal pain suggesting the effectiveness of using behavioral medicine treatment (BMT) within the PT context (120-126). The SBU report from 2010 concluded that BMT leads to greater improvement in the ability to perform activities than physical activity/exercise alone, and the report recommended that BMT be incorporated into the treatment of musculoskeletal pain in adults

in primary care (127). In a study by Lee et al., a combination of PT, pain education and cognitive behavioral therapy was found to be successful for both short-term and long-term outcomes when targeting children with pain (114).

In BMT, the focus of treatment is the activity goals for each child; such treatment enhances motivation and engagement in restoring good health. When applying a behavioral medicine approach to treatment, the focus is shifted from body structures and functions and to the consequences of pain on activity and participation in everyday life. Treatment according to behavioral medicine principles strengthens patients' confidence in their own ability to function in everyday life and to independently manage pain, which has long-term benefits (121, 125, 126). However, BMT within a PT context targeting adolescents has not yet been studied. Because this approach has been successfully applied to adult patients experiencing pain, it is relevant to study whether the results from adults can be extrapolated to the pediatric population.

The rationale for this thesis

The understanding of pediatric pain in children has increased in past decades. Epidemiological studies have contributed data on pain prevalence, including gender- and age-associated differences in the general pediatric population. Insights into the specific problems experienced by children and adolescents with severe pain conditions have often been gained from studies in tertiary care settings. Therefore, the available data may not always be representative of children and adolescents at the primary care level.

A biopsychosocial model provides the theoretical framework for this thesis, which involves embracing a broad perspective on pain and PRD. To date, little is known about the target population of this thesis: what factors characterize children and adolescents seeking primary care for a pain condition? The physical and psychosocial well-being of children and adolescents seeking primary care for pain has been scarcely studied. The nature of pain, daily functioning and biopsychosocial factors related to pain in this population remain unclear. Few, if any, studies have evaluated PT interventions in this population that is suitable for use in primary care settings. There is also limited experience with BMT targeting childhood pain, indicating the need to explore the feasibility of BMT in this context.

Aims

The aims of this thesis were to explore pain conditions and the feasibility of behavioral medicine treatment in samples of children and adolescents and their parents admitted to primary care physical therapy. The aims were to describe the associations between pain, HRQoL and pain-related disability; and differences in these variables by pain location and age (Study I), to describe pain coping and biopsychosocial determinants and their association with pain-related disability (study II), and to study the feasibility and effect when applying a tailored behavioral medicine treatment targeting adolescents experiencing musculoskeletal pain (Study III and IV).

Methods

This thesis comprises four studies based on two samples of children and their parents. In Study IV, the sample was complemented by data from the three PTs who delivered the intervention. A methodological overview of the studies is presented in table 1.

Table 1. Methodological overview of the included studies.

Study	Sample	Design	Data collection method	Point of measure
I	154 children	Cross-sectional, descriptive, correlational, comparative	Self-report	At first visit to PT
II	133 [#] children	Cross-sectional, descriptive, correlational	Self-report	At first visit to PT
III	32 children 32 parents	Experimental, randomized, controlled feasibility trial	Self-report	Pre- and post-treatment
IV	25 children 25 parents 3 PTs	Descriptive, Quantitative and qualitative	Self-report and semistructured interview	Post-treatment

[#]Data from the same participants as in Study I

Settings and recruitment procedure

All the participants in the four studies were recruited when requesting PT in primary care for a pain-related problem. Primary care is the health service provided in the local community and is intended for patients who do not require the technical and medical resources of hospitals or specialist care. Many parents seek primary care as a first step in obtaining help for their child's pain problem. In most parts of Sweden, primary care PT may be accessed through either patient self-referral or physician referral,

and many children and adolescents with pain visit a PT before seeing a general practitioner or pediatrician (128).

The data from Studies I and II were collected at a primary healthcare center in a city of 200,000 inhabitants in central Sweden. The data from Studies III and IV were collected at the same center and from a primary healthcare center in a rural catchment area.

In Studies I and II, information was sent to the parents when an appointment was scheduled. Oral (child and parent) and written (parent) consent was obtained during the first visit to the primary healthcare center.

In Studies III and IV, information regarding the study was provided orally in connection with scheduling the first appointment, and the willingness to learn more about the study was explored. If permission to make another telephone call was obtained, the researcher contacted the family to provide additional information about the study. Information was emailed to the families, and written consent was obtained from both the adolescents and their parents during their first visit to the primary healthcare center.

Participants

Studies I and II

Studies I and II were based on the same sample. Patients who sought primary care PT for a pain-related condition lasting for more than one week were eligible for inclusion. At the time of inclusion, access to PT was limited by a waiting list, which caused a delay of several weeks. As a result, none of the patients included in Studies I and II had pain durations of less than two to three weeks.

Patients between 8 and 16 years of age who were admitted to PT during a 10-month period were consecutively invited to participate. Patients with poor knowledge of Swedish, experience of recent trauma or ongoing treatment for psychiatric illness or cognitive problems were excluded.

A total of 170 children and adolescents as well as their parents were approached for participation. Nine patients or parents declined participation, and seven were excluded because of poor Swedish language skills or cognitive problems. Altogether, 154 participants and their parents were included in Study I.

Study II includes data from 133 of the 154 participants; the data from 21 participants were excluded because of missing data on the pain coping questionnaire. There were no differences between the participants and non-participants or between the patients with complete data and those with missing data with regard to age, gender or pain location. Descrip-

tions of the demographic data of the participants in Studies I and II are presented in table 2.

Table 2. Descriptions of the study participants (the children and adolescents) in Studies I–IV.

	Study I/II n = 154	Study I/II n = 133	Study III/IV n = 32
Mean age (sd)	12.9 (1.9)	12.9 (1.9)	14.3 (1.2)
Gender	n (%)	n (%)	n (%)
Girls	113 (73)	96 (72)	26 (81)
Boys	41 (27)	37 (28)	6 (19)
Primary pain location(s)			
Head	38 (25)	35 (26)	11 (34)
Neck/Shoulder	9 (6)	7 (5)	2 (6)
Back/hip	22 (14)	19 (14)	7 (22)
Limb	47 (30)	38 (29)	5 (16)
Stomach	4 (3)	4 (3)	- -
Multiple Pain Locations*	34 (22)	30 (23)	7 (22)
Pain duration			
< 3 months	20 (13)	16 (12)	- -
3–6 months	26 (17)	22 (16)	4 (12)
7–12 months	34 (22)	30 (23)	4 (12)
More than one year	74 (48)	65 (49)	24 (75)

sd = standard deviation * more than three pain locations

Study III

Adolescents (age 12–16 years, school grades 6–9) who sought primary care PT for the first time for a pain-related condition of a duration exceeding three months and who reported perceiving pain-related interference in daily activities, regardless of pain location, were asked to participate in the study. Adolescents with a poor understanding of written and oral Swedish, experience of recent trauma or ongoing treatment for psychiatric illness or severe cognitive problems were not included. Forty-two eligible teenagers and their parents were approached about participation in the study; thirty-two agreed to participate. There were no differences in age, gender or pain location between the participants and non-participants. Descriptions of the demographic data of the participants in

Study III are presented in table 2. The participants' parents (28 mothers and four fathers) were also included. The participant flow, including the reasons for dropouts, is shown in figure 1.

Study IV

Study IV comprised 25 participants and their parents who completed post-treatment questionnaires in Study III. The three primary care PTs who delivered the intervention also participated in the study; they are referred to as PTs throughout the text to avoid confusion. The PTs had between 10 and 31 years of experience in the profession and a special interest in the treatment of children and adolescents but no formal training in pediatrics.

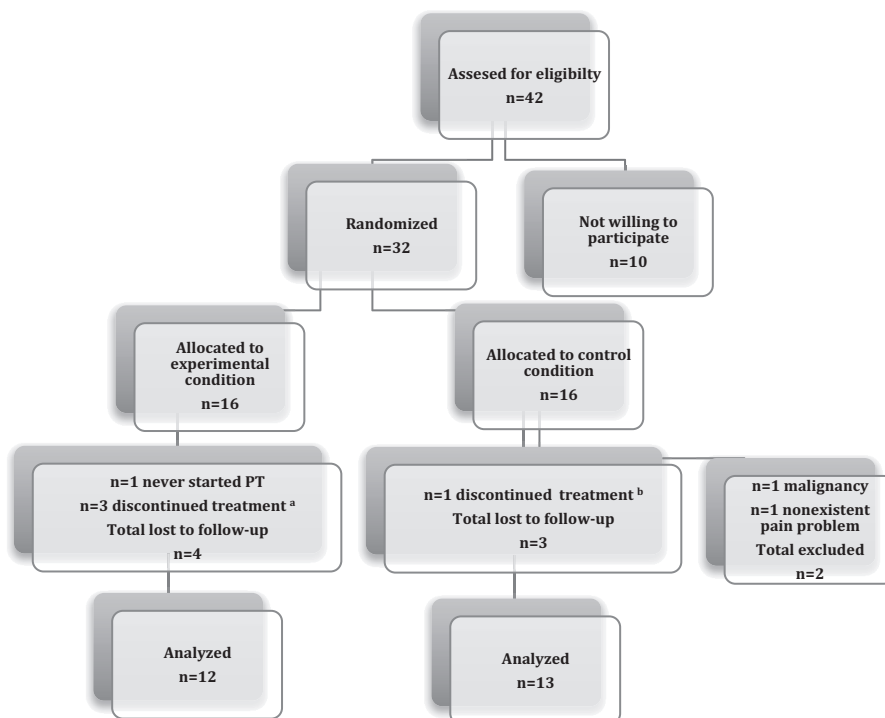


Figure 1. Participant flow in Study III ^a One participant reported a family crisis, and two participants reported difficulties prioritizing time for treatment as their reasons for withdrawing from the study. ^b One participant's mother expressed discontent with the randomization as the reason for withdrawing from the study.

Data collection procedures

Studies I and II

Data were collected during the first visit to a primary care PT. Questionnaires were presented in the waiting room, and the participants and their parents were asked to complete them separately without communicating.

Studies III and IV

Data were collected through study-specific web-based questionnaires. Before treatment, each participant and his/her parent were e-mailed a link to the study questionnaires along with an individual code; the same code was used for each participant and his/her parent. Data were collected both before and after treatment. After completing the pre-treatment questionnaires, the participants were allocated to either the BMT or control treatment (CT). The randomization results were sent to the parents, participants and PTs via e-mails or texts. For details on the randomization procedure, see Study III.

In Study IV, additional data were collected during one semistructured group interview with the three study PTs. The PTs were interviewed at one of the primary healthcare units three weeks after the feasibility RCT was completed.

Description of the interventions in Study III

A detailed manual provided instructions to the PTs concerning their work with the participants. The manual was divided into two sets of instructions: one for the experimental condition for BMT and one for CT. One workflow for each condition was described, and the manual was constructed around two or three treatment “tracks,” depending on the condition.

The exercise track: Best clinical practices for exercise (both conditions).

