Psychosocial Assessment and Self-Management of Chronic Pain

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Psychosocial Assessment and Self-Management of Chronic Pain
PhD thesis, Erasmus MC University Medical Center

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CHAPTER 1

General introduction

Mrs. Janssen is a 35-year-old woman who was in a car accident two years ago. Her arm was broken and she was treated in the hospital. Mrs. Janssen still has pain in her arm and hand. The pain has even increased during the past two years. Medical examination shows no abnormalities. However, Mrs. Janssen finds it hard to do her job and the housekeeping properly, and to care for her two young children. These pain-related disabilities make her feel frustrated and sad. She thinks her pain will never abate.

Julia is a 13-year-old girl who has been going to secondary school for just a few months now. Recently, she started to experience headaches and abdominal pain. Additionally, she often stays home from school. At home, Julia stays in her room all day and sleeps more than usually. Julia's mother is worried about Julia's health. Therefore, she took Julia to a GP two weeks ago. The GP could not find a medical explanation for the pain and suggested it could be caused by stress. Julia's mother is calmer, for the moment, but Julia's pain does not abate.

1.1 AIM OF THE THESIS

Chronic pain is prevalent in both children and adults and has major negative consequences for daily life, e.g. reduced participation in activities, depressive feelings, or anxiety. Therefore, it is important to signal and treat chronic pain in its early stage. This thesis aims to provide answers to two important questions: 1. How to improve early signaling and assessment of chronic pain in adolescents?; and 2. How to improve self-management of chronic pain in both adolescents and adults?

1.2 CHRONIC PAIN

In 1986, the International Association for the Study of Pain (IASP) formulated the following definition of pain: 'Pain is an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage. Note: pain is always subjective. Each individual learns the application of the word through experiences related to injury in early life'. The model of Loeser [97] describes four different components of pain; nociception, pain sensation, pain interpretation, and pain behavior. Simply stated, if you burn your hand on the gas stove, the pain receptors in the fingers are stimulated. The firing of these pain receptors means that the painful stimulus is registered (nociception). Next, the painful stimulus is transferred to the sensory cortex, where pain sensation takes place; the location, nature, and intensity of the pain become clear. The painful stimulus is also transferred to the prefrontal cortex, where it is interpreted; harmless, dangerous or depressing. Finally, you act upon it, for example by taking medication, taking a rest, or visiting the hospital.

In adults, chronic pain is understood to be pain that is present for more than six months, recurrently or continuously (IASP, 1986). Approximately 20% of the adult European population is affected by chronic pain [18,19,88]. Chronic pain is more common among women, older people, and people with a low socio-economic status. Chronic pain negatively influences people's quality of life. Many people with chronic pain are often no longer able to take perform activities, such as household chores, working

outside home, or maintaining relationships with family and friends. Additionally, approximately 20% of the patients with chronic pain report to have become depressed because of their pain. Besides these individual consequences, the burden to society is enormous, in view of the escalating health care costs and loss of productivity. In Europe chronic pain seems to cost society billions per year [19]. Unfortunately, the results of chronic pain management remain insufficient.

1.2.1 Chronic pain in children and adolescents

Most chronic pain studies have been performed among adults. However, chronic pain is also highly prevalent among children and adolescents [133]. Most pediatric studies define chronic pain as pain that persists for at least three months, continuously or intermittently [105,133,147]. Approximately a quarter of the children and adolescents in the open population suffer from chronic pain [133]. The prevalence of chronic pain is higher among girls than among boys and peaks in girls aged 12 to 14 years. Limb pain, headache, and abdominal pain are the most frequently reported types of pain. The combination of headache and abdominal pain often occurs [60]. Chronic pain in children also seems to persist over years [16,109,136]. In a longitudinal study, Brattberg and colleagues [16] reported that 59% of the girls and 39% of the boys who reported pain in childhood also reported pain in early adulthood. Another study found that 30% of the children and adolescents with chronic pain at baseline still experienced chronic pain two years later [136].

Chronic pain reduces the quality of life of children and adolescents [71] because they often cannot meet with friends or pursue hobbies [60]. Moreover, chronic pain is often associated with disturbed sleep, loss of appetite, less emotional well-being, and high medication use. Last but not least, these youngsters often stay home from school [43,119,127,134].

Attending school for children and adolescents can be seen as compared to going to work for adults. Therefore, school absenteeism is one of the most important measures of pain-related disability among children and adolescents. Only a few studies have, however, examined whether school absenteeism for this reason affects academic performance. The results of these studies are conflicting. One study suggests that adolescents with chronic pain have lower academic performance than adolescents without chronic pain [99]; another suggests that they score within average range on academic performance measures [65]. Furthermore, the mechanisms (e.g., attention, emotional well-being) that could explain the relation between chronic pain and academic performance are unknown. This thesis addresses relationships between self-reported disabling pain, emotional well-being, and attention and mentor-reported grades in a large normal population sample of Dutch adolescents. Additionally, the mediating mechanisms in the relationship between pain and school functioning is examined.

Psychosocial stress seems to be a risk factor for the occurrence of pain [30,102,123,178]. For example, childhood sexual and physical abuse, parent-reported family conflict, and being bullied during childhood were found to be risk factors for chronic pain at adult age [13,42,49,92,94,146,147,177]. However, the association between adolescent-reported negative life events and adolescent pain has not been studied before. This thesis examines whether negative life events, e.g. bullying, abuse, and family conflict, are risk factors for chronic pain in adolescents.

In summary, chronic pain is a big problem among children and adolescents. The high prevalence rates over time show that results of pain management for children and adolescents, as for adults, still remain insufficient [184].

1.3 THE BIOPSYCHOSOCIAL MODEL OF PAIN

For a long time pain has been explained by the biomedical model, which assumes that pain is always caused by physical damage. Over the years, however, the perspective on pain has changed from a biomedical to a biopsychosocial perspective, as studies have shown that the levels of pain and disability are only partly explained by physical abnormalities [51,175]. On the other hand, psychological disability and pain-related disability are not fully explained by the intensity of pain.

The biopsychosocial model [166] states that pain is caused by a complex interaction between biological, psychological, and social variables. Differences in the interaction between biological, psychological, and social variables may explain differences in pain expression between individuals. People with identical levels of pain intensity may cope very differently with their pain symptoms. For example, some will quit their jobs and become inactive, whereas others continue working and performing other activities. Non-physiological factors also seem to be crucial in the development and maintenance of chronic pain [95,170]. Additionally, psychosocial factors can influence treatment response [170].

1.3.1 Biological factors

Pain symptoms that are not or not sufficiently explained by physical damage may originate from the phenomenon of central sensitization (CS). CS involves an abnormal and intense enhancement of pain experience by mechanisms in the central nervous system. CS is characterized by allodynia (painful sensation to a normally non-painful stimulus, such as touch), hyperalgesia (excessive sensitivity to a normally painful stimulus, such as pressure), expansion of the receptive field (pain that extends beyond the area of peripheral nerve supply), and unusually prolonged pain after the stimulus has been removed (usually burning, throbbing, tingling, or numbness). Dysregulation of both ascending and descending central nervous system pathways due to physical trauma and sustained pain impulses has been implicated in CS, as well as the chronic release of pro-inflammatory cytokines by the immune system as a result of physical trauma or viral infection [186]. It is known that psychosocial factors may influence CS [185].

1.3.2 Psychological factors

1.3.2.1 Cognitions

A person's beliefs about the controllability of pain can influence the level of pain and related disability [93]. Many people believe they cannot exert personal control over their pain; it just happens to them. Such a negative thought may cause feelings of helplessness and demoralization. As a consequence, the pain is felt more intensely and efforts to become active again are discarded. Self-efficacy is closely related to

beliefs about controllability, and as a cognitive factor influences chronic pain too. Pain self-efficacy refers to the belief that one is capable to tolerate pain and to perform activities despite pain [4,9]. Research has shown that high self-efficacy regarding pain control is related to a higher tolerance of pain [41,96].

Additionally, cognitive errors, such as catastrophizing, overgeneralization, and selective abstraction, are related to maladaptive pain behaviors. Catastrophizing refers to the tendency to think extremely negatively about pain and its consequences. Examples of catastrophic thoughts are "my thoughts are always concentrating on the pain" and "the pain will never subside". Overgeneralization refers to the conviction that similar events always result in the same amount of pain. For example, when a walk to the mall has caused extreme pain once, one may be convinced that this will happen again. Selective abstraction refers to the inclination to focus on the negative aspects of an event. For example, only remembering that the pain increased at a party, but not thinking back to the nice conversations with friends. Of all the cognitive errors, catastrophizing seems to influence pain, pain-related disability, and distress the most [80]. In children and adolescents, pain-related disability also seems to be strongly influenced by catastrophizing [68].

1.3.2.2 Emotions

To suffer from chronic pain and its consequences is emotionally burdensome. Approximately 40-50% of the people with chronic pain report depressive symptoms [143]. Especially those who feel they cannot function anymore and cannot control their pain become depressed. Besides depressive symptoms, anxiety is very common among people with chronic pain. Many tend to attribute their pain to a specific trauma, for example an accident. Posttraumatic stress disorder (PTSD) is two to three times more prevalent among people with chronic pain than among the general population [5,38]. PTSD is caused by a traumatic event and is characterized by nightmares, avoidance of thoughts or activities related to the traumatic event, and increased arousal. Furthermore, anger is a common emotion [47], and so is frustration, for instance because it is not known what causes the pain, previous interventions failed, or lack of control became apparent [126]. Twenty-eight percent of children and adolescents with unexplained chronic pain were found to suffer from a psychiatric disorder, which proportion is thrice as much as that among healthy children and adolescents [83]. Anxiety (12%), affective (mainly depressive disorders; 19%), and disruptive disorders (ODD and ADHD; 9%) were most prevalent. Children and adolescents with headaches have a higher risk of psychiatric disorder than children and adolescents with other types of pain.