The lifestyle track: Informational brochures on sleep improvement, healthy eating and stress reduction (both conditions).

The behavior change track: A set of standardized psychological techniques supporting behavioral change (experimental condition only)

The tracks contained specified information on treatment techniques, such as loads and/or durations, applications of techniques and examples of homework assignments. The application of the tracks is described in detail under the description of each treatment condition in Appendices 1 and

2. For each treatment component or technique, a brief theoretical background and rationale, followed by stepwise instructions, were provided in the manual.

The use of passive, manual, acupuncture or electrophysical treatments were not allowed in either condition. The instructions allowed the number of treatment sessions to vary from 6 to 12 according to the individual needs of the participants.

Description of the experimental condition (BMT)

The BMT in this study was based on previous behavioral medicine research on adults with chronic pain (121, 122, 124). The treatment program was adjusted to be appropriate for an adolescent population regardless of pain location and was individualized according to the specific activity problems of the participants. Pain education, based on a biopsychosocial model, was introduced and discussed with both participants and parents. The participant's parents were invited to a session, without their child, on how to support their child in changing behaviors.

Workflow for the BMT

The workflow was based on learning principles (10-13) aimed at improving behavioral function and enhancing self-efficacy in performing goal activities. An overview of the workflow is presented in Table 3, for details on the BMT, see Appendix 1.

Table 3. Overview of the workflow and content of BMT.

Pain history

Anamnestic interview using a biopsychosocial approach.

Identification of problem and goal activity

Identification of individual important, pain-related problem activities.

Physical examination

Screening for red flags and a thorough physical examination of importance for the pain-related problem.

Self-monitoring

Self-monitoring of goal activity for one week.

Functional behavior analysis and identification of behavioral goals

A functional behavior analysis was formulated with information from the anamnesis, physical examination and self-monitoring.

Goal setting

SMART goal setting, for behaviors in the goal activities.

Determined together with the participant.

Treatment for behavior change

Assisted behavior change of importance for goal activity. Basic, applied and generalized skills were acquired by gradually practicing physical and psychological skills. Physical exercises as outlined in the exercise track, information according to the lifestyle track. Relevant content from the behavioral change track was used in all treatment phases.

Maintenance and relapse prevention were identified and discussed.

Homework assignments were given in all the treatment phases.

Description of the CT

Workflow for the CT

The CT workflow included physical exercises and informational brochures. This treatment was individualized according to the participants' pain-related physical and musculoskeletal problems. A brief biomedical explanation related to pain was provided to both the participants and their parents. The CT included no behavior change techniques. An overview of the structure and content of the control treatment is provided in Table 4; for more details, see Appendix 2.

Table 4. Overview of the structure and content of the control treatment.

<p><i>Pain history</i> Anamnestic interview based on a biomedical approach.</p> <p><i>Identification of problem</i> Identification of the pain problem.</p> <p><i>Physical examination</i> Screening for red flags and a thorough physical examination.</p> <p><i>Goal setting</i> General goal for treatment and goals for physical exercises. Determined together with the participant.</p> <p><i>Treatment</i> Physical exercises as outlined in the exercise track. Information according to the lifestyle track.</p>
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Measurements

A biopsychosocial model and a behavioral medicine perspective provided the foundation for the choice of measures in this thesis. These measurements were chosen to capture features of the central construct in the behavioral medicine approach.

Children and adolescents

Demographic data were collected through study-specific questionnaires, information regarding age, sex, school grade and nationality was obtained.

Table 5 presents an overview of the other measures used in Studies I–IV.

Table 5. Overview of the measures used in Studies I–IV.

Variable	Measures	No. of items	Response scale	I	II	III	IV
Pain location	Study specific questions	2	nominal scale	X	X	X	
Pain duration	Study specific questions	1	4/7-point Likert scale	X	X	X	
Pain Intensity	Numerical Rating Scale	4	0–10 rating scale	X	X	X	
Pain-related disability	Functional Disability Inventory	15	5-point Likert scale	X	X	X	
School attendance	Study specific questions	1	4/6-point Likert scale	X	X	X	
Health-related quality of life	KIDSCREEN-27	27	5-point Likert scale	X			
Pain coping	Pain Coping Questionnaire	39	5-point Likert scale		X		
Pain catastrophizing	Pain Catastrophizing Scale - Child	13	5-point Likert scale			X	
Fear of movement	Tampa Scale of Kinesiophobia - C	11	5-point Likert scale			X	
Self-efficacy	Self-Efficacy for Daily Activities	21	0–10 rating scale			X	
Depression	The Children's Depression Inventory	2	4-point Likert scale	X			
	Center for Epidemiological Studies Depression Scale	20	4-point Likert scale			X	
Treatment satisfaction	Study specific questions	3	0–10 rating scale				X

Pain location

In all studies, the reason for consulting primary care PT was recorded through an open-ended question.

In Studies I and II, the participants' main pain location was recorded using an open-ended question, and additional pain location sites (head, back, limb, neck/shoulder or stomach) were determined based on a multiple-choice question.

In Study III, the participants' main and additional pain location(s) (head, neck, shoulder, back, hip, upper extremity, lower extremity, abdomen or entire body) were determined based on multiple-choice questions.

Pain duration

Pain duration was categorized as < 3 months, 3–6 months, 6–12 months or >12 months in Studies I and II and as 3–6 months, 7–12 months, 1–2 years, 2–3 years or more than 3 years in Study III.

Pain intensity

Pain intensity was rated using a Numerical Rating Scale (NRS-11), with anchors of 0 = no pain and 10 = the worst pain imaginable. NRS-11 has a high correlation with VAS and good validity for assessing pain intensity (129, 130). Present pain was reported in all the studies, and Studies I and II assessed the minimum, maximum and average pain intensity during the previous two weeks. In study II the perceived ability to reduce pain was rated on a numerical rating scale with the anchors 0 = I have no possibility to reduce the pain at all to 10 = I can get pain free (131).

In Study III, the average pain over the previous month was assessed.

Pain-related disability

To measure PRD, the Functional Disability Inventory (FDI) was used to assess self-reported difficulties in the performance of activities in a variety of contexts. The FDI consists of 15 items describing limitations in common, daily activities as a result of pain experienced during the previous two weeks. Each item is scored on a 5-point scale (0–4) categorized by descriptors (no trouble, a little trouble, some trouble, a lot of trouble and impossible) with a total score of 0–60, with higher scores indicating greater disability. The suggested cut-off scores are 0–12 for minimal PRD, 13–29 for moderate PRD, and 30 and above for severe PRD (132). The FDI has demonstrated sound psychometric properties for children

with pain (132-134). In the PedIMMPACT recommendations (a consensus statement on measures recommended for use in clinical trials in pediatric chronic pain), the FDI was recommended for the assessment of physical functioning (135).

School attendance

Lapses in school attendance during the previous month caused by the pain problem were reported using multiple-choice categories. School absences were identified as 0, 1–10, 11–20 or > 20 h (in Studies I and II) or as none, 1 day, 2–3 days, 4–5 days, more than 5 days or every day/almost every day (in Study III).

Health-related quality of life

KIDSCREEN-27, a shorter version of KIDSCREEN-52, measures self-reported HRQoL in healthy or chronically ill children and adolescents (136). This questionnaire consists of 27 items measuring five dimensions: physical well-being, psychological well-being, autonomy and parental relationships, social support and peers, and school environment. Items are scored on a five-point Likert-type scale (1–5) and assess either the intensity (not at all, slight, moderate, high or extremely high) or the frequency (never, seldom, quite often, very often or always) of a behavior, thought or feeling based on a one-week recall time. The total score on each dimension varied between 0 and 100 (when transformed into T-values), with a higher score indicating a higher HRQoL. The data are reported as T-scores to facilitate comparisons with other samples. KIDSCREEN-27 is considered to have moderate to high levels of validity and reliability (137)

Coping with pain

The Pain Coping Questionnaire (PCQ) (138) was used to assess pain coping. The PCQ consists of 39 questions regarding different coping strategies used by a child or an adolescent experiencing pain. The PCQ consists of eight subscales: information seeking (4 items concerning learning about pain), problem solving (6 items concerning actions one can take to address pain), seeking social support (5 items concerning discussions about how one is feeling), positive self-statements (5 items related to telling oneself that the situation will improve), behavioral distraction (4 items related to participating in enjoyable activities), cognitive

distraction (5 items concerning thinking actively about things other than pain), externalizing (5 items related to screaming or throwing things to release tension) and internalizing/catastrophizing (5 items concerning exaggerated negative thoughts, such as “the pain will never go away”). Responses are rated on a five-point scale (1 = never, 2 = hardly ever, 3 = sometimes, 4 = often and 5 = very often). The subscale scores are summed, and the mean is computed. Higher scores indicate greater use of coping strategies. The PCQ has demonstrated good content validity, good construct validity and high internal consistency ($\alpha = .79-.89$) in international samples of children and adolescents (138, 139). The psychometric properties of the PCQ were reported to be satisfactory for all factors in a sample of adolescent Swedish schoolchildren, with moderate test-retest reliability (140).

Pain catastrophizing

Pain catastrophizing was assessed using a Swedish translation of the child version of the Pain Catastrophizing Scale, the PCS-C. The PCS-C consists of 13 items describing different thoughts and feelings that children may experience when in pain. Children rate to which extent they experience each thought or feeling, using a 5-point scale (0 = not at all; 4 = extremely). The PCS-C yields a total score ranging from 0 to 52 and contains three subscales: rumination, magnification and helplessness. The PCS-C has been shown to be reliable and valid in children aged 9 to 15 years (62).

Depression/sadness

In Study II, worrying and sadness/depression was examined using two questions from the Children’s Depression Inventory (141). The participants were asked to choose one of three sentences that best described their experience of the last two weeks: I sometimes, I often or I always worry/feel sadness.

In Study III, the child version of the Center for Epidemiological Studies Depression Scale (CES-DC) (142) was used for descriptive purposes. This scale has been reported to be valid and reliable for screening depressive symptoms in teenagers (143). Using a four-point Likert scale, the participants scored their symptoms during the previous week from 0 (not at all) to 3 (a great deal). A total score was calculated and ranged from 0 to 60, with a higher total score indicating more depressive symptoms.

Fear of movement

Fear of movement was assessed using the Tampa Scale of Kinesiophobia for Children (TSK-C), an 11-item questionnaire aimed at assessing the fear of movement. This instrument is an adaptation of the Tampa Scale of Kinesiophobia (144). The TSK-C consists of statements with scoring alternatives ranging from strongly disagree (1) to strongly agree (4). The total scores range from 11 to 44, with higher scores indicating a greater fear of movement. Validation and reliability testing of the TSK-C is currently ongoing at centers in the Netherlands, Canada and Sweden.

Self-efficacy

Self-efficacy in daily activities was measured using the Self-Efficacy for Daily Activities (SEDA) scale, which was developed for this study. SEDA was constructed based on the recommendations of Albert Bandura (145) and consists of 21 activities that are normally included in a Swedish high school student's day. The questions began with the stem "How confident are you in performing ... despite your pain?" The answers were rated using NRS-11, with anchors of 0 = very unconfident to 10 = very confident. A total score was calculated with a minimum of 0 and a maximum of 210, with higher scores indicating greater self-efficacy for daily activities. The validation and reliability testing of SEDA is presently ongoing.