1.3.3 Social factors

An important social factor that influences chronic pain is social learning. Social learning is defined as learning by observation of others [8]. The behavioral responses of family members to chronic pain affects a persons' own behavioral responses to chronic pain [172]. For example, if a parent responds with withdrawal, the child is primed to use the same strategy when experiencing pain.

A second social factor that influences chronic pain is operant learning. Operant learning means that behavioral responses are used more if these responses frequently have been reinforced by the environment [153]. For example, when a partner responds to a persons' chronic pain in an attending and caring manner, the chronic pain itself will be reinforced. And then, more solicitous or encouraging responses of parents (e.g. frequent attending to pain symptoms, giving permission to stay home from school) seem to increase pain intensity and functional disability of children and adolescents with chronic pain [137].

In summary, chronic pain is a multi-dimensional syndrome. Chronic pain has many psychosocial consequences, which, in turn may sustain pain symptoms. Therefore, patients with chronic pain should be assessed and treated from a biopsychosocial perspective. This thesis therefore, places a focus on the psychosocial assessment and self-management of chronic pain.

1.4 PSYCHOLOGICAL TREATMENT OF CHRONIC PAIN

1.4.1 The consequences model

Different behavioral models for the psychological treatment of chronic pain different behavioral models exist. In this thesis the so-called consequences model takes a central place. This model underlines that one should stop looking for causes of pain and instead be focusing at the consequences. The original model of Speckens and colleagues [157] assumes that pain-related thoughts arise first. These thoughts lead to physical, cognitive, emotional, behavioral, and social consequences. Subsequently, these consequences would sustain or worsen the pain. According to the consequences model the aim of treatment is to reduce the consequences of pain; especially the consequences that hamper natural recovery, e.g. depressive feelings and social isolation. Zonneveld and colleagues [187] hypothesize that patients will have difficulty accepting this non-physical perspective. Patients do perceive their pain symptoms first; not their pain-related thoughts. Zonneveld and colleagues [187] modified the consequences model to assume that the consequences of pain arise first and lead to pain-related thoughts. Subsequently, the consequences (sometimes in reaction to pain-related thoughts) worsen the pain. According to this modified consequences model the aim of treatment is to better deal with the consequences of pain and thereby improving quality of life.

1.4.2 Cognitive-behavioral therapy for adults with chronic pain

Cognitive behavioral therapy (CBT) refers to a set of psychotherapeutic techniques to help patients understand how thoughts and emotions influence behavior. CBT fits in with the model of consequences and is used to reduce pain and improve quality of life. Research has shown strong evidence for CBT reducing headaches [132]. A systematic review has made clear that CBT is also an effective treatment for adults with non-headache chronic pain [183]. CBT improves quality of life and reduces psychological distress and, only to a small extent, pain. So far, however, the effect sizes of CBT for persons with non-headache pain are modest; perhaps because it is still unclear what types of patients with chronic pain would benefit the most from CBT.

1.4.3 Cognitive-behavioral therapy for children and adolescents with chronic pain

CBT and relaxation exercises could effectively reduce pain frequency and pain intensity in children and adolescents, with large effect sizes for recurrent headaches and medium effect sizes for non-headache recurrent pain [44,45]. Only few studies have examined if psychological treatment also enhances adolescents' well-being; it is unclear whether levels of anxiety and depression also decrease after treatment.

1.5 MINIMAL CONTACT INTERVENTIONS FOR CHRONIC PAIN

Access to CBT is still limited because of high costs, mobility limitations, long waiting lists, and availability of too few health professionals specialized in chronic pain. Greater access could be achieved by using so-called minimal contact versions of psychological interventions; i.e. with shorter contact time with the therapist. This means that patients need to take a more active role in their own treatment, which also may improve treatment outcome. Minimal contact interventions have proven to be at least equally effective as vis-à-vis treatments in treating chronic pain [58,145].

1.5.1 Behavioral management training provided by lay trainers

One specific example of a minimal contact intervention is behavioral management training (BMT) provided by lay trainers. BMT for migraine was converted to a self-management format and provided to small groups at the lay trainers' home [118]. The intervention was targeted at identifying migraine triggers and internalizing self-regulation skills. This approach seemed effective in increasing self-efficacy and perceived control over migraine attacks [117,118]. In this thesis the sustained effects of BMT by lay trainers at the long-term are addressed.

1.5.2 Internet interventions for adults with chronic pain

Another example of a minimal contact intervention is internet therapy. An important advantage of internet therapy is that is available at any place and any time. Recent studies have evaluated internet therapy for chronic pain and found reductions in the levels of pain and pain-related disability [10,107]. Results are inconsistent, however, when it comes to emotional well-being. Some studies found significant reductions of depression and anxiety levels [17]; others did not [20].

1.5.3 Internet interventions for adolescents with chronic pain

To adolescents psychological interventions using an Internet format might be more attractive than face-to-face therapy since Internet is the top health information resource for this age group [56]. Stinson and colleagues [161] concluded in their review that Internet-based self-management interventions seem to improve health outcomes, including pain, in children and adolescents with various health conditions. In 2005, Hicks and colleagues [64] tested the efficacy of online CBT treatment for adolescents with chronic pain in the open population. The main therapeutic elements were psycho-education, relaxation techniques (i.e., deep breathing, relaxation, visualization/imagery), cognitive strategies (e.g., self-talk),

and promoting healthy lifestyle choices. Parents were also involved in the intervention; they received education on how to encourage healthy behavior (e.g., do not reward pain behavior). One month after treatment the adolescents in the experimental group reported significantly more pain reduction than did adolescents in the waitlist group. More recently, Palermo and colleagues [128] evaluated an Internet-delivered family CBT intervention. The children were instructed how to identify stress, to apply deep breathing and progressive muscle relaxation, and to modify cognitions about pain and functional ability. The children's sleep habits and activity pacing were addressed as well. The parents were encouraged to reinforce positive behavior of their child (e.g., staying active) of their child. Results showed that pain related disability and pain intensity were more reduced in the Internet group than in the waitlist group. These effects were sustained through the three months follow-up period. In summary, these findings suggest that Internet interventions may be a promising alternative for face-to-face therapy in teenagers with chronic pain. However, more research is needed. For example, most studies have examined internet therapy for adolescents with chronic pain without clinician contact. In other populations (e.g., suffering from depression) Internet therapy with therapeutic support seems to be more efficacious than Internet therapy without support [35,158]. In this thesis the efficacy of a guided online self-management intervention for adolescents with chronic pain (Move It Now) is studied.

1.6 AIM AND RESEARCH QUESTIONS OF THE CURRENT THESIS

The first part of this thesis (**chapters 2-3**) focuses on the prevalence and psychosocial assessment of chronic pain. The following research questions are addressed:

- Are negative life events, e.g. bullying, abuse, and family conflict risk factors for chronic pain in adolescents? (**chapter 2**)
- Is disabling pain associated with lower grades, reduced emotional well-being, and more attention problems? (**chapter 3**)

The second part of this thesis (**chapters 4-6**) explores the efficacy of self-management interventions for chronic pain. The following research questions are addressed:

- Was improvement in migraine frequency, self-efficacy in managing headache episodes, and headache specific internal control resulting from BMT sustained or even more marked two to four years after BMT? (**chapter 4**)
- What are the effects of a guided interactive internet intervention, Move It Now, for adolescents with chronic pain? (**chapter 6**)

In **chapter 5** the study protocol for the Move It Now studies is presented.

CHAPTER 2

Bullying, abuse, and family conflict as risk factors for chronic pain among Dutch adolescents

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Bullying, abuse, and family conflict as risk factors for chronic pain among Dutch adolescents. Eur J Pain, Mar 5. doi: 10.1002/ejp.689.

ABSTRACT

Purpose

Psychosocial stress seems to serve as an important risk factor for the occurrence of pain. The present study aims to examine if early adversities, e.g. bullying, abuse, and family conflict are risk factors for chronic pain in adolescents. The secondary aim of the present study is to describe the pain characteristics of chronic pain in adolescents in a community sample of Dutch adolescents.

Methods

Participants in the present study were 15,220 adolescents, attending schools (grade 7 and 8) in Rotterdam, the Netherlands. Chronic pain was measured with a newly developed questionnaire; the Pain Barometer. Early adversities were measured using single item questions from the Rotterdam Youth Monitor, a longitudinal youth health surveillance system. Cross-sectional associations between early adversities and chronic pain were investigated using logistic multilevel analysis, adjusted for potential confounding.

Results

In school year 2010-2011, 9.2% of the 15,220 adolescents reported chronic pain. Physical abuse by others (OR=1.51, 95% CI=1.07-2.14), sexual abuse (OR=1.46, 95% CI=1.05-2.05), family conflict (OR=1.79, 95% C=1.61-1.99), and being bullied (OR=1.23, 95% CI=1.17-1.29) are more common in adolescents with chronic pain. Physical abuse (OR=1.28, 95% CI=.95-1.71) by parents and parental divorce (OR=1.07, 95% CI=.93-1.22) were not significantly related to chronic pain.

Conclusions

The results of the present study suggest that bullying, abuse, and family conflict may be risk factors for chronic pain in adolescents. Early signaling these stressors might prevent chronic pain.

INTRODUCTION

Chronic pain is a common experience among adolescents [133]. Perquin and colleagues reported that 25% of the children and adolescents (0-18 years) in the general population suffer from chronic pain. Chronic pain is defined as pain that has persisted continuously or intermittently for at least three months. The most common types of chronic pain seem to be limb pain, headache, and abdominal pain [133]. The most prevalent combination is headache plus abdominal pain [60]. The prevalence of chronic pain is higher among girls than among boys and is positively related to age [133]. Chronic pain is most frequent in girls aged 12 to 14 years old. Furthermore, chronic pain seems to persist over years [16,109,136]. A study by Brattberg [16] reported that 59% of the women and 39% of the men who reported pain in childhood also reported pain in early adulthood. A different study reported that 30% of the children and adolescents with chronic pain at baseline, still complained of chronic pain after two years [136].

Chronic pain negatively influences adolescents' quality of life [72]. Many children and adolescents with chronic pain report sleeping problems, an inability to pursue hobbies, and less intense relationships with friends than their contemporaries [60]. Approximately 40% of these children and adolescents also report having been absent from school because of pain. Additionally, in a study by Kashikar-Zuck and colleagues [78] most children with chronic pain reported mild to moderate levels of depression and 15% reported severe levels of depression. The relationship with school absence and quality of life is an important argument to improve the detection and prevention of chronic pain in children and adolescents. To optimize this detection and prevention, it is necessary to know the risk factors for chronic pain in these adolescents.

Psychosocial stress may serve as an important risk factor for the occurrence of pain. In the general population, childhood adversities (e.g. physical abuse, sexual abuse, neglect, parental death, and parental divorce) seem to be risk factors for the development of chronic physical conditions [148], and more specifically headaches during adulthood [91]. Additionally, several studies have shown that childhood sexual and physical abuse increases the risk of occurrence of chronic pain in adult men and women [13,42,49,92,94,177]. A retrospective study of Fillingim and Edwards showed that pain complaints in early adulthood are associated with self-reported history of childhood abuse, sexual or physical [49]. The results of a study by Sansone and colleagues [146] showed that being bullied in childhood was associated with higher pain ratings in adulthood. A retrospective study by Scharff and colleagues showed that distress and disability among children with chronic pain is associated with high levels of parent-reported family conflict [147]. In summary, previous research suggests that early adversities, e.g. bullying, abuse, and family conflict, are related to chronic pain in adolescents. To our knowledge, the association between adolescent-reported early adversities and adolescent pain has not been studied before.

Therefore, the main aim of our study is to examine if early adversities, e.g. bullying, abuse, and family conflict are risk factors for chronic pain in adolescents. The secondary aim of the present study is to describe the pain characteristics of chronic pain in adolescents in a community sample of Dutch adolescents.

METHODS

Participants

Until 2011, the Municipal Health Services (GGD) in Rotterdam-Rijnmond preventively examined health conditions and developmental problems annually in approximately 15,000 adolescents in grades 7 and 9 of most secondary schools (±85%) in the Rotterdam area. They did this using the Rotterdam Youth Monitor (RYM), which is a longitudinal youth-health surveillance system. For the present study we used data on 15,220 participants of the RYM from the year 2010-2011, which represents a response rate of 87.41%.

Measures

Demographic data

Demographic data, including adolescents' gender, age, education level, and Dutch vs. non-Dutch origin, were measured using self-constructed questions. Age was classified into eight age groups: ≤11, 12, 13, 14, 15, 16, 17 and ≥ 18 years old. Education level was classified into six levels: three levels of pre-vocational secondary education (with increasing difficulty), higher general secondary education, a combination of higher general education and pre-university education, and pre-university education. Adolescents were defined as non-Dutch when at least one of their parents was born outside the Netherlands (CBS, 2012).

The Pain Barometer

The Pain Barometer is a self-constructed signaling instrument for chronic pain in adolescents aged 12 to 17 years. The instrument is based on the Pain Questionnaire [133], literature research, and on expert knowledge (e.g., a medical doctor and researcher from Rijndam rehabilitation center, psychologists from the Erasmus MC University Medical Hospital, and researchers, nurses, and a medical doctor from the Preventive Youth Health Care in Rotterdam). The Pain Barometer consists of 12 questions about several characteristics of pain. The first question is: "Did you experience pain in the previous three months?". If the answer is "no" no further questions are asked. The second question is: "For how long do you already experience pain?". If the answer is "less than three months" no further questions are asked. The third question is: "Only for girls: Is your pain related to your menstrual cycle". If the answer is "My pain is exclusively related to my menstrual cycle" no further questions are asked. The next six questions concern the location (10 predefined answer categories), location of most pain (10 predefined answer categories), frequency (6 predefined answer categories), onset of pain (4 predefined answer categories), and cause of their pain (7 predefined answer categories and 1 open category). The final three questions concern the intensity and the impact of chronic pain, i.e. the interference of pain with daily activities and the subjective burden of pain. These three questions were answered on a 0-10 (11-point) numerical rating scale. The Pain Barometer has not yet been validated. A copy of the Pain Barometer is included in appendix 1.

Early adversities

Early adversities were measured by six single items included in the RYM. The early adversities measured were physical abuse by parents, physical abuse by others, sexual abuse, being bullied, parental conflict, and parental divorce. Abuse and family conflict were measured with the question: "Do or did you ever experience the following negative events? 1. Regular arguments between parents 2. Divorce of parents 3. Physical abuse by (one of) your parents 4. Physical abuse by other adults 5. Sexual abuse (e.g., against your will forced into sexual activities, sexually assaulted, raped). The abuse and family conflict items were answered on a dichotomous scale (with *1* = no, never experienced and *2* = yes, experienced or experiencing now). Bullying was measured with the following question: "How frequently have you been bullied at school during the last three months?". This item was answered on a 5-point scale (with *1* = never, *2* = less than twice per month, *3* = two or three times per month, *4* = once a week, and *5* = multiple times per week). The items measuring the psychosocial stressor were self-constructed. No psychometric properties are available.

Study design and procedure

The design of the present study was cross-sectional. Between September 2010 and July 2011 the RYM included a chronic pain measure, i.e. the Pain Barometer. Data was collected throughout the school year, except during the summer holidays. Most questionnaires were administrated in the period November-December. Approximately 85% of the secondary schools in Rotterdam participated in the RYM. Specially trained youth health care nurses carried out the administration of the RYM during class hours. Informed consent was not requested, as the data was gathered from within, and as part of, the government approved routine preventive health examination of youth. Only anonymous data were used. Parents received written information on the RYM and were free to object to their child's participation. The study was approved by the Ethics Committee of the Erasmus MC, Rotterdam, The Netherlands.

Data analysis

Participants were classified as having chronic pain if they reported having pain for more than three months (all day or intermittently), and if pain in girls was not associated with their menstrual cycle. A simple algorithm was used to combine the first three questions of the Pain Barometer to derive the outcome variable chronic pain. Participants were considered as having chronic pain if they answered "yes" to the first question, "more than three months all day long" or "more than three months, intermittently" to the second question, and "this pain is not exclusively related to my menstrual cycle" to the third question (girls only).

The frequencies describing the chronic pain characteristics are presented. To examine the association between the early adversities and chronic pain, binomial logistic multilevel analysis was applied, with school location as upper level and children as lower level using the GENLIN module in SPSS 21. The multilevel approach was used to account for school dependent differences. Age, gender, and Dutch vs. non-Dutch origin were taken as covariates, because these demographic variables were significantly correlated with chronic pain.

RESULTS

Participant characteristics

Of the participating adolescents, 52% were Dutch. Gender was almost evenly distributed (49% female). Most adolescents were aged 12 years old (35%) and involved in pre-vocational secondary education (52%). The frequencies of all participant characteristics are reported in table 1.

TABLE 1 Chronic pain characteristics and frequencies of the early adversities for adolescents with chronic pain vs. no chronic pain

	Total	Chronic pain	
		No	Yes
	N = 15,220	N = 13,802	N = 1,398
Total	100%	91%	9%
Participant characteristics			
Gender (Female)	49%	89%	11%
Dutch origin	52%	90%	10%
Age			
<11	1%	1%	1%
12	35%	35%	30%
13	22%	22%	20%
14	25%	25%	26%
15	15%	15%	19%
16	3%	3%	4%
17	<1%	<1%	<1%
>18	<1%	<1%	<1%
School level			
Prevocational secondary education (three levels)	56%	56%	58%
Higher general secondary education	11%	11%	12%
Higher general secondary education + Pre university education	15%	15%	14%
Pre-university education	18%	18%	16%
Chronic pain characteristics			
Pain location			
Headaches			42%
Eyes			9%
Throat, nose, and ears			9%
Teeth			3%
Neck			17%
Breast			10%
Hips, pelvis, or abdomen			21%
Back			30%
Arms, shoulders, or hands			14%
Legs, groins, or feet			43%

TABLE 1 (continued)