Treatment satisfaction

Treatment satisfaction was rated using the NRS-11 scale (scores of 0–10) (135), with the anchors 0 = not satisfied at all and 10 = very satisfied. The following questions were asked: How satisfied are you with your treatment result? How satisfied are you with the treatment content? How satisfied are you with your own performance during treatment?

Parents

Studies I and II

The parents reported the demographic data concerning their child's medical conditions, family structure and school-related problems. The parents also reported whether they themselves had suffered from recurrent pain

problems during the previous year and, if so, which parent(s) was/were affected.

Study IV

The parents completed a study-specific, web-based satisfaction questionnaire in conjunction with the collection of post-treatment data. Their satisfaction with their child's treatment content and results, perceived changes in the adolescent's pain thoughts/talk and the parents' views of their own performance during treatment were explored using the same questions that were posed to the adolescents.

Physical therapists

In study IV, the PTs completed a paper-based, study-specific satisfaction questionnaire in conjunction with the participants' last session. The PTs' satisfaction with the adolescents' treatment content and results, perceived changes in the adolescents' pain thoughts/talk and the PTs' views of their own performance during treatment were explored using the same questions that were posed to the adolescents and parents.

The PTs were also interviewed about their experiences of the BMT and semi-structured interview guide with open-ended questions was used. The following three areas were covered: the perceived benefits and challenges of applying the BMT, thoughts on the education and practical training of the BMT and as the usefulness and challenges of using a treatment manual and views on its content. Follow-up questions were used when further clarification was needed.

Data management and analyses

Missing values

Generally, the amount of missing data in the studies was low. When data were missing for one or two items and were assumed to be missing at random, they were replaced with information from the remaining items in the same subscale or dimension. If more than 3 missed items were found in a measurement, the data from that individual were excluded from the analysis. For details on missing data, see each individual paper.

The analyses were performed using the Statistical Package for the Social Sciences (SPSS), versions 17.0–20.0 (Studies II-IV) (SPSS Inc., Chicago,

IL, USA). The level of statistical significance was set as a p-value = 0.05, and exact p-values are presented when appropriate. For an overview of the data analysis methods used, see table 6.

Table 6. Overview of data analysis methods in Studies I-IV.

Analyses		I	II	III	IV
Descriptive statistics	Mean (SD)	X			
	Median (IQR/min-max)		X	X	X
Inferential analyses					
Parametric	Pearson's correlation	X			
	Unpaired Student's t-test	X		X	
	One-way analysis of variance	X			
	Rasch analysis	X			
	Multiple linear regression		X		
Non-parametric	Mann-Whitney U test		X	X	X
	Spearman's correlation		X		
	Fisher's exact test			X	
	Wilcoxon's signed-rank test			X	
	Area under the receiver operating characteristics curve			X	
Internal consistency	Cronbach's alpha			X	
Qualitative analysis	Content analysis				X

Descriptive statistics

Descriptive data are presented for all the studies. Continuous variables are presented as the means and standard deviations (SD), and ordinal data are presented as the medians and interquartile ranges (IQRs) or as ranges (min-max). Nominal data are presented as absolute numbers and/or proportions (%).

Other statistical methods used for specific studies are described below.

Study I

In the analysis of HRQoL (KIDSCREEN-27), the algorithms recommended by the developers of the instrument were followed. For each

dimension, Rasch scores were computed and transformed into T-values with a mean of 50 and SD of 10 (136).

Pearson's correlation was used to assess the associations among the level of disability, HRQoL, number of pain sites, pain intensity and to assess the association between parental education level and pain duration. Student's unpaired t-tests were used for between-group comparisons of the different age groups.

A one-way analysis of variance was used for the between-group analyses of HRQoL and PRD for different pain locations. Tukey's honestly significant differences was used for post hoc analyses.

A parametric analysis was used according to the recommendations for the KIDSCREEN. Because of the nature of the data, the parametric testing was complemented by nonparametric testing where relevant, and the results were similar.

Study II

In Study II, the Mann-Whitney U test was performed to analyze the differences between age and gender on the PCQ subscale results.

Two bivariate correlations were performed using Spearman's rank-order correlation test. The first bivariate correlation was between the dependent variable PRD (measured with the FDI) and the PCQ subscales, and the second bivariate correlation was between the FDI and individual, pain-related and psychosocial factors.

Two separate multiple regression analyses were performed to assess the impact of independent variables on the FDI. In the first analysis, the PCQ subscales were regressed onto the FDI. In the second multiple regression analysis, individual and pain-related variables were regressed onto the FDI. A moderate relationship between the dependent variable and the independent variables (iv) was assumed, with α set at 0.05 and β set at 0.20. To reduce the risk of suppressor effects, a backward selection method was used. To assess multicollinearity, variance inflation factors (VIFs) were calculated. The VIFs for the two regression analyses were acceptable (range of 1.05–1.81), suggesting the absence of multicollinearity. The residuals were normally distributed, and no standardized residuals above 3 were found.

Study III

Wilcoxon's signed-rank test was used to analyze within-group changes (from pre-treatment to post-treatment) in all the outcome variables. The Mann-Whitney U test was performed to analyze differences between

treatment groups with respect to changes in their scores from pre-treatment to post-treatment.

Fisher's exact test was used for the recovery rate. Student's t-test was used to assess between-group differences in the number of treatment sessions.

In this sample, the PRD data, which were measured with the FDI, were not normally distributed. Therefore, a non-parametric option was used for the calculation of effect size. The difference between the participants' pre- and post-treatment data from the FDI was used to determine the effect size by calculating the area under the receiver operating characteristics curve (AUC). The AUC indicate the probability that a patient in one group has a better response than a randomly chosen patient in the other group. The null hypothesis was that the true area = 0.5, with higher numbers being favorable for the experimental group. When translating Cohen's standards for effect sizes (146), $AUC > 0.55$ is equivalent to a small effect, $AUC > 0.64$ is equivalent to a medium effect and $AUC > 0.71$ is equivalent to a large effect (147).

Study IV

The interview was transcribed verbatim by a research assistant, and a content analysis was performed according to the guidelines of Graneheim and Lundman (148). The interview was read several times (by SH and AS) to acquire an overall sense of the content, with a focus on the manifest content. The text was analyzed in several steps. Meaning units were identified, condensed and labeled with codes that were abstracted into subcategories and categories, focusing on the manifest content. The authors repeatedly discussed the meaning and context of the categories and the similarities and differences in the content. All the steps in the analysis process were repeated and compared to the transcribed text from the interview for content verification until full consensus was achieved.

The quantitative data were described by medians and IQRs and were displayed as boxplots. Mann-Whitney U tests were performed for between-group analyses.

Ethical approval

Ethical approval was provided by the Regional Ethics Review board of Uppsala, Sweden, for Studies I and II (Dnr 2011/120) and Studies III and IV (Dnr 2010/047). All participants and their parents were informed that they could terminate their participation in the studies at any time without providing an explanation. The written information regarding the studies that was provided contained contact details for the researchers responsible for the studies.

The intervention trial (Study III) was registered at ClinicalTrials.gov under clinical trial number NCT01381263.

Results

Studies I and II

Pain conditions in children and adolescents admitted to primary care PT

The most commonly reported pain site was limb pain, which was experienced by 47% of the participants in Study I. Limb pain was closely followed by headache (46%), back pain (40%), neck/shoulder pain (34%) and stomachache (17%). Among the participants, 58% experienced pain in two or more locations, and 22% experienced pain in three or more pain locations. Many children and adolescents had experienced pain for an extended period, according to the self-reported pain duration. In Study I, 87% of the participants had a pain duration of three months or more, and almost half of the sample had a pain duration of more than one year. Pain-free episodes (i.e., recurrent pain) were reported by 48% of the participants.

Pain-related disabilities

The levels of PRD measured using the FDI were low in this sample, with a mean FDI score of 10.4 (SD 7.6). Among the participants, 64% (n=99) showed scores indicating mild PRD (FDI scores ≤ 12), 35% (n=54) indicated moderate PRD (FDI=13–29) and the score of one participant in this primary care sample indicated severe disability (FDI=35). Moderate to severe PRD represents a substantial influence on daily living. Patients with multiple pain locations had significantly higher PRD levels (FDI scores) than those with only one pain location ($p=0.03$). No significant difference in gender was found ($p=0.108$), but the older (age 12–16 years) children reported higher PRD scores (FDI mean=11.4, SD 7.6) than the younger children (age 8–11 years) (FDI mean=8.0, SD 7.3, $p=0.01$).

Pain intensity, worrying and the ability to reduce pain explained 21% of the variance in the FDI (adjusted $R^2 = 0.21$, $p=0.03$; β values are reported in Figure 2).

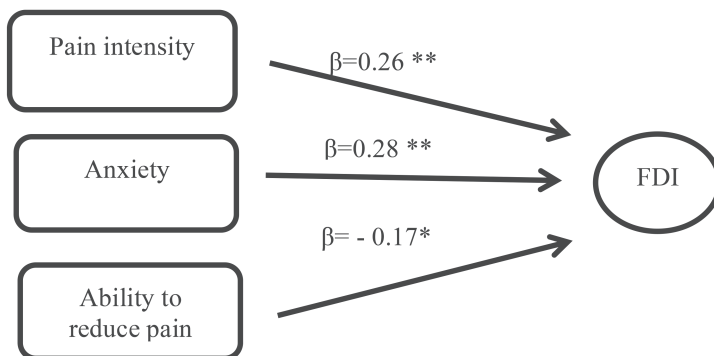


Figure 2. β - values for independent variables regressed on dependent variable Functional Disability Inventory *= $p < 0.05$, **= $p < 0.01$.

Health-Related Quality of Life

(Study I)

The reported mean score for HRQoL (KIDSCREEN) in the study sample was close to or above 50 (the mean score for an international population sample) for all dimensions, except physical well-being and psychological well-being. Descriptive data are shown in table 7. Participants with multiple pain locations reported lower HRQoL than children with a single pain location ($p= 0.01$). Differences between age groups were found for all KIDSCREEN subscales except social support and peers, for which the scores were higher for younger children, indicating a better self-reported HRQoL; see paper 1 for details. Differences in HRQoL based on gender are displayed in Figure 3.