	Total	Chronic pain	
	N = 15,220	No N = 13,802	Yes N = 1,398
Pain frequency			
<1 per month			6%
Once per month			4%
2-3 per month			12%
Once per week			13%
>2 per week			35%
Daily			31%
Onset of pain			
3-6 months			35%
6-12 months			29%
1-5 years			27%
>5 years			10%
Idea of cause of pain			
No idea			23%
Injuries			25%
Stress			19%
Physical overuse			18%
Disease			5%
Innate			4%
Accident or operation			3%
Consulted healthcare providers			
No healthcare provider visited			32%
G.P.			43%
Hospital			22%
Physiotherapist			24%
Psychologist or social worker			4%
Early adversities			
Physical abuse			
By parents	2.8%	3%	6%
By others	2.6%	3%	5%
Sexual abuse	1.7%	2%	4%
Regular arguments	35%	33%	51%
Parental divorce	21.2%	21%	27%
Bullying			
Never	84%	85%	76%
Less than twice per month	9%	9%	12%
Two or three times per month	2%	2%	3%
Once per week	2%	2%	3%
Multiple times per week	3%	3%	5%

Chronic pain characteristics

In the school year 2010-2011, 9.2% (1,398 out of 15,200) of the adolescents reported chronic pain. Most of these adolescents reported having pain in their legs, groin or feet (43%; 573 out of 1,349), headaches (42%; 568 out of 1,349), back pain (30%; 401 out of 1,349), and/or pain in their hips, pelvis or abdomen (21%; 289 out of 1,349). A third (35%; 493 out of 1,398) of the adolescents reported having pain twice or more times per week. Of all adolescents with chronic pain, 35% (479 out of 1,390) reported being in pain from 3-6 months. Most of the adolescents with chronic pain reported that they had an idea about the cause of their pain (77%; 1,080 out of 1,396). Frequently reported causes were: (sports) injuries (25%%; 354 out of 1,395), stress (19%; 266 out of 1,395), physical over-exertion (18%; 246 out of 1,395), disease (5%; 64 out of 1,395), innate (4%; 62 out of 1,395), and an accident or an operation (3%; 46 out of 1,395). More than half (68%; 944 out of 1,396) of the adolescents had visited a healthcare provider in the previous year because of their pain: 43%% visited a G.P. (600 out of 1,396), 22% (311 out of 1,396) visited the hospital, 24%% (335 out of 1,396) visited a physiotherapist, and 4% (51 out of 1,396) visited a psychologist or social worker. Half of the adolescents (53%; 745 out of 1,397; $M = 5.85$, $SD = 5.07$) who reported chronic pain scored 5 or higher (on a scale from 0 to 10) on the item 'how severe was their pain' during the last week. On the item 'if their pain interfered with school, work, hobbies, and/or sports' 41% (572 out of 1,396; $M = 4.90$, $SD = 5.21$) scored a 5 or higher. More than half of the adolescents (57%; 797 out of 1,396; $M = 6.17$, $SD = 5.24$) scored 5 or higher on the item indicating the level of subjective pain-related burden. The frequencies of most pain characteristics are shown in table 1.

Pain and its association with participant characteristics

Among adolescents with chronic pain there were more girls than boys (OR = 1.57, CI 95% = 1.38-1.78). Additionally, chronic pain increased with age (OR per year = 1.14, CI 95% = 1.08-1.20). More Dutch than non-Dutch adolescents reported chronic pain (OR = 1.29, CI 95% = 1.14-1.47). No differences were found for school level (OR = 1.02, CI 95% = .93-1.11). The associations between pain and participant characteristics are shown in Table 2.

Pain and its association with early adversities

Physical abuse by others (OR = 1.51, CI 95% = 1.06-2.13), and sexual abuse (OR = 1.46, CI 95% = 1.04-2.05) are more common in adolescents with chronic pain. Additionally, adolescents with chronic pain are more at risk of being bullied at school (OR per point on the 5-point scale = 1.22, CI 95%= 1.16-1.29). Regular arguments between parents (OR = 1.79, CI 95% = 1.61-1.99) also occurred more in adolescents with chronic pain than in adolescents without chronic pain. Physical abuse by parents and parental divorce were not significantly related to chronic pain (OR = 1.27, CI 95% = .95-1.71 and OR = 1.07, CI 95% = .93-1.22 respectively). The reported OR's are adjusted for confounding by age, gender, and Dutch vs. non-Dutch background. The frequencies of the early adversities for the adolescents with chronic pain versus adolescents without chronic pain are shown in table 1. The details of the binomial logistic multi-level regression analysis are reported in table 2.

TABLE 2. Association of the early adversities with chronic pain, crude and adjusted for potential confounding

Effect	Crude estimates			Adjusted estimates		
	p-value	Odds ratio	[95% CI]	*p*-value	Odds ratio	[95% CI]
Constant	<0.001	0.010	[0.007-0.014]	< 0.001	0.004	[0.003, 0.007]
Girl vs. boy				< 0.001	1.569	[1.383, 1.780]
Age				< 0.001	1.142	[1.082, 1.204]
Dutch vs. non-Dutch				< 0.001	1.292	[1.138, 1.468]
School level				0.65	1.020	[0.934, 1.114]
Being bullied	<0.001	1.199	[1.142-1.260]	< 0.001	1.226	[1.166, 1.290]
Regular arguments between parents	<0.001	1.908	[1.725-2.111]	< 0.001	1.791	[1.614, 1.986]
Parental divorce	0.36	1.065	[0.931-1.218]	0.34	1.067	[0.933, 1.221]
Physical abuse by parents	0.10	1.285	[0.954-1.730]	0.11	1.275	[0.951, 1.709]
Physical abuse by other adults	0.10	1.333	[0.947-1.877]	0.02	1.511	[1.066, 2.142]
Sexual abuse	0.001	1.717	[1.238-2.383]	0.03	1.463	[1.046, 2.048]

Binomial logistic multi-level regression analysis.

CONCLUSION AND DISCUSSION

The main aim of the present study was to examine whether early adversities are associated with chronic pain. The results suggest that bullying, physical abuse by others, sexual abuse, and regular arguments between parents are more prevalent in adolescents with chronic pain than in adolescents without chronic pain. Although it was found that physical abuse by parents and parental divorce are more prevalent in adolescents with chronic pain, these associations were no longer significant after controlling for gender, age, Dutch vs. non-Dutch origin, and school. These findings are partly in accordance with the findings in the studies by Scott [148] and colleagues and Lee and colleagues [91]. Both of these studies found an association between physical abuse by parents and chronic pain. Scott and colleagues [148] did find an association between parental divorce and chronic pain, but Lee and colleagues [9] did not.

A relatively new and important finding is that bullying seems to be more common among adolescents with chronic pain. Theoretically, both directions of effect seem possible. There is some evidence that children with chronic physical disability are at risk of becoming victims of bullying [138]. On the other hand, it seems also possible that being bullied causes so much stress that children become more vulnerable to experiencing (unexplained) pain.

The secondary aim of the present study was to describe the characteristics chronic pain in adolescents in a community sample of Dutch adolescents. The findings show that a substantial group of the Dutch adolescents (9.2%) experience chronic pain. The most common types of pain reported were pain in the legs, groin, and feet, headaches, and back pain. Most adolescents suffering from pain reported pain twice per week, from 3-6 months. More than half of these adolescents reported that they experienced their pain as a big problem. Additionally, many adolescents perceived their pain as interfering with their daily activities, e.g. hobbies and/or sports. The prevalence rates found in our study are hard

to compare with the rates found in other studies, because of the large methodological differences. For example, in the study by Perquin and colleagues [133] a much wider age-range was used. Additionally, they did not exclude the chronic pain cases that were exclusively related to the menstrual cycle. These differences might be the reason for the much larger prevalence (25%) found by Perquin and colleagues. Roth-Isigkeit [144] and colleagues found a prevalence of chronic pain of 9.5%, which is quite comparable with the prevalence found in the present study. Van Dijk and colleagues found a prevalence of chronic pain of only 6% [3]. An explanation for the somewhat lower prevalence might be found in the age group studied (9-13 years). Previous studies have shown that the prevalence of chronic pain increases with age [60,133]. Also, the results of our study suggest that the prevalence of chronic pain is higher for adolescents aged 14-16 years than for adolescents aged ≤ 13 years.

It is noteworthy that we found chronic pain to be more common among Dutch adolescents than among non-Dutch (48%) adolescents. Our finding is hard to compare with previous research, because studies examining the relationship between chronic pain and immigration in adolescents are rare [54,102]. Furthermore, the definitions used for immigration vary enormously. For example, a Finnish study by Luntamo and colleagues [102], found that immigrants experienced more chronic pain. The adolescents in this study self-identified themselves as either an immigrant or Finnish. In the present study adolescents were defined as non-Dutch when at least one of their parents was born outside the Netherlands. Our finding that chronic pain among non-Dutch adolescents is less common, might be explained by the 'healthy immigrant effect', which refers to the finding that immigrants just arriving in their new country are in better health than their native counterparts [162]. However, in our study only 9% of the adolescents were born outside the Netherlands.

An important strength of this epidemiological study is the large sample of a non-referred population, which increases the reliability and generalizability of the results. A limitation of the present study is first, its cross-sectional design, which makes it impossible to make inferences about cause and effect. A longitudinal study is needed to find out whether early adversities cause chronic pain or the other way round. A second limitation of the present study is that chronic pain was measured with a non-validated instrument; the Pain Barometer. This questionnaire includes 0-10 numerical rating scales, commonly used to assess the intensity and impact of pain [34,76,101]. Furthermore, early adversities were measured using single items only. The reliability and validity of our measures for chronic pain and early adversities need further investigation. Additionally, the questions about early adversities were retrospective, which increases the risk of data distortion by memory biases. Another limitation is that early adversities, especially abuse, might be underreported. The offender might have pressurized the adolescent not to tell anyone about what happened. Also, feelings of shame may have prevented the adolescents from reporting such emotional information.