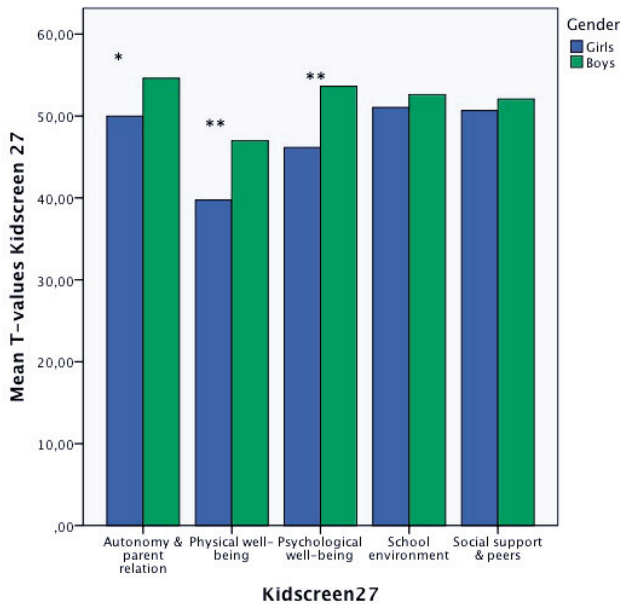


Figure 3. HRQoL, mean KIDSCREEN T-scores by gender
 * = $p < 0.05$, ** = $p < 0.01$.

Pain coping strategies

(Study II)

The self-reported coping strategies are presented in Table X. Gender differences were observed; girls scored higher than boys on the PCQ subscales seeking social support ($p < 0.01$) and internalizing/catastrophizing ($p < 0.01$). On the behavioral distraction (doing something active or fun) subscale, boys scored higher than girls ($p = 0.02$). The coping strategies of behavioral distraction, externalizing and internalizing/catastrophizing explained 13% of the variance in the FDI (adjusted $R^2 = 0.13$, $p = 0.02$; β values are reported in Figure 4).

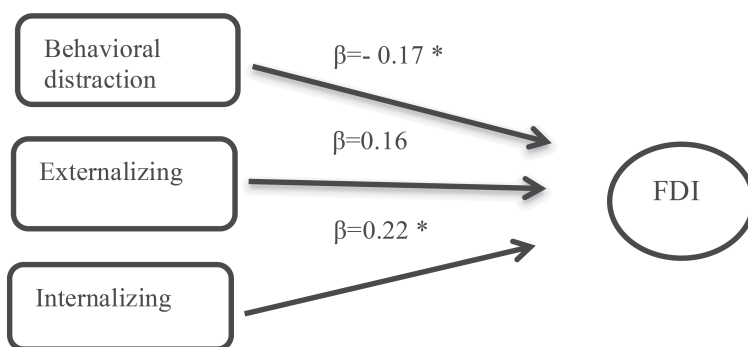


Figure 4. β - values for independent variables regressed on dependent variable Functional Disability Inventory *= $p < 0.05$.

Descriptive data on HRQoL and pain coping are provided in table 7.

Table 7. Descriptive data on HRQoL (T-values) and pain coping.

<i>KIDSCREEN dimensions</i> n=138/137	median	min/max
Physical well-being	41.8	12–64
Psychological well-being	48.3	23–74
Autonomy & parent relations	51.3	32–74
Social Support & peers	51.1	11–66
School environment	51.5	16–71
<i>PCQ subscales</i> n=133	median	min/max
Information seeking	2.2	1.0–4.3
Problem solving	3.0	1.2–4.8
Seeking social support	2.8	1.0–5.0
Positive self-statements	2.8	1.0–4.6
Behavioral distraction	3.2	1.0–5.0
Cognitive distraction	3.0	1.0–5.0
Externalizing	1.2	1.0–4.8
Internalizing/catastrophizing	2.2	1.0–4.6

KIDSCREEN T-values range from 0 to 100; a higher score indicates better HRQoL. The PCQ sum for each subscale ranges from 1 to 5, with a higher score indicating more frequent use of each strategy.

Feasibility of the tailored behavioral medicine treatment (Studies III and IV)

Treatment fidelity

The workflows of the treatments were delivered as proposed, but the number of treatment sessions varied between four and 14 rather than the outlined six to 12 sessions. A difference between the conditions regarding the mean number of treatment sessions was found, with a mean of 10 sessions for the BMT and a mean of 6 for the CT ($p=0.007$).

There were low levels of agreement between the participants' scores on catastrophizing, fear of movement and self-efficacy in daily activities and the implementation of intervention techniques addressing these factors. Among the participants with scores indicating a need for specific techniques to reduce catastrophizing or fear of movement or to increase self-efficacy, 40% ($n=6$) were introduced to these techniques; thus, the tailoring of the intervention was insufficient.

Recruitment, retention and data collection

The recruitment of participants was slower and the number of dropouts higher than estimated; 1.3 participants were recruited per month rather than the estimated 2–4 participants. The a priori estimate of treatment compliance was set at 90%, but in practice, 78% of the included participants completed treatment.

The online data collection method resulted in a low number of missing items and was thus feasible for this population. Specifically, 4% of the participants had 1–3 items with missing data and one patient had more than five items missing data on the TSK post-treatment measure.

Treatment effects

A significant decrease in PRD was found for both the BMT (score change -18, $p = 0.003$) and CT groups (score change -11, $p = 0.001$), resulting in a significant difference between the conditions ($p=0.019$). The in-between effect size for the FDI (AUC) was 0.77. Figure 5 displays the pre-and post-treatment data from the FDI. The school attendance of the participants in both BMT and CT increased, but no significant difference between the conditions was detected ($p=0.25$). Figure 6 displays the pre-and post-treatment school attendance data.

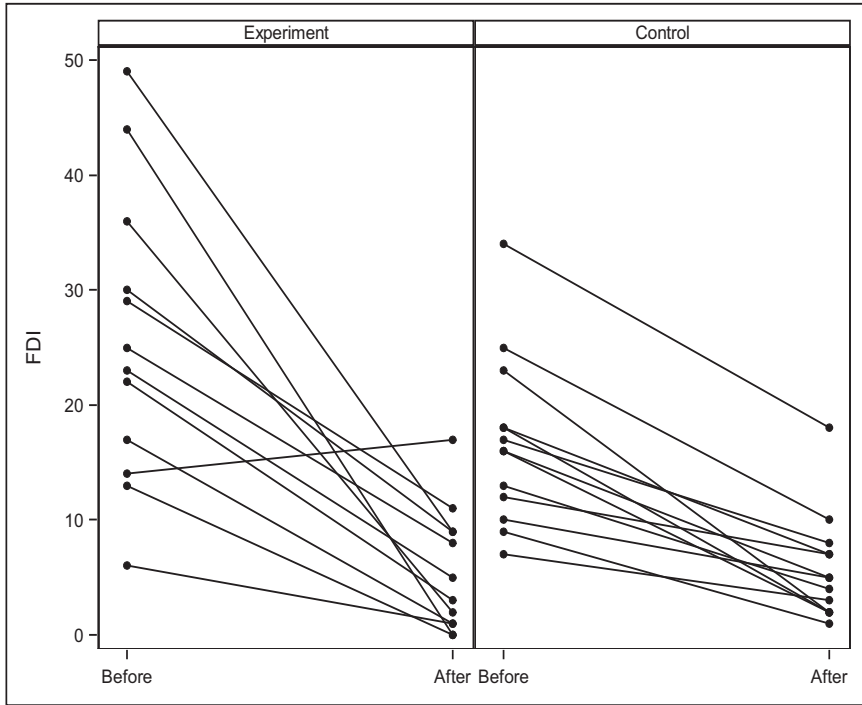


Figure 5. Pre-and post-treatment PRD data measured by the FDI for the experimental and control conditions.

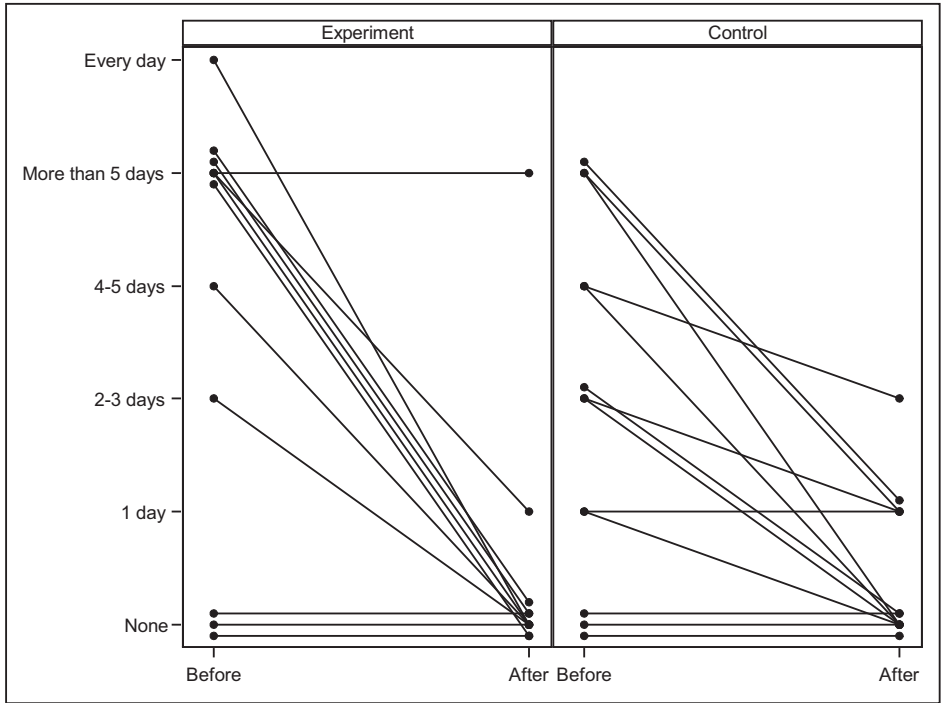


Figure 6. Pre- and post-treatment lapses in school attendance due to pain, data for the experimental and control conditions.

The participants in both conditions improved on all other measurements, and no significant between-group differences were detected. The pre- and post-treatment data for pain intensity, pain catastrophizing, fear of movement and self-efficacy for daily activities are shown in table 8.

Table 8. Pre- and post-treatment data and within- and between-group changes.

Measures	Experiment n=12			Control n=13			Between groups p
	Pre	Post	p	Pre	Post	p	
	Md (min- max)	Md (min- max)		Md (min-max)	Md (min-max)		
Intensity (0–10)	4.5 (0–8)	0.0 (0–5)	0.005	4.0 (0–10)	0.0 (0–6)	0.013	0.574
PCS-C (0–52)	25.5 (8–47)	11.5 (0–20)	0.004	20.0 (10– 49)	15 (1–39)	0.009	0.087
TSK-C (11–44)	26 (16–37)	22 (3–27)	0.047	25 (17–39)	20 (13–29)	0.030	0.820
SEDA (0–210)	127 (33–201)	200 (128–210)	0.008	151 (78–195)	191 (84–204)	0.006	0.320

Treatment satisfaction

The participants' satisfaction with the treatment results and content was high for both the control and experimental conditions, although it was significantly higher for the experimental condition ($p=0.05$). The parents were equally satisfied with both treatment conditions; no significant differences were found for any of the parents' ratings.

The PTs were satisfied with the treatment results and their own performance in both treatment conditions. However, they rated their satisfaction with the treatment content more highly for the experimental condition than for the control condition ($p=0.04$).

PTs experiences of BMT

The analysis of the group interview of the PTs regarding their BMT experiences identified three content areas: (1) learning behavioral medicine, (2) delivering the BMT and (3) the scientific context. The categories for each content area are presented in table 9.