In conclusion, our findings suggest that chronic pain is a common problem among Dutch adolescents to the extent that that interferes with their daily activities. This result stresses the importance of early signaling and preventing chronic pain. Since early adversities might be risk factors for experiencing chronic pain during adolescence, early signaling these stressors might, prevent chronic pain.

Since the present study is a first step in understanding the relationship between early adversities and chronic pain in adolescents, future studies should also include other early adversities, e.g., psychological abuse and neglect and death of a family member. Additionally, a measure of severity of abuse should be included. The severity of abuse might be more important than simply the occurrence of abuse in determining health effects. Furthermore, the Pain Barometer should be validated.

APPENDIX 1: PAIN BAROMETER

Everyone has experienced pain; everyone falls or stumbles sometimes. Usually, the pain quickly subsides. There are also people with **pain with a longer duration**. Some of these people have pain **all day**; other people have pain that comes **in episodes**, such as migraine or tension headaches. The following questions are about these kinds of **long-term pain**.

1. Have you experienced pain during **the last 3 months**?
   - □ Yes
   - □ No → *you do not need to fill out the other questions*

2. **For how long** have you been in pain?
   - □ More than 3 months, all day long
   - □ More than 3 months, intermittently
   - □ Shorter than 3 months → *you do not need to fill out the other questions*

3. **For girls:** is this pain exclusively related to your period?
   - □ This is pain is not exclusively related to my period
   - □ This pain is exclusively related to my period → *you do not need to fill out the other questions*

You should only answer the following questions, if you have been in pain **for more than 3 months**.

4. What is **the location** of the pain you have been experiencing for more than 3 months? (More than one answer is allowed)
   I am experiencing pain in:
   - □ my head
   - □ my eyes
   - □ my throat, nose or ears
   - □ my teeth
   - □ my neck
   - □ my thorax
   - □ my hips, abdomen or pelvis
   - □ my back
   - □ my arms, shoulders or hands
   - □ my legs, groins or feet

5. If you experience pain at more than one location, please indicate which pain you experience as the **most burdensome**. This pain exists for more than 3 months.
   My pain is most burdensome in:
   - □ my head
   - □ my eyes
   - □ my throat, nose or ears
   - □ my teeth
   - □ my neck
   - □ my thorax
   - □ my hips, abdomen or pelvis
   - □ my back
   - □ my arms, shoulders or hands
   - □ my legs, groins or feet

Please answer the following questions for the pain location you **checked at question 5**. This is the pain you experience as the **most burdensome** and exists for **more than 3 months**.

6. **How frequently** have you been experiencing this pain?
   - □ Less than once per month
   - □ Once per month
   - □ 2 or 3 times per month
   - □ Once per week
   - □ Twice or more per week
   - □ Daily

7. **Since when** do you experience this pain?
   - □ 3 to 6 months ago
   - □ Half a year to a year ago
   - □ 1 to 5 years ago
   - □ More than 5 years ago

8. Do you have any idea about **the cause** of the pain? (More than one answer is allowed)
   - □ No
   - □ Injury, for example caused by sports
   - □ Physical overuse
   - □ Accident or operation
   - □ Illness
   - □ Inherited
   - □ Stress, tensions
   - □ Otherwise, namely……

9. Have you visited any **health care provider** because of your pain during the last six months? (More than one answer is allowed)
   - □ No
   - □ The G.P.
   - □ The hospital
   - □ A physiotherapist, caesar therapist, manual therapist or a mensendieck therapist
   - □ A psychologist or a social worker
   - □ Otherwise, namely……

When answering the following questions, think of the **most burdensome** pain that you have been experiencing **for more than 3 months**. A **higher** number means your pain is **severe**. A **lower** number means your pain is **less severe**.

10. **How intense** has your pain been during the last week?

No Pain	0	1	2	3	4	5	6	7	8	9	10	Worst pain I could think of
	□	□	□	□	□	□	□	□	□	□	□	

11. How much has your pain **interfered with your daily activities**, such as school, work, hobbies or sport, during the last week?

I could do all my activities	0	1	2	3	4	5	6	7	8	9	10	I could do nothing
	□	□	□	□	□	□	□	□	□	□	□	

12. How **burdensome** is your pain to you?

Not burdensome	0	1	2	3	4	5	6	7	8	9	10	Very burdensome
	□	□	□	□	□	□	□	□	□	□	□	

CHAPTER 3

Pain is associated with lower grades, reduced emotional well-being, and attention problems in adolescents

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Pain is associated with lower grades, reduced emotional well-being, and attention problems in adolescents (submitted).

ABSTRACT

Objectives

The purpose of the present study was to determine whether pain is associated with specific aspects of academic performance, i.e. poorer grades, and with factors critical to an adolescent's academic performance, i.e. decreased emotional well-being and attention problems. We hypothesized that the association between pain and school grades is mediated by emotional well-being and attention problems.

Methods

In a large cross-sectional study, we collected data from 2215 pupils, aged 12-13 years old. Pain (no, occasional, and frequent), emotional well-being, and attention problems were measured with self-rating scales. Dutch, English, and math grades were taken as an index of academic performance.

Results

Frequent pain in adolescents was associated with poorer grades (Dutch p=.02 and math p=.01). Both occasional and frequent pain were associated with reduced emotional well-being (p=<.001) and reduced self-reported attention (p=<.001). Furthermore, the association between pain and poorer grades was partially mediated by reduced self-reported attention and for a large part by emotional well-being (p-values=<.001).

Discussion

The present study shows that the association between pain and grades can be explained by reduced emotional well-being and also by attention problems. The results suggest that an intervention targeted at pain in adolescents could have a positive effect on emotional well-being and school performance.

INTRODUCTION

In the general population, approximately 25% of all children and adolescents suffer from chronic pain [133]. Pain in children and adolescents is usually not caused by a serious physical disease [23,53]. Pain-related complaints are commonly described as 'functional' or 'medically unexplained'. Nevertheless, recurrent pain negatively influences the quality of life of these adolescents and their families [71]. Children and adolescents with chronic pain participate less in physical activities and report reduced emotional well-being in comparison with their peers.

Adolescents with recurrent pain are also frequently absent from school [43,119,127,134]. The results of a study by Konijnenberg and colleagues showed that almost half of the children with unexplained chronic pain who were clinically referred had been absent from school for at least 1 to 3 days per month [84]. Even more concerning is that approximately 1 out of 10 of the children with chronic pain reported that they had not attended school for a period of 3 months or longer. Similar results have been found in community samples [144]. The results of a study by Roth-Isigkeit and colleagues showed that almost half of the children and adolescents reporting pain in the preceding three months had missed school because of this pain [144].

Only a few studies have examined whether school absenteeism in adolescents with chronic pain results in problems in academic performance. The results of these studies are conflicting. A cross-sectional study by Logan and colleagues reported that the current grades attained by 4 out of 10 of adolescents with chronic pain were poorer than before the onset of chronic pain. Additionally, adolescents with chronic pain perceived pain to interfere with their school success [99]. In contrast, Ho and colleagues found youngsters with chronic pain to score within the average range on both cognitive and academic performance measures (e.g., word reading, mathematical reasoning and written expression) [65]. These mixed results might be explained by differences in methodology (e.g., sample size, instruments and population characteristics) and differences in outcome measures (e.g., grades and assessment batteries).

Some studies suggest that the association between pain and problems in academic performance might be explained by reduced emotional well-being and/or attention problems. A cross-sectional study by Logan and colleagues [98] showed an association between emotional well-being and poorer academic performance in clinically referred adolescents (aged 12-17 years) with chronic pain. Emotional well-being seemed to have more impact on academic performance than factors such as pain intensity and pain duration. Diminished attention might also negatively affect learning and academic performance in chronic pain patients [40]. Adult chronic pain patients often report attention problems [124]. To our knowledge, data on self-perceived attention problems is not yet available for children and adolescents with chronic pain.

Building on previous work, the present study focuses on the relationship between self-reported pain, emotional well-being, and attention and mentor-reported grades in a large normal population sample of Dutch adolescents in grade 9 (12-13 years old). The following hypotheses were tested: 1. Pain is associated with poorer grades, reduced emotional well-being, and more attention problems. 2. Part of

the association between pain and grades is explained by reduced emotional well-being and/or attention problems. The present study is the first to examine mediating mechanisms in the relationship between pain and school functioning. Knowledge of mediating mechanisms is of value for youth health practitioners and for school teachers, as it may be used to guide interventions.

MATERIALS AND METHODS

Population

The present study is part of the large cross-sectional study that examines risk and protective factors associated with good academic performance and school functioning in the Netherlands. Subjects were recruited between January and April 2011 in grade 7 of ten secondary schools in the Netherlands. The schools were located in the provinces Limburg, Noord-Holland, and Gelderland. Adolescents were sent consent forms, with parents providing informed written consent and children assenting prior to participating in the research. Approximately 83% of the invited adolescents finally participated in the study. Data were collected for 2,215 pupils and 1854 mentors. In the Netherlands, a mentor is a teacher that guides a specific class and helps when the pupils experience problems at school. Adolescents were not compensated for taking part in de study.

Instruments

Demographics

Demographic data included the pupil's gender, age, ethnicity (Dutch, Western minorities, and non-Western minorities), and education level (pre-vocational secondary education, higher general secondary education, a combination of higher general secondary education and pre-university education, pre-university education, and pre-university education with additional classical languages) and was measured using a survey.

Pain

Pain was measured by asking the following question: "During the last three months did you experience any intense pain (e.g. headache or abdominal pain) that interfered with your academic performance?" (0 = *no*, 1 = *occasionally*, and 3 = *frequently*).

Attention

The Attention subscale of the Amsterdam Executive Function Inventory (AEFI) was used to measure self-reported attention problems [169]. The Attention subscale consisted of 6 items (e.g., 'I am easily distracted'). The responses for the items were presented on a 3-point Likert scale (1 = *not true*, 2 = *partly true*, and 3 = *true*). A higher score was indicative of more attention problems (range: 6-18). The psychometric qualities of the AEFI are acceptable [169].

Emotional well-being

The Emotional Well-being subscale of the Maastricht Cognition Questionnaire for Children and Adolescents (MCQCA) was used to measure self-reported emotional well-being [169]. The emotional well-being subscale originally consisted of 8 items (e.g., 'I often worry'), but we removed the item about headaches ('I often suffer from pain in my head'). This item would conceptually overlap too much with the pain measure. The responses for the items are presented on a 3-point Likert scale (with *1* = not true, *2* = partly true, and *3* = true). A higher score was indicative of more emotional problems (range 8-24). The emotional well-being subscale has not yet been validated.

Grades

The schools had monitored the mean English, Dutch, and math grades of the first trimester of the school year. Grades were used to index academic performance. The mentors of the pupils provided the data. In the Netherlands, schools grade on a scale from 1 (very poor) to 10 (excellent), with 5.5 being the cut-off between satisfactory and unsatisfactory.

Procedure

In March or April 2011 the participating pupils filled out a digital questionnaire in a classroom. Answering the questions took approximately 40 minutes. During the administration of the questionnaire there was always a researcher present to answer pupils' questions. The mentor of each pupil was also present and filled out one questionnaire per pupil, that approximately took three minutes. Ethics approval for the present study was obtained from the Scientific and Ethical Committee of the Faculty of Psychology and Education of the VU University, Amsterdam.

Data analyses

For the present study 2,658 pupils were invited to participate in the study. Pupils of parents who did not return a signed consent form and pupils who did not fill out the questionnaire completely, were excluded from data analysis (n = 443, 17%). A complete mentor questionnaire was available for 84% of the remaining pupils (n = 1,854). Chi square analyses and ANOVA were used to examine the relationship between population characteristics and the three categories of pain: no, occasionally, and frequently pain. Multi-level analyses were used to test the relationship between pain and grades, pain and emotional well-being, and pain and attention problems. Schools were taken as an upper level. The covariates we used were gender, age, ethnicity, and school level, since these variables were significantly related to chronic pain in univariate analyses. The English, Dutch, and math grades were root-transformed and the variable emotional well-being was transformed with a Blom transformation [12] to obtain normally distributed outcomes. For all differences effect sizes are reported, with d = .2 indicating a weak effect, d = .5 indicating a medium effect and d = .8 indicating a strong effect [25]. Mediation of attention and emotional problems in significant relations between pain and intellectual performance was analyzed with multilevel Sobel tests.

RESULTS

Participant characteristics

The participating pupils were primarily Dutch (80%) and male (53%). The mean age of the pupils was 12.6 years (*SD* = .62). The majority of the pupils were involved in a combination of higher general secondary education and pre-university education (30%).

Pain

A third of the pupils (32%) reported that they were in pain occasionally. Six percent reported that they experienced pain frequently. Girls were more likely to report pain than boys. Non-Western minority adolescents were more likely to report occasional pain and Western minorities were more likely to report frequent pain. Furthermore, pain was positively associated with age. Additionally, pain differed for education level. Adolescents at the preparatory vocational education level more frequently experienced occasional pain, whereas adolescents at the pre-university educational track level with additional classical languages experienced occasional pain less frequently. Descriptive statistics are presented in table 1.

TABLE 1 Descriptive statistics for no pain, occasional pain, and frequent pain

	Total	**Pain-No**	**Pain-Occasionally**	**Pain-Frequently**	
	n (%)	n (%)	n (%)	n (%)	$\chi^2(df)$ *p*
Total	2115 (100%)	1371 (62%)	713 (32%)	131 (6%)	
Gender					
Boys	1170 (53%)	779 (67%)	331 (28%)	60 (5%)	$23.10_{(2)}$ <.001
Girls	1045 (47%)	592 (57%)	382 (37%)	71 (7%)	
Ethnicity					
Dutch	1753 (79%)	1117 (64%)	542 (31%)	94 (5%)	
Western-minorities	207 (9%)	125 (60%)	59 (29%)	23 (11%)	$25.4_{(4)}$ <.001
Non-Western minorities	236 (11%)	122 (52%)	101 (43%)	13 (5%)	
Education level					
Pre-vocational secondary education	447 (20%)	246 (55%)	174 (39%)	27 (6%)	
Higher general secondary education	177 (9%)	101 (57%)	64 (36%)	12 (7%)	
Higher general secondary education + pre-university education	602 (30%)	373 (62%)	192 (32%)	37 (6%)	$32.08_{(10)}$ <.001
Pre-university education	165 (8%)	119 (72%)	40 (24%)	6 (4%)	
Pre-university education with additional classical languages	620 (31%)	428 (69%)	159 (26%)	33 (5%)	
	mean (sd)	mean (sd)	mean (sd)	mean (sd)	F(df) *p*
Age	12.56 (.62)	12.53 (.62)	12.59 (.60)	12.66 (.69)	$3.61_{(2;2212)}$ 0.03

Academic performance

Frequent pain was significantly related to poorer Dutch (d = .22) and Math grades (d = .24). English grades were not related to frequent pain. Occasional pain was not significantly related to grades (p-values = >.05). Estimated means and standard deviations are reported in table 2. The multi-level models for grades are presented in table 3.

Additionally, both frequent pain and occasional pain were significantly related to reduced emotional well-being (respectively d = 1.40 and d = .68). Both frequent and occasional pain were significantly

TABLE 2 Estimated means and standard deviations of grades, attention, and emotional problems

	Pain-No	**Pain- Occasionally**		**Pain-Frequently**	
	mean (sd)	mean (sd)	F(df) p	mean (sd)	F(df) p
Dutch	6.80 (1.05)	6.68 (0.99)	$2.59_{(1;1637)}$ 0.11	6.56 (1.11)	$5.55_{(1;1636)}$ 0.02
English	7.08 (1.31)	6.99 (1.36)	$0.39_{(1;1637)}$ 0.53	6.87 (1.37)	$1.80_{(1;1634)}$ 0.18
Math	7.01 (1.19)	6.81 (1.11)	$3.44_{(1;1636)}$ 0.06	6.67 (1.24)	$6.29_{(1;1635)}$ 0.01
Attention problems	10.63 (2.81)	11.60 (2.68)	$237.5_{(1;1995)}$<0.001	12.92 (2.80)	$259.9_{(1;1988)}$<0.001
Emotional problems	11.96 (2.54)	14.54 (2.96)	$44.6_{(1;1995)}$<0.001	17.47 (3.30)	$80.9_{(1;1984)}$<0.001

TABLE 3 Multilevel models for grades

	Dutch		**English**		**Math**	
	Estimate	p-value	Estimate	p-value	Estimate	p-value
Occasional pain	-0.232	0.11	-0.122	0.80	1.039	0.03
Frequent pain	-0.079	0.02	-0.033	0.53	-0.094	0.06
Age	-0.221	0.69	-0.134	0.18	-0.242	0.01
Girls	-0.014	<0.001	-0.029	0.44	-0.107	0.003
Education level	0.069	<0.001	0.105	<0.001	0.097	<0.001
Western-minorities[a]	-0.086	0.25	0.060	0.45	0.020	0.79
Non-Western minorities[a]	-0.055	0.54	0.123	0.20	-0.080	0.39

[a] Reference category Dutch

TABLE 4 Mulitlevel models emotional well-being and attention problems

	Attention problems		**Emotional problems**	
	Estimate	p-value	Estimate	p-value
Occasional pain	0.322	<0.001	0.675	<0.001
Frequent pain	0.860	<0.001	1.401	<0.001
Age	-0.004	0.92	-0.027	0.40
Girls	-0.145	0.001	-0.072	0.07
Education level	-0.060	<0.001	-0.008	0.53
Western-minorities[a]	0.073	0.29	0.039	0.56
Non-Western minorities[a]	0.090	0.16	0.178	0.03

[a] Reference category Dutch

related to attention problems (respectively $d = .81$ and $d = .32$). Estimated means and standard deviations are reported in table 2. Multi-level models for emotional well-being and attention problems are presented in table 4.

In the present study, the internal consistency of both the Attention subscale of the AEFI and the Emotional Well-being subscale of the MCQCA was acceptable (respectively $\alpha = .72$ and $\alpha = .72$).

Mediation

Two significant pain-performance relations were observed, namely frequent pain with Dutch and math grades. The association between frequent pain and Dutch and math grades is partially mediated by reported attention problems (table 5, respectively 56% explained and 27% explained). Additionally, the association between frequent pain and Dutch and Math grades is for a large part mediated by emotional well-being (94% explained and 94% explained).

TABLE 5 Mediation of emotional and attentional problems between pain and grades

	Dutch			**Math**		
	Sobel statistic	*p*-value	% of the total effect that is mediated	Sobel statistic	*p*-value	% of the total effect that is mediated
Attention problems	-4.86	<.001	56%	-2.93	.003	27%
Emotional problems	-5.28	<.001	94%	-5.60	<.001	92%

DISCUSSION

Our results suggest that pain in early adolescence is associated with poorer Dutch and math grades, reduced emotional well-being, and increased attention problems. The effect sizes for the relationship between pain and the different grades are quite small. Furthermore, the association between frequent pain and poorer grades seems partially mediated by self-reported attention and for a large part by self-reported emotional well-being. In other words, adolescents with pain might only develop problems in academic performance when emotional problems and/or attention problems exist. Our findings link to previous studies that found pain to interfere with school success [99].