Learning the behavioral medicine approach was perceived as demanding, and learning a new vocabulary was considered laborious. Concrete examples and supervision facilitated learning, and the theories were viewed as first becoming anchored when meeting patients.

The delivery of the behavioral medicine intervention was perceived as challenging but rewarding by the treating PTs. The biopsychosocial approach, tailoring of the treatment and dialogues with parents were identified as key aspects of the BMT. The PTs had dual experience of self-monitoring. When the self-monitoring diaries worked well; they facilitat-

ed conversation, treatment and participants' insight. However, it was challenging when a participant had entered sparse information in the diary, and the information was insufficient. With the information obtained from anamnesis, physical examination, and self-monitoring individual FBAs was formulated. The process of producing the FBAs was perceived as strenuous, and with rich information from self-monitoring diaries this formulation was less troublesome.

A detailed manual, which provided both the theoretical background and explicit treatment techniques, was considered a prerequisite for delivering BMT, but also for the ability to conduct a study. The manual also contributed to the structure of work, and explanations from the manual were used in conversations with the participants and parents.

The scientific context itself posed challenges, especially the restrictions on treatment content and session numbers.

Table 9. Overview of the content areas and categories from the analysis of the interview with the PTs

Learning behavioral medicine	Delivering the BMT	The scientific context
Learning the theoretical concepts	Holistic approach	Experiences of the control treatment
Support	Frustration	Restrictions on treatment sessions
Understanding in practice	The positive experience of working with prioritization and goal activities	The dosing of physical exercises
The need to feel confident	The dual experience of self-monitoring	The importance of the manual
The importance of supervision	The content of treatment tracks	
The treatment manual	The concept of consequences	
	Satisfaction with tailoring	
	The meaningfulness of parental work	
	The need for a manual	
	The experiences of outcomes	
	Time	
	Prerequisites in the primary healthcare unit	
	Behavioral medicine in daily practice	

Discussion

This thesis has provided new insights into pain and PRD and their associations with different aspects of the daily lives of children and adolescents seeking primary care for pain-related problems. By including the first randomized trial to implement a tailored BMT targeting adolescents with persistent pain in a PT framework, this work also provides information regarding the feasibility of such a treatment program.

Summary of results

Among the children and adolescents seeking PT in primary care in this work, some patients were slightly influenced by pain, whereas others had experienced a profound impact of pain on their lives. Many had experienced pain for a long duration of time, and multiple pain locations were common. Approximately one third of the participants reported moderate levels of PRD, indicating a substantial impact on daily living. The coping strategies reported by the children in primary care corresponded to those of tertiary care samples. The observed variation in PRD was partly explained by pain intensity, worrying, and the perceived ability to reduce pain, and by pain coping. Boys reported more behavioral distraction while girls reported more catastrophizing.

BMT for adolescents was found to be feasible when delivered by PTs in primary care, with promising outcomes. Tailoring of the treatment was suboptimal but the effect of behavioral medicine treatment in PRD exceeded the effect of the control treatment. The satisfaction with treatment content and results were high for both the control- and experimental condition, significantly higher for the experimental condition as rated by participants.

Learning and delivering the behavioral medicine intervention was perceived as challenging by the treating PTs, but was also perceived as rewarding. The biopsychosocial approach, the tailoring of treatment and dialogues with parents were identified as key beneficial aspects of the BMT program.

Pain, pain-related disability and health-related quality of life

Many of the participants in the included studies had experienced pain for long durations (48% in Study I and 66% in Study III had pain durations exceeding 12 months). We did not ask the participants in our first sample about previous contacts with the health care system for the same pain-related problem; therefore, we do not know whether the current visit to a PT was the first such consultation or had been preceded by many visits to a GP or other healthcare providers. Many individuals who suffer from pain conditions do not seek medical care at all, or they wait a long time before seeking care (149). In a Finnish birth cohort study, 16% of boys and 20% of girls reporting pain during the preceding six months had sought medical care. In a Spanish sample of healthy schoolchildren, 38% had consulted with a healthcare professional for a pain complaint during the preceding 3 months (32). In Dutch and German studies of general populations, 57% and 51%, respectively, of children with chronic pain had visited a general practitioner for their pain (34, 150). The Swedish system of direct access to PT may be one explanation for why Sweden's incidence of consultations with general practitioners for low back pain was approximately half of the consultations in England (128). Approximately 3% to 5% of children and adolescents with pain are estimated to require tertiary multiprofessional care for their pain condition (27, 32, 54). Thus, among the many individuals who experience pain during childhood, substantially fewer seek medical care in a primary care setting, and a small number seek care at tertiary care clinics.

Study I revealed a weak correlation between pain intensity and pain location; thus, reported pain intensity does not appear to be related to a specific pain location. As in many other studies from the past decade (27, 30, 31, 151, 152), multiple pain locations were common in our study. The reasons that many children and adolescents experience pain at more than one location are not fully understood. Pain at multiple sites has been suggested to have genetic origins, which is referred to as pain vulnerability (151), or to be caused by combinations of factors, such as environmental and psychological vulnerabilities or comorbidities (153). Pain at multiple sites is increasingly prevalent and is regarded as a risk factor for compromised long-term health (154, 155). In our study, participants with multiple pain locations reported lower HRQoL and higher rates of PRD than children with a single pain location. Clinicians should thus pay special attention when children in primary care report multiple pain sites.

The mean degree of PRD was low in our samples (mean FDI values of 10.4 in Study I and 19.3 in Study III) compared with tertiary care-based studies (54, 59, 86, 132). In samples of schoolchildren, lower scores have been re-

ported (mean FDI values of 4.4–8.2) (32, 61). In Study I, approximately one-third of the participants reported a degree of PRD that is sufficiently high to interfere with activities of daily living. The mean HRQoL score on the physical dimensions of KIDSCREEN confirmed that pain causes problems for youth with pain because the reported scores in our sample (for both the younger and older age groups) were below the expected scores for the age-matched general population (136). Thus, children with pain in primary care are affected by their pain and likely require treatment, but they are not likely to require highly specialized care, such as that provided by tertiary care pain clinics.

Study II examined the relative contributions of different independent variables in explaining the variance in FDI. Behavioral distraction, externalizing and catastrophizing explained 13% of the variance in PRD, and pain intensity, worrying and the perceived ability to reduce pain explained 21% of the variance. Hence, although the proportion of explained variance was significant in our sample, several unknown factors likely also contributed to PRD. Across studies of pediatric pain samples, the most salient factors predicting PRD have been pain intensity (100, 156), depression (55, 88, 100) and catastrophizing (61, 62). Other factors previously reported to partly contribute to PRD but not included in our analysis are low global self-worth (157), a deficient family environment and parental distress (97), and fear of pain (158). Our findings add to previous findings and could guide the choice of treatment content in future intervention studies in primary care, such as the benefits of addressing catastrophic or negative thinking and teaching distraction skills in combination with physical intervention components.

KIDSCREEN has been used as a generic HRQoL measure in a dozen studies targeting children with, for example, cancer, psychosomatic health complaints and environments with high migrant density (112, 159, 160). In Study I, the scores for both physical and psychological well-being were lower than those of children with cancer and the healthy control group in a study by Jervaeus et al. (160).

Powers et al. (161) compared the HRQoL scores of children with headache (a common childhood pain) to those of children experiencing chronic illness, cancer or rheumatoid conditions. The researchers found that the levels of HRQoL did not differ according to diagnosis, but there were some differences within subscales. Thus, children seeking primary care for a pain condition may have HRQoL scores that are similar to those of children with serious diseases or illness.

Psychosocial factors and pain

Children's choice of strategies for attempting to cope with pain are associated with their success in adjusting to pain and the effects of pain on their lives (37, 54, 56). We found that children in primary care exhibited similar patterns of coping strategies, but they appeared to report less frequent use of such strategies than those in tertiary care. One possible conclusion is that children in primary care have not yet fully developed their coping strategies. Thus, it is important to assess the pain coping strategies of children receiving treatment in primary care.

PRD was partly explained by catastrophizing and externalization, both of which are considered maladaptive coping strategies in both adult and child studies across pain conditions (56, 60, 111). Pain catastrophizing has been found to be associated with lower pain tolerance (73) and greater emotional distress (54), and it contributed to increased pain 6 months later in a prospective study by Vervoort et al. (63). Behavioral distraction (which also partly explained PRD in Study II) is generally regarded as a positive coping strategy (64, 73, 162) and is associated with greater perceived pain control (163). Thus, treatment should aim to decrease catastrophizing and to increase the use of behavioral distraction techniques.

Differences between genders were found. Compared with boys, girls reported a higher incidence of catastrophizing and were more likely to seek social support, whereas boys reported greater use of behavioral distraction. These gender differences in catastrophizing and distraction are consistent with the results of studies of chronic pain samples (35, 36) but could not be confirmed in a community-based sample (163). The greater inclination of girls (compared with boys) to seek social support (i.e., to engage in discussions with friends and family about their pain) has been confirmed in both chronic pain and community samples (35, 36, 163). Clinicians should be aware of such gender differences because they may be relevant to treatment.

Many of the participants in our studies reported symptoms of sadness or depression. Depression has repeatedly been found to be a predictor of PRD in children with pain (55, 88, 100), and it could also be an important factor to consider at the primary care level. Comorbidity between psychiatric disorders such as depression and pain is also common (164-167). Future studies, should evaluate whether decreases in pain and PRD also change depression scores or whether changes in depression scores could mediate treatment effects. The healthcare staff at primary care units should be aware of the possible comorbidity between pain and depression, include an assessment of

depression in the anamnestic interview and refer patients to the appropriate care if depression is suspected.

In our studies, we found no significant correlation between parents' own pain and their children's PRD. Mixed findings have been reported regarding the relationship between parental pain and children's pain. Pain has been reported to be more common among children who have a caregiver with a chronic pain condition in some studies (77-79) but not in others (37, 80, 81). The social environment is regarded as important for a child's pain perception (54), and the influence of the social context is believed to be greater for children than for adults (168). According to SCT, learning is thought to occur through both parental modeling and parental reinforcement (83). Therefore, an assessment of how parents respond to their children pain may be important to fully understand the factors contributing to a child's PRD (84, 86, 169).

The social dimensions of KIDSCREEN revealed high scores, which may indicate good relationships between the studied children and their parents as well as age-appropriate autonomy from parents (136). We did not study parental catastrophizing in our first sample. However, the children and adolescents reported relatively high levels of autonomy, which could indicate that there is no need for general parental intervention in primary care. However, PTs adopting a biopsychosocial treatment approach should be encouraged to assess parents' thoughts and beliefs regarding their children's pain because protective parental behaviors (88, 91) and parents' catastrophizing (86, 170) appear to be associated with PRD in children.

Guidance from learning theories and the correlations between parental behaviors and PRD found in previous studies led us hypothesize that targeting parents' beliefs regarding their children's pain could be valuable. Asmundsen and colleagues suggested that treatments aiming to reduce PRD among children with pain conditions should also address parental psychological responses and behaviors (67). When parents who report catastrophizing thoughts (about their child's pain) are encountered, the provision of advice and/or a referral to a psychologist may be important to increase the success of BMT for the child. In Study IV, working with parents was identified as one of the key aspects of BMT by the treating PTs. Many pain treatment programs have reported including parents in children's treatment (96, 171-173), but few have reported specific effects or explored parents' experiences of such treatments. Future studies could address if teaching parents to systematically use operant techniques for reinforcement of new, positive health behaviors in their child is effective.

The behavioral medicine treatment for adolescents with pain

In Study III, 75% of the participants receiving BMT and 62% of those receiving CT considered themselves fully recovered from their pain problem after treatment. A significant difference between the treatment conditions (in favor of BMT) was found in the scores for the primary outcome variable FDI (an indicator of PRD). All but one of the participants with pretreatment school absences had increased school attendance post-treatment (the other primary outcome variable), but no difference was found between conditions. Measuring treatment satisfaction is important because a patient's experience is associated with treatment adherence and effectiveness. In a review of 55 studies, a consistently positive association between patients' treatment satisfaction and health outcomes was found across diseases and settings (174). Satisfaction with treatment was high for both conditions, but the participants in the experimental condition assigned higher scores to their satisfaction compared with those in the control condition. Parents were also highly satisfied with both treatment conditions.

BMT appears to be a promising method for addressing both the physical and psychological aspects of pain in adolescents receiving PT in primary care. However, many improvements in the implementation of BMT could be applied in future studies.

During the planning for the BMT in Study III, theories of pain and exercise physiology (6-9), learning theories (10-13) and theories of health psychology (14) emerged as applicable to comprehensive treatment. Thus, the theoretical basis for the BMT was extensive. The PTs' brief education in the theoretical underpinnings of the behavioral medicine approach was likely insufficient, as partly indicated by the results of Study IV. The PTs perceived the theoretical education in behavioral medicine as well as the theoretical content and new vocabulary as challenging. Nonetheless, the PTs concluded that the amount of theory in their education was adequate. However, therapists' theoretical understanding of the components included in a treatment program is critical to their adherence to the treatment protocol (175).

The BMT could have been structured differently and likely would have benefited from being less comprehensive; the treatment comprised many components and treatment techniques. Individual tailoring is a key component of BMT. In fact, individually tailored BMT has previously been reported to be useful for adults receiving treatment for musculoskeletal pain (121, 122) and whiplash-associated disorders (123) as well as for enhanced physical activity and healthy eating (176). The individual tailoring of treatment based on individual assessment is intended to reach an individual based on his or her personal characteristics (177). In our study, the tailoring of the treatment was

based on the individual FBAs of the participants. However, the tailoring in our study did not fully function as intended. There was a suboptimal level of agreement among the scores on measures of pain-related fear, catastrophizing and self-efficacy for daily activities and who had been introduced to the specific behavioral change techniques targeting these constructs.

Pain-related fear has been found to be associated with higher levels of PRD and more frequent use of healthcare (178), and has also been found to be a stronger predictor of physical activity limitations compared with pain intensity or depression (169). Graded exposure combined with the use of self-reinforcement strategies to increase activities was promoted in the treatment manual, but these techniques were not introduced to all the participants with high scores on TSK-C. The fear-avoidance model (68) has been amended several times, but it was only recently first conceptualized within a pediatric pain framework (67). The fear-avoidance model could serve as an explanation of the processes and mechanisms through which pain becomes chronic; therefore, fear-avoidance could be an important target of interventions for persistent pain in children/adolescents (67).

Not all participants in the BMT condition who had high scores on the PCS-C were introduced to techniques for handling catastrophizing or negative thoughts. Catastrophizing is associated with increased pain and PRD (62). Cognitive restructuring (179), challenging negative thoughts (173), positive self-talk (171) and problem-solving (180) have been used to address catastrophizing, frequently as part of a more comprehensive cognitive behavioral treatment program aiming to reduce PRD. The participants in both treatment conditions decreased their PCS-C scores post-treatment, and the reductions in the scores were more substantial for the participants in the BMT (the mean change in the score was -17.5 for the BMT group and -7 for the CT group). However, the differences were not sufficiently large to reach significance.

In adults, the goal of enhancing self-efficacy has been successfully incorporated into tailored BMT (121) and cognitive behavioral pain management programs, with corresponding increases in exercise levels, pacing and relaxation training (181, 182). In Study III, self-efficacy for daily activities was significantly increased in BMT and CT groups after the treatment. It is possibly that the increase in self-efficacy, since it happened in both groups, was generalized by the perceived success from the physical exercises within the exercise track. The experience of success increases self-efficacy according to SCT (19, 76). However, the participants in the BMT group could have

been exposed to several other techniques for increasing self-efficacy that were included in the treatment manual, such as modeling, which could have produced additional increases in self-efficacy and possibly entailing differences between the treatment groups.

In conclusion, the information regarding the levels of pain-related fears, catastrophizing and self-efficacy for daily activities provided during the anamnestic interview was insufficient for the PTs to fully tailor the treatment. One can only speculate whether more precise tailoring would have resulted in different outcomes. In future studies, tailoring components could be provided by PTs who are not blind to the participants' scores on measures of pain-related fear, catastrophizing and self-efficacy for daily activities or by an independent person providing the PTs with risk profiles for the participants. Such approaches would require further validation and meaningful cut-off scores for the applied measurements, and such scores do not currently exist. Another option would be the use of specific anamnestic assessments at the clinic to promote high levels of accuracy in identifying psychosocial problems. Also, the PTs would probably need to be provided with specific guidance regarding the appropriate treatment to be used, i.e. assisted in translating findings into treatment.

We currently have no information regarding the long-term effects of the BMT; however, following the participants over time will provide these data. Applying a behavioral medicine approach involves the enhancement of self-management skills to address pain during treatment and the formation of plans for maintenance and relapse prevention. An increased ability to self-manage pain has been reported to be associated with improvements in function and psychological well-being (183). The effects of an intervention providing self-management techniques may certainly continue after the treatment is completed and after participants have presumably learned skills that may prove valuable if they encounter similar or new pain-related problems in the future. Further studies are needed to confirm these associations.

Methodological considerations

The use of valid measures that mirror biopsychosocial determinants is critical for measuring BMT effects and for understanding the broad impact of pain. FDI is the most frequently recommended and used measure of pediatric PRD. In Study III, only participants who exhibited substantial PRD were recruited; however, 5 of the 25 participants received low scores on the FDI. Thus, a potential floor effect was found for the FDI. A study of adolescents with idiopathic pain also found floor effects for the FDI (70). The FDI was

originally designed to assess limitations in daily life among children with abdominal pain rather than to specifically measure PRD in children and adolescents with persistent musculoskeletal pain. Further psychometric testing and possible revision of the FDI to better capture PRD in children in primary care is advisable. The use of measures specifically designed to assess mild pain-related interference in daily activities or a patient-specific disability measure could complement the FDI in future studies. There are several other options for measuring PRD, including The Child Activity Limitations Interview (a specific assessment of PRD) (184), the physical dimensions of the Bath Adolescent Pain Questionnaire (a comprehensive assessment of the effects of chronic pain across physical and psychosocial constructs) (185) and the PROMIS Pediatric Pain Interference Scale (which assesses pediatric pain interference with daily functioning) (186). These options are relatively new; when our project began, none of them were available in Swedish, they had not been validated for the pediatric primary care population, and they were not primarily intended for use in patients with musculoskeletal pain. However, these measures are promising in many ways, and research is needed to validate them for use in Scandinavia and in the pediatric primary care context.

Some of the other measures used in this work are also worthy of further reflection. The measures used in Studies I and II largely followed the recommendations of the PedIMPACT consensus statement (135). However, KIDSCREEN, the instrument that we chose for measuring HRQoL, was not among the recommended measures for HRQoL. We chose to use KIDSCREEN because it was an age-appropriate measure with a Swedish translation, because a short-form version was available and because the subscales measured constructs that were relevant to this project.

Pain intensity ratings are almost always used in clinical studies, although it is a great simplification to assess a complex phenomenon this way (129, 130). The most commonly used self-report measure in adults is the NRS-11, which has demonstrated sound psychometric qualities for use in assessing pain intensity in pediatric populations (129, 130, 187). However, the studies on children and adolescents recall time for reporting pain intensity is sparse. Pain intensity scores have been demonstrated to be stable over a 2-week period (187), but retrospective scores have been reported to show inflated values (188, 189). In our studies the participants reported both present pain and retrospective pain intensity. We chose to use present pain in our analysis, with the purpose to avoid recall bias. But, since pain intensity often is fluctuating to its nature, by this, we might not have captured the whole picture. The use of pain diaries during one or two weeks before starting treat-

ment (and after for effect), preferable with electronic devices, could be an option to acquire accurate data (190, 191).

In Study IV treatment satisfaction was measured by study specific questions. There is a recent interest in measuring treatment satisfaction from the children's viewpoint, but the established measures are few (135, 192, 193) and none specifically designed for use in our sample of interest. However, open-ended questions or interviews pertaining children's and parent's reasons for global ratings could have been used to provide more comprehensive data concerning treatment satisfaction.

Both the PCS-C and TSK-C are pediatric measures extrapolated from the adult versions. The validity and reliability of the TSK-C have not yet been studied, whereas the reliability of the PCS-C was confirmed in a study of Flemish-speaking children (62). In a recently published study, an 11-item English version of the PCS-C was found to provide a statistically better fit than the presently existing 13-item measure. (194). Thus, there is a need for further testing and the establishment of cut-off values for the PCS-C and TSK-C.

No difference between the treatment conditions was identified on SEDA in the post-treatment data. The SEDA instrument has not yet been validated in a normal population or in clinical samples. A question remains concerning whether a measure of specific goal activity related self-efficacy would have generated more reliable results and between-group differences than the SEDA, which was developed to be used in this study. Another possible problem when assessing self-efficacy is whether the targeted age group really can differentiate between the *belief* in one's capabilities in performing an activity and the actual performing of the activity. This remains to be resolved in future studies.

There is an increased risk of type II error when drawing conclusions from studies with small samples. In Study III, both treatment conditions led to significant improvements in fear of movement, catastrophizing and self-efficacy for daily activities, which were measured with the TSK-C, PCS-C and SEDA, respectively. A larger sample size might have detected between-group differences in these measures. When an active control group is used, as in our Study III, differences may be too small to detect in a small sample. Unfortunately, there are no data available to help determine the cut-off values for clinically relevant changes; nevertheless, we observed that both groups exhibited prominent changes in scores.

A limitation of the first two studies was that the results were based on cross-sectional data, which makes it impossible to determine the direction of the relationship between variables. Prospective longitudinal designs with repeated measures could have revealed more information regarding the development of PRD and the factors influencing PRD.