An important strength of this epidemiological study is its large sample size and the fact that the sample is homogeneous with respect to grade and age. This increases the reliability and generalizability of the results. Another strength is that the present study was the first to examine self-reported attention in adolescents with pain.

A limitation of the present study is that pain was measured only as a single item. In the present study we do not have any information about the duration, frequency, cause, and consequences of pain. We cannot make inferences about the severity of the pain experienced by adolescents. A second limitation is that grades might not be the best index for school performance. It might be that adolescents with pain do not only have poorer grades, but are also at a lower educational level than could be expected, based on their intelligence.

The results of the present study stress the importance of highlighting pain in the adolescent population at an early stage. With early warning signs, problems in academic performance might be prevented in adolescents with pain. Furthermore, the results can also be used to develop appropriate psycho-educational techniques in order to help these adolescents to cope with chronic pain and related emotional problems. Psycho-educational techniques should be aimed at improving executive functions (e.g. planning and problem solving), by including cognitive coping strategies and strategies to improve attention regulation [66]. According to a multidimensional model for stress reactions in adolescents there are three types of voluntary coping: primary control, secondary control, and disengagement [29]. Primary control is defined as attempts to alter one's emotions or the stressor itself. Secondary control coping is characterized by modifying cognitions or regulating attention. Disengagement coping is defined as removing oneself from the stressor or the emotions related to the stressor. Disengagement coping is associated with higher levels of pain and primary and secondary control are found to be associated with lower levels of pain [28,164]. Since the development of primary and secondary control coping is largely influenced by higher order executive functions [27], psycho-educational techniques might be aimed at improving executive functions, to improve coping and attention regulation.

While the present study constitutes a first step in understanding the relationship between pain and the academic performance of adolescents with pain, future research should examine the relationship between pain and academic performance in a longitudinal design or in a controlled intervention study, which makes it possible to draw inferences about cause and effect. In a controlled intervention study it could be examined whether the reduction of subjective pain in adolescents may also lead to improved emotional well-being and improved academic performance. It is possible that adolescents present their psychological problems in terms of a physiological dysfunction (i.e. pain). In this case, pain would be a proxy for decreased well-being. Additionally, future research should include the relationship between pain and underachievement in adolescents.

CHAPTER 4

Long-term follow-up of home-based behavioral management training provided by migraine patients

Voerman, J.S., De Klerk, C., Mérelle, S.Y.M., Aartsen, E., Timman, R., Sorbi, M., Passchier, J. (2014) Long-term follow-up of home based behavioural management training provided by migraine patients. Cephalalgia, 34, 357-64.

ABSTRACT

Background

Behavioural migraine approaches are effective in reducing headache attacks in migraine. Availability of treatment might be increased by using migraine patients as trainers. Therefore, Mérelle and colleagues developed and evaluated a home-based behavioural management training (BMT) delivered by migraine lay trainers [118]. The maintenance of effects at two to four years follow-up is studied in the present study.

Method

Measurements were taken pre-BMT (T0), post-BMT (T1), at 6-month follow-up (T2), and at long-term follow-up, i.e. two to four years after BMT (T3). Data of 127 participants was analyzed with longitudinal multi-level analyses.

Results

Short-term improvements in attack frequency and self-efficacy in managing headache episodes post-BMT were maintained at long-term follow-up ($d_{T0\text{-}T3}$=-.34 and $d_{T0\text{-}T3}$=.69 respectively). The level of headache specific internal control which increased during BMT decreased from post-BMT to long-term follow-up ($d_{T0\text{-}T3}$ =.18). Quality of life and migraine-related disability improved gradually over time ($d_{T0\text{-}T3}$ =.45 and $d_{T0\text{-}T3}$ = -.26 respectively).

Conclusions

Although the results should be interpreted with caution due to the lack of a follow-up control group, the findings of this study suggest that behavioural management training for migraine is beneficial at the long-term.

INTRODUCTION

Migraine is a chronic neurovascular disease, which is characterized by severe headache attacks [48,55]. Therefore, migraine interferes with work activities and social life [69,72]. Since the 1970's behavioral migraine interventions have been developed [141]. These interventions are targeted at increasing participants' perceived control over migraine, reducing the frequency and intensity of headache attacks, and decreasing migraine-related disability [70]. Behavioral migraine approaches, such as relaxation training and cognitive-behavioral techniques, are effective in reducing headache attacks [111,131,141,159]. Minimal contact versions of these approaches have proven to be equally effective as vis-à-vis treatments [58,145]. An important advantage of minimal contact approaches is increased availability of therapy [3,31,145,163]. Availability might even be more increased by using migraine patients as trainers. Additionally, lay trainers may decrease drop-out by being more motivating [2]. For these reasons Mérelle and colleagues developed and evaluated a home-based behavioral management training (BMT) delivered by migraine lay trainers (MLTs) [116]. BMT was primarily aimed at reducing the migraine attack frequency and increasing internal control over and self-efficacy in attack prevention.

The short-term efficacy of BMT was examined in a randomized controlled trial [118]. The results showed that participants in the treatment group experienced 21% less migraine attacks immediately after the training. Migraine patients in the waitlist control (WLC) group experienced 6% less attacks. The difference between the treatment group and the WLC group did not reach significance ($p = .07$). However, participants in the treatment group did report significantly more self-efficacy in managing headache episodes and headache-specific internal control than participants in the WLC group. Moreover, effect sizes were large. No differences were found for quality of life and migraine-related disability. The WLC group received BMT directly after their waitlist period. In a 6-month follow-up study, Mérelle and colleagues found that attack frequency in the total group of participants significantly decreased from post-BMT to 6-month follow-up (-23%). Post-BMT improvements in headache specific internal control and self-efficacy in managing headache episodes were maintained [117]. Furthermore, quality of life improved from pre-BMT to 6-month follow-up.

To our knowledge no long-term follow-up studies on BMT delivered by migraine patients have been conducted yet. Therefore, the primary aim of the present study was to study if the changes in migraine frequency, self-efficacy in managing headache episodes, and headache specific internal control were maintained or improved from post-BMT to two to four years after BMT. The secondary aim of the present study was to study if the change in quality of life was maintained, or improved. Furthermore, we evaluated if any changes occurred over time in migraine-specific disability.

MATERIALS AND METHODS

Participants

At the time of inclusion [118], participants' age ranged from 18 to 66 years old (*M* age = 44). All patients met the International Headache Society criteria [73] for migraine with (G43.1) or without (G43.0) aura. Headache attack frequency had to be between one and six attacks per month. Exclusion criteria were a headache frequency of 15 days or more per month, migraine duration of less than a year, a score of 178 or higher on the Symptom Checklist-90 (SCL-90) [39], and migraine onset above age 50. Migraine onset at an older age can be indicative of an underlying organic disease. The major reason to set up this study was an optimal applicability of behavioral strategies. Therefore, we did not restrict the study to ideal participants of 2-4 attacks per month. Instead, we chose a broad range of patients that allowed us to generalize the findings to the general population and could enhance the external validity of the study. We choose to exclude high frequency to minimize the risk of including patients with medication-overuse headache.

Behavioral Management Training

The BMT protocol was based on an established protocol for individual behavioral treatment [113,155,156]. The two main strategies used to prevent migraine attacks were: 1) Detecting migraine triggers and premonitory symptoms (using registration assignments) 2) Using skills for preventive self-regulation, including autogenic relaxation, diaphragmatic breathing, cue-controlled self-relaxation, positive thinking, and time-management. Treatment consisted of seven small-group (three to four migraine patients) sessions at the trainer's home. The sessions had a duration of two hours each and were provided within a period of ten weeks. Session 1 to 3 took place once per week and focused on the acquisition of self-management skills regarding the early detection of triggers and premonitory symptoms and a stepwise training in self-relaxation. Session 4 to 6 took place every two weeks and focused on the application of these skills in daily life and in migraine-provoking conditions. In addition, participants developed a personalized action plan with lifestyle changes and self-regulation skills to be executed in case of a risk condition. The last session was used for evaluation and relapse prevention. In between the group meetings participants had to read some theoretical background information, to do their autogenic and breathing exercises, and to do their registration assignments.

Migraine Lay Trainers

MLTs received BMT themselves before becoming a trainer [116]. Participants that were able to manage their own migraine attacks and were motivated to become a trainer, provided BMT to one new patient under supervision of the third author. Next, the potential MLTs trained a small group of new patients under continued supervision. MLTs received an elaborate manual for every BMT session. MLTs briefly evaluated every session on a standard evaluation form, which was used for supervision after sessions 1,3, and 6. Finally, MLTs participated in three workshops on trainer skills and a workshop on relaxation training. MLTs were paid a limited fee of 20 euro's per BMT session, which also covered expenses of the BMT provision at their homes.

Measures

Headache Diary

Migraine frequency was measured with a prospective headache diary. The use of a headache diary is in accordance with the guidelines for clinical trials in prophylactic treatment and the guidelines for behavioral trials [131,160]. Participants recorded their headaches for four weeks per 24 hours. In case of a migraine attack pain intensity was rated per 6-hour unit and was scored on a three-point scale: 1 (*mild headache – I can do everything but with some difficulty*), 2 (*moderate headache – I cannot do all or most things*), and 3 (*severe headache – I am unable to do things, bed rest is necessary*). Participants were asked to register additional characteristics and associated symptoms once a day. The data from the headache diary were prepared in the same way as in the study of Mérelle and colleagues [118].