One limitation of Study III is that it was originally intended to be a full-scale RCT rather than a feasibility study. A study that was specifically designed to evaluate the feasibility of testing this rather complex intervention could have provided more detailed results. However, the findings from the two feasibility studies may be applied to guide future research in this context and to potentially guide the clinical implementation of BMT in the primary care setting. For future BMT studies our findings imply that the knowledge, skills and competencies of PTs are requirements for delivering BMT, and treatment fidelity must be monitored. Supervision, including comprehensive discussions, patient cases and educational outreach visits with direct feedback in clinical situations, may be useful strategies to facilitate the learning of a new treatment approach.

Another limitation is that the same PTs delivered both treatment conditions, which may have posed a threat to the internal validity through contamination. However, during the treatment, consideration and actions were taken to avoid contamination between the treatment conditions. During each supervision session, efforts were made to distinguish between the two conditions by discussing them and ensuring that the correct workflow was used for each participant. Although it would have been advantageous for different PTs to have delivered the BMT and CT, such an approach was not possible for practical reasons.

In Study III, the randomization procedure did not function fully as intended because the pretreatment FDI data were unevenly distributed between the groups in the relatively small sample. However, there is no reason to believe that the outcome of this randomization procedure would have been unreliable in a full-scale study. One reason to perform a feasibility or pilot study is to estimate an effect size to guide the calculation of sample size for a full-scale study. The small sample size and the uneven distribution of FDI scores between the groups led us to calculate the effect size for treatment outcomes using a non-parametric method: the area under the receiver operating characteristics curve (AUC). The AUC represents the probability that a patient in one group will have a response that is preferable to that of a randomly chosen patient in the other group. The null hypothesis is that the true area = 0.5,

with higher numbers favoring the experimental group. When translating Cohen's standards for effect sizes (146), an AUC of 0.55 is equivalent to a small effect, an AUC of 0.64 is equivalent to a moderate effect, and an AUC of 0.71 is equivalent to a large effect (147). The AUC found in Study III was 0.77, but the large effect size should be interpreted with caution because the groups were not exposed to the same number of treatment sessions. If this effect size is used for sample size calculation, a conservative approach is advisable. Our results also indicate that high withdrawal rates should be considered when planning a study within this field of interest.

External validity and transferability

The results from the first two studies are valid for children and adolescents who seek PT for a pain condition and who are willing to participate in studies within settings in countries with similar social contexts and healthcare systems. The majority of the participants were female. This uneven gender distribution was consistent with previously reported prevalence rates (27, 28); therefore, there is no reason to believe that our sample differed from the patients typically presenting in clinical settings. In Study I, a small number of randomly distributed items had missing data. In Study II, only participants with complete PCQ data were included; the excluded group did not differ from the included group with regard to age, gender or pain location.

The generalizability of the results of Study III is low because of the small sample size. However, the purpose of conducting a feasibility or pilot study is not to produce generalizable results but to test the study concept and intervention. Larger studies are warranted, preferably with participants recruited from a variety of primary care settings. The transferability of the findings from Study IV is difficult to estimate. The variation among PTs was small, with only three PTs from two primary care centers. It is likely that some aspects of the experience of a PT learning and delivering BMT in a primary care setting were not discussed and included in the data analysis. Future studies are needed, preferably with larger numbers of PTs and the inclusion of specific research questions concerning the implementation processes.

Conclusions

The children and adolescents seeking primary care PT for a pain-related problem shared many of the common features of children admitted to tertiary pain clinics. In this work, the results indicate that pain has an impact on HRQoL in children and adolescents admitted to primary care PT. Physiological and physical factors partly determine pain and PRD, but psychological aspects, such as coping also, affect PRD. Thus, in children seeking primary care for pain, the factors leading to PRD are complex and interrelated. BMT targeting adolescents with pain is feasible for use in primary care with the exception of the tailoring components, which require further development. BMT poses great challenges on the PTs. Substantial theoretical knowledge and understanding of theory and supervision during long periods of time is likely integral for successful implementation of BMT.

Likely, when patients, parents, and PTs have a broad understanding of biopsychosocial factors influencing pain, improvement of activities and participation may be facilitated, thereby helping children and adolescents to take an important step toward regaining good health.

Clinical implications for primary care pain treatment of children and adolescents

- Primary care providers should, along with the physical examination also assess patients for psychosocial prognostic factors for long-term PRD at the first visit and tailor treatment guided by the identified prognostic factors.
- Children with pain at multiple sites should be regarded as high-risk patients.
- Special attention should be given to girls with pain since their greater tendency to catastrophize places them at higher risk for PRD.
- The use of measures of patients' psychosocial risk profiles is likely needed for adequate tailoring of treatment.
- Self-monitoring can help increase awareness what influences pain but it should be brief and simple to increase the children's adherence.
- A combination of physical exercise and psychological behavioral change techniques is promising treatment. By providing behavioral medicine training for PTs, behavioral medicine treatment can be tested and implemented in clinical work.

Future research

The results of this work prompt several suggestions for future studies. The following suggestions concern some of the essential research questions that should be addressed in the future.

- A full scale RCT should be performed considering the insights from the feasibility studies.
- Validation and reliability testing of measures should be performed, including cut-off scores for measures used to determine BMT treatment outcomes.
- The most important components for providing a sufficient but less comprehensive BMT should be determined.
- The use of e-health and modern technology for self-monitoring and delivering of treatment should be examined in the context of BMT.
- Interventions for parents should be studied, particularly to determine the specific effects of teaching parents to systematically use operant techniques.

Svensk sammanfattning (Swedish summary)

Smärta är vanlig förekommande under uppväxtåren. Tidigare studier har visat att långvarig smärta kan ge allvarliga konsekvenser för barn och deras familjer. Smärta påverkar många olika delar av livet och smärtrelaterad aktivitetsnedsättning har visat sig ha en negativ påverkan på hälsorelaterad livskvalitet, skolnärvaro och skolresultat, ge känslomässigt lidande, minskat deltagande i sociala aktiviteter och en ökad användning av sjukvårds resurser. I den här avhandlingen har barn och ungdomar som sökte fysioterapi inom primärvården för ett muskuloskelettalt smärtproblem studerats. Syftet med avhandlingen var att undersöka de smärttillstånd som barn och ungdomar sökte fysioterapi i primärvård för, och hur biopsykosociala faktorer är associerade till smärtrelaterad aktivitetsnedsättning i dagligt liv. Syftet var också att undersöka genomförbarheten av en beteende medicinsk behandling för den här målgruppen.

Resultaten i studie I visade att många hade haft sin smärta under lång tid, nästan hälften av de som sökte fysioterapeut i primärvård hade haft ont i mer än ett år. Studiedeltagarna rapporterade en lägre hälsorelaterad livskvalitet gällande fysiskt och psykologiskt välbefinnande än barn och ungdomar i normalpopulationen. Många barn hade ont på flera ställen och de med multipla smärtlokalisationer hade än lägre hälsorelaterad livskvalitet än de som hade smärta på ett ställe. Smärtintensitet, upplevelse sin förmåga att själv kunna minska smärtan, oro och hur man hanterar smärta förklarade delvis smärtrelaterad aktivitetsnedsättning. Resultaten indikerar också en könsskillnad mellan flickor och pojkars sätt att hantera smärta. Flickor rapporterade en högre förekomst av katastrofierande medan pojkar använde mer beteende distraktion, vilket generellt sätt anses vara en positiv smärthanteringsstrategi.

Beteendemedicinsk behandling har sin grund en biopsychosocial syn på hälsa och sjukdom, och i den här avhandlingens studie III och IV studerades en beteendemedicinsk behandling med ungdomar med långvarig muskuloskelettalt smärta som målgrupp. Syftet med studie III var att studera genomförbarheten av bland annat datainsamling och rekrytering av patienter samt att studera effekten av det beteendemedicinska behandlingsprogrammet

inriktat mot tonåringar med långvarig smärta. Kontrollbehandlingen bestod av fysisk träning enligt gällande praxis. Trettio-två patienter och deras föräldrar accepterade deltagande, två studiedeltagare blev senare exkluderade. Resultaten visade att datainsamling med hjälp av webb-baserade enkäter fungerade bra i den här målgruppen, med lågt antal uteblivna svar. Rekryteringen av patienter gick långsammare än planerat, med 1.3 patienter per månad från två vårdcentraler och fler än förväntat hoppade av under behandlingens gång. Trots att arbetsgången var omfattande upplevde de behandlande fysioterapeuterna att behandlingsprogrammet möjligt att genomföra. Skräddarsyningen av behandlingen var problematisk och överensstämmelsen mellan de studiedeltagares behov av en viss behandlingssinsats och den utförda behandlingen var låg, 40 %. Den beteendemedicinska behandlingen resulterade i en större minskning av poängen för smärtrelaterad funktionsnedsättning (det primära utfallsmåttet) än i kontrollbehandlingen. Resultaten ska dock tolkas försiktigt eftersom studien hade få deltagare och grupperna hade olika utgångsvärden. Gällande skolfrånvaro (det andra primära utfallsmåttet) och för sekundära utfallsmått (katastrofierande, rörelserädsla och tilltro till egen förmåga i aktiviteter) förelåg inga signifikanta skillnader mellan grupperna efter behandling. I den fjärde delstudien undersöktes hur nöjda studiedeltagarna, deras föräldrar och behandlande fysioterapeuter var med behandlingarnas innehåll och resultat, nöjdhet med sin egen insats under behandlingen samt i vilken utsträckning studiedeltagarna hade förändrat sitt sätt att tänka om smärta. Studiedeltagarna i båda grupperna var nöjda med den behandling de hade fått, deltagarna i beteendemedicin gruppen skattade sin nöjdhet signifikant högre än deltagarna i kontrollgruppen. Fysioterapeuterna intervjuades om sina upplevelser av att lära sig och leverera beteendemedicinsk behandling. Att lära beteendemedicinsk behandling var arbetsamt men givande. Den biopsychosociala grundsynen, att skräddarsy behandlingen och arbetet med föräldrarna identifierades som nyckelaspekter av behandlingen. Att sammanställa funktionella beteendeanalyser upplevdes svårt och krävande och handledning under behandlingstiden värderades högt.

Sammanfattningsvis visar resultaten att barn och ungdomar som söker primärvård för ett smärtproblem i flera avseende liknar dem som behandlas vid specialistkliniker. Flera olika biopsykosociala faktorer samvarierar med smärtrelaterad aktivitetsnedsättning. Att använda ett biopsykosocialt synsätt i undersökning och beteendemedicinsk behandling av ungdomar med smärta är lovande för att hjälpa ungdomar återfå sin hälsa.

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Appendix 1 The Behavior medicine treatment

Pain history

A detailed anamnestic interview was conducted with the participants and their parents, and all the important aspects of the pain problem, including physical, emotional and social triggers of pain, were addressed using a bi-opsychosocial approach (5).

Identification of problems and goal activities

The participants were asked to list 3–5 frequent and important activities in which pain interfered with their performance. The activities were prioritized by their importance to the participant, and self-efficacy was estimated for the performance of each activity.

The participant and PT then worked together to choose the first activity. The activity should be of importance for the participant, and be one with a self-efficacy rating of no less than 4–5 (on a 0–10 rating scale, where 10 indicates “I am very sure I can perform the activity”) to ensure the possibility of success in performing the chosen activity.

Physical examination

The participants underwent a thorough physical examination focusing on areas of importance for the pain-related problem (7, 195), including screening for red flags (potentially dangerous physiological conditions) (196).

Self-monitoring

Self-monitoring of the goal activities was aimed at increasing the participants’ awareness of their actions and thoughts in the goal activity (197) and was recorded in a paper-based diary. The participants were asked to rate their self-efficacy before the activity and to monitor their bodily sensations, thoughts and feelings during the activity. Ideas pertaining to adjustments, thoughts and feelings about the activity performed and the physical and social context were noted in the diary after the activity. Each activity was monitored for one week (or at least 5 times if the goal activity was a non-daily activity).

FBA and identification of behavioral goals

The aim of the FBA (198) was to formulate hypotheses regarding the relationships among the factors (physical, emotional, cognitive and environmental) related to behaviors in the goal activity. The FBA (198) was formulated from the information obtained from the anamnesis, physical examination and self-monitoring.

S-I-R-C (Situation-Individual-Responses-Consequences, which correspond to the ABC from operant learning theory) was used for sorting and summarizing the findings (199). In S-I-R-C, the S stands for situational factors, including antecedents; the I for individual factors (physical and psychological resources and restrictions); the R for behavioral responses; and the C for consequences.

The information from the S-I-R-C was used to form hypotheses regarding the associations among the antecedents, behavioral responses and consequences of the participants' behaviors related to the goal activity. The FBA summarized the reasons that it was not possible to perform the goal activity as desired. The hypotheses formed in the FBA directed the choice of behaviors in the goal activity that required behavioral change, and they formed the foundation of the individually tailored treatment plan. The FBA, goal activities, target behaviors and, accordingly, the treatment plan were continuously modified throughout the treatment process.

Goal setting

SMART (Specific, Measurable, Activity related, Realistic and Time-specified)(200) were set in communication between the participant and physiotherapist, and concrete action planning was initiated. Generally, each goal activity comprised numerous behaviors that required changing; at all times, the behavioral changes were specifically aimed at improving function in the goal activity.

Skills acquisition

This part of the treatment was tailored to the participants' specific problems based on the hypothesis in the FBA, and the SMART goals provided directions for the required training. Pain education based on a biopsychosocial model (1, 5) was introduced and discussed. No exercise or skills were introduced without an explanation of their relationship to the goal activity. Physical and psychological skills were gradually expanded by slowly increasing the number of repetitions, load or level of challenge (7, 8). The purpose was to avoid feelings of failure and to increase the self-efficacy for the exercises, skills or activities rehearsed. The PT provided verbal positive reinforcement when possible.

To increase skills, the treatment followed four phases (121, 122, 124): basic skills acquisition, applied skills acquisition, generalized skills acquisition, and maintenance and relapse prevention.

The objective of the *basic skills* acquisition was to rehearse the basic physical, cognitive or behavioral skills required for improvements in the goal ac-

tivity/ies. Examples of basic skills include performing strengthening exercises for the upper arm and shoulder muscles and learning to recognize a catastrophic thought.

In the *applied skills* phase, basic skills were combined to shape the required behavior by applying and practicing in the goal activity/ies. Examples of applied skills include maintaining the correct position of the scapula when hitting a forehand on the tennis court and handling catastrophic thoughts by restructuring them.

In the *generalization phase*, the application of skills was extended to other activities and situations important to the adolescents. Examples of generalized skills include using the new coordination of shoulder movements, handling catastrophic thinking while playing basketball or helping a neighbor to move heavy objects.

Before the end of treatment, during the *maintenance and relapse prevention phase*, future “risk situations” were identified and discussed. The participants and PTs formed strategies for the maintenance of new behaviors, and ways in which the adolescents and parents could further support positive behaviors were recorded.

Home assignment

Home assignments were given in all sessions during the acquisition of basic skills (the components required for the improvement of basic physical, cognitive or behavioral skills), applied skills (combinations or advances of basic skills in the goal activity) or more generalized skills practiced in activities/behaviors similar to/other than the goal activity. Every session included a follow-up of home assignments from the previous session, and the PT provided positive feedback on performance and demonstrated and practiced new home assignments.

Application of tracks in the BMT

The Exercise Track

The exercise track was primarily applied during the basic skills phase and represented the best clinical practices and was guided by recommendations for repetitions, load and intensity (7, 8). Exercises to improve strength, endurance, circulation, posture control, range of motion, stabilization/muscular control, coordination and aerobic fitness could be chosen. The PT choices from this track were based on the participants’ FBA; every choice should be specifically directed to change a target behavior to incrementally achieve good function in a goal activity.

The Lifestyle Track

The lifestyle track comprised information and techniques aimed at changing sleep behavior, healthy eating and stress reduction to influence goal activities. Standardized information on sleep, healthy eating and stress was provided orally. The participants received a brochure containing the same information to be read as a homework assignment.

The techniques available for addressing sleep problems included information, monitoring sleep in a diary, changes in sleep hygiene and sleep restriction.

The techniques available for healthy eating included information, monitoring emotions and thoughts, and discussions of the causes of unhealthy eating and solutions for healthier eating habits.

The techniques available for stress reduction included information, listing of the causes of stress, and discussions of the causes of and possible solutions for stressful situations.

The PT could use techniques from the behavior change track to support changes for a healthier lifestyle.

The Behavior Change Track

Psychological techniques from the behavior change track were used to support behavior change in the goal activity/ies. In this track, cognitive and behavioral skills promoting behavior change for the activity-related problems were presented. The PTs chose techniques from the set of standardized techniques described below based on each individual participant's FBA and identified barriers to behavior change.

Motivation

The assessment of motivation was performed according to the principles of motivational interviewing (201). Depending on each participant's state of readiness for change (202), the activation and enhancement of motivation were achieved by the PT asking questions regarding the different aspects of taking action for change. Thereafter, the PT initiated the identification of barriers to change, the facilitation of strategies to overcome these barriers. (202).

Problem solving

When a participant lacked ideas or found it impossible to identify any solutions concerning where and how to initiate the introduction of a new, healthier behavior, two problem-solving techniques could be applied. In the first technique, the participant identified all possible solutions to the perceived problem, estimated the positive and negative consequences of each solution

and finally chose the best possible solution. In the second technique, a traffic light analogy was used to prevent the participant from rushing into performing the first action that entered his/her mind (stop – red traffic light) and to urge the participant to plan for action (plan what to do – yellow light) and to proceed with the plan (green light) (203, 204).

Systematic increase in self-efficacy

Increases in SE (19, 76) and operant learning (11, 12) principles were integrated into the workflow. However, in situations in which low SE for an activity was a primary area of concern, the manual provided support on how to structure the increase in SE. Graded exercises or tasks were prompted to increase SE. Beginning with basic, easily performed tasks enabled the participants to succeed. The tasks were then planned to gradually increase in difficulty while remaining achievable (19, 76). When applicable, the PT provided examples of other similar patients who experienced similar struggles and successful solutions according to the principles of model learning (21). Participants who were hesitant to perform a behavior were encouraged to make new attempts. Instant feedback on performance from the PT or parent, as well as from the participants themselves in subsequent stages, was used to reinforce new behaviors.

Automatic thoughts and catastrophizing

With these techniques, the participants learned to identify automatic thoughts and catastrophizing and/or negative thoughts that disturbed performance of the goal activity. When a participant mastered the ability to recognize these thoughts using basic techniques to challenge thoughts, he/she was prompted to think realistically about these thoughts (204). The participants were encouraged to attempt to think differently when performing the activity, and they learned that new thoughts may lead to different feelings. To challenge automatic catastrophizing or negative thoughts during the performance of an activity, the participants were prompted to use positive self-talk (aloud or silently) (205) or distraction (205) before and during the target behavior. Distraction techniques were also taught as an alternative focus for attention when the participants experienced difficulties in attempting to avoid thinking about the pain.

Fears and worries

When fears of movement and/or reinjury hindered behaviors in the prioritized activity, systematic graded exposure (67) in target behaviors was prompted and accompanied by positive verbal reinforcement for every small success provided by the PT, parents and/or participant at home. A small dai-

ly increase in home assignments (for example, increases in time, load or number of repetitions) was negotiated with the participant; this minor increase was to be performed regardless of pain. In addition, “worry time” with a thought-stopping technique for more general worrying was described in the treatment manual.

Relaxation

The relaxation techniques used were based on Fichtel and Larsson’s studies of adolescents with headaches (206). To suit our purpose, a less comprehensive program with new, somewhat shorter recordings was used. The participants accessed these recordings using CDs, MP3 files or an Internet link. Basic, applied and generalized training was followed by the identification of situations in which muscle tension could lead to pain or could interfere with activities as well as strategies for use in case of relapse. The recorded relaxation consisted of six parts:

- 1: Progressive muscle relaxation, longer version
- 2: Progressive muscle relaxation, shorter version
- 3: Relaxation without tensing muscles
- 4: Relaxation without tensing muscles; deep breathing paired with the cue word “relax”
- 5: Relaxation in activity
- 6: Relaxation in activity, shorter version

The parents

The parents of each participant in the experimental condition were invited to a session without their child. Pain education that was based on a biopsychosocial model was introduced and discussed. The education included discussion of the causes of pain, operant techniques, coping, the importance of everyday activities, planning for individual pain management and advice on how parents could support their children in changing behaviors. The agreed-upon forms of support were written on a preprinted worksheet for the parents to take home. On the back of this worksheet, general recommendations for parents were provided (207).

Most participants attended the PT sessions without their parents; therefore, the PT also called the parents every few weeks to inform them about their children’s progress and to plan ahead.

Appendix 2 The control treatment

Pain history

A detailed anamnestic interview was conducted with the participants and their parents, and the important aspects of the pain problem were addressed using a biomedical approach. Psychosocial aspects of the problem were not inquired about but were noted in the medical record if spontaneously mentioned by a participant or parent.

Identification of problem

The participants and their parents were questioned about their reasons for seeking PT, the nature of the problem and what they wanted assistance with.

Physical examination

The participants underwent a thorough physical examination, including screening for red flags potentially dangerous physiological conditions) (196).

Goal setting

The treatment goals were determined together with the participants. The goals were not described in terms of activity-related problems (for example, “pain in the shoulder will decrease from NRS 7 to NRS 3 in three weeks’ time”).

Treatment and application of tracks

The Exercise Track

The exercise track for the control condition consisted of the same instructions as for the experimental condition regarding the recommendations for repetitions, load and intensity (7). For the CT, the exercise track was complemented by a hold/let go muscular relaxation technique and a brief biomedical explanation of pain. The exercises were individualized and based on the participants’ pain-related physical musculoskeletal problems and treatment goals.

The Lifestyle Track

For the control condition, the lifestyle track contained standardized oral information on sleep, healthy eating and stress reduction according to best practices. In addition, the participants received a brochure containing the same information. No techniques for behavior change were applied.

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