Headache Management Self-Efficacy Questionnaire

To assess participants' self-efficacy in managing headache episodes, the Headache Management Self-Efficacy Questionnaire (HMSE) [52] was used. The HMSE consists of 25 items. All items are evaluated on a seven-point scale (from 1 = *strongly disagree* to 7 = *strongly agree*). A total score (ranging from 25 to 175) has been calculated by summing all the items. A higher score indicates a higher level of self-efficacy. Mérelle and colleagues translated the HMSE into Dutch and found its reliability to be satisfactory [118].

Headache Specific Locus of Control Scale

The Headache Specific Locus of Control Scale (HSLC) [110] was used to measure perceived control over migraine attacks. The HSLC consists of 33 items and three subscales: internal locus of control (11 items), external control chance (11 items) and health care professional locus of control (11 items). The health care professional locus of control scale was not used in this study. Every item is scored on a five-point scale (from 1 = *strongly disagree* to 5 = *strongly agree*). For the HSLC the subscale scores were calculated (ranging from 11 to 55). The psychometric abilities of the HSLC are satisfactory [171]. Mérelle and colleagues translated the HSLC into Dutch. This Dutch version yielded good to excellent reliability [118].

Migraine Specific Quality of Life Questionnaire

The quality of life of the participants was measured by the Dutch version of the Migraine Specific Quality of Life Questionnaire (MSQOL) [72,115,176]. The MSQOL consists of 20 items, each of which is scored on a four-point scale. Scores can vary from 20 to 80, with lower scores indicating a lower quality of life. The Dutch version of the MSQOL has a good reliability and validity [72].

Migraine Disability Assessment Scale

The Migraine Disability Assessment Scale (MIDAS) [160] was used to asses disability related to migraine attacks in migraine patients. The total score reflects the number of days lost during the past three months

due to migraine in different settings (e.g., at work or at home). The reliability and validity of the MIDAS are acceptable [160].

Study design and Procedure

The present study is a follow-up study (i.e. two to four years after BMT) of the randomized controlled trial Mérelle and colleagues performed between July 2003 and March 2007 [117,118]. All participants of the original study [116] were invited to take part in this long-term follow-up study. Participants were informed about the follow-up study by an information letter, which was sent in November 2007 (two to four years post-BMT). This letter was sent by post, together with the headache diary and questionnaires. The study was approved by the Ethics Committee of the Erasmus MC, Rotterdam, The Netherlands.

As the WLC group received BMT directly after the waiting period we added their data to those of the original BMT group.

Data analysis

Multi-level modelling (SPSS 17, MIXED module) was applied for longitudinal analysis. This analysis allows handling incomplete data with a minimal loss of information, as incomplete cases can be included in the analyses. Results of the measurements pre-BMT (T0), post-BMT (T1, 10 weeks), at 6-month follow-up (T2), and at long-term follow-up (circa 3 years) are presented. Linear splines were used to test the differences in outcomes between time points [46]. In addition to a linear time effect, knots were postulated at post-BMT (10 weeks) and at 6-month follow-up (26 weeks), thereby respecting the differences in time span between the follow-ups. This procedure allows the estimated course of the dependent variable to bend at the intermediate time points (post-BMT and 6-months follow-up). Non-significant knots (p-out > 0.05) were deleted from the models until a final parsimonious model was reached that did not differ significantly from the original saturated model. This significance was determined with the deviance statistic [152]. Migraine-related disability was log-transformed; other dependent variables were root-transformed because they were not normally distributed. There was no transformation needed for the HSCL subscale for external control chance (subscale HSLC). For all differences effect sizes are reported, with $d = .2$ indicating a weak effect, $d = .5$ indicating a medium effect and $d = .8$ indicating a strong effect [25].

RESULTS

Patient characteristics

In the original study [118] 129 migraine patients were randomized (BMT: n = 61, WLC: n = 68). Two patients were excluded before the start of the study because of a diagnostic error. In the present study data of 127 patients were analyzed. Twenty-three patients did not start or withdrew from BMT. The characteristics of all patients at baseline and drop-outs at long-term follow-up are presented in Table 1. Patients who

TABLE 1. Demographic, psychological and migraine characteristics at baseline for all patients and dropouts at long-term follow-up

	Baseline	Retained at long-term follow-up	Dropouts at long-term follow-up	Difference retained/dropout p [a)]
N	127	77	50	
Female, n (%)	111 (87.4%)	67 (87.0%)	44 (88.0%)	1.00 [b)]
Education (%)				
low	7 (5.5%)	3 (3.9%)	4 (8.0%)	0.67 [c)]
middle	37 (29.1%)	23 (29.9%)	14 (28.0%)	
high	83 (65.4%)	51 (66.2%)	32 (64.0%%)	
Age, M (sd)	44.4 (9.9)	44.9 (9.9)	43.6 (10.1)	0.47
Primary outcomes				
self-efficacy	89.2 (23.3)	87.7 (23.3)	91.9 (23.6)	0.40
attack frequency	3.04 (1.76)	2.84 (1.73)	3.36 (1.76)	0.10
internal control	35.4 (10.3)	37.2 (10.0)	32.5 (10.4)	0.012
external control chance	35.0 (8.65)	34.9 (8.89)	35.3 (8.34)	0.79
Secondary outcomes				
quality of life	56.9 (9.1)	55.3 (8.91)	59.3 (8.94)	0.015
migraine-related disability	23.2 (18.1)	22.6 (18.1)	24.1 (18.3)	0.37

[a)] Independent samples t-test, unless otherwise indicated

[b)] Fisher's exact test

[c)] Chi square test

did not participate at long-term follow-up had significantly lower scores for headache specific internal control ($p = 0.01$) and higher scores for quality of life ($p < 0.02$) at baseline.

Changes in primary outcomes

Attack frequency and self-efficacy in managing headache episodes improved from pre-BMT to post-BMT ($d_{T0\text{-}T1} = -.37$ and $d_{T0\text{-}T1} = .80$) and were maintained at 6-month follow-up ($d_{T0\text{-}T2} = -.36$ and $d_{T0\text{-}T2} = .79$) and long-term follow-up ($d_{T0\text{-}T3} = -.34$ and $d_{T0\text{-}T3} = .69$). The level of perceived headache-specific internal control over migraine attacks increased during BMT ($d_{T0\text{-}T1} = .60$), but decreased from 6-month follow-up to long-term follow-up (.$d_{T2\text{-}T3} = -.38$). The gain in perceived headache specific internal control between pre-BMT and long-term follow-up, was nearly significant ($d_{T0\text{-}T3} = .18$, $p = 0.059$; not in table). External control chance decreased during BMT ($d_{T0\text{-}T1} = -.68$) and was maintained at long-term follow-up ($d_{T0\text{-}T3} = -.57$). Details about the mixed models and their regression weights are presented in Table 2. Estimated means, standard deviations, effect sizes, and p-values are presented in Table 3. Graphic displays of the results are presented in Figure 1.

Changes in secondary outcomes

Quality of life improved from pre-BMT to long-term follow-up ($d_{T0\text{-}T3} = .45$). Migraine-related disability also improved gradually over time ($d_{T0\text{-}T3} = -.26$). Estimated means, standard deviations, effect sizes, and

TABLE 2. Parsimonious mixed models of transformed primary and secondary outcomes

	Estimated regression weights (*p*)				Deviance [a)]
	Intercept	General slope	Knot T1	Knot T2	Chi^2 (df) *p*
Primary outcomes					
Attack frequency	2.21 (<.001)	-0.381 (<.001)	0.383 (<.001)		0,31 (1) 0.57
Self-efficacy	4.66 (<.001)	0.120 (<0.001)	-0.121 (<0.001)		2.30 (1) 0.13
Internal control	2.73 (<.001)	0.079 (<.001)	-0.83 (<.001)		1.63 (1) 0.20
External control chance	35.00 (<.001)	-0.591 (<.001)	0.598 (<.001)		0.53 (1) 0.47
Secondary outcomes					
Quality of life	7.52 (<.001)	0.0244 (<.001)		-0.022 (.001)	0.02 (1) 0.89
Migraine-related disability	3.14 (<.001)	-0.008 (<.001)		0.008 (.001)	2.05 (1) 0.15

[a)] -2 loglikelihood difference between complete and final model

TABLE 3. Estimated means and effect sizes of primary and secondary outcomes

	Estimated means				Cohen's *d*				
	T0 baseline	T1 end BMT	T2 6 months	T3 2-4 years	T0-T1	T0-T2	T0-T3	T1-T2	T2-T3
Primary outcomes									
Attack frequency	2.88	2.24	2.24	2.29	-0.37	-0.36	-0.34	0.00	0.03
Self-efficacy	90.4	109.2	108.9	106.6	0.80	0.79	0.69	-0.01	-0.10
Internal control	36.8	42.9	42.5	38.6	0.60	0.55	0.18	-0.04	-0.38
External control chance	35.0	29.1	29.2	30.0	-0.68	-0.67	-0.57	0.01	0.10
Secondary outcomes									
Quality of life	57.5	58.5	60.1	61.7	0.11	0.28	0.45	0.17	0.17
Migraine related disability	19.1	17.1	14.5	14.4	-0.10	-0.25	-0.26	-0.15	-0.01

p-values are presented in Table 3. Details about the mixed models and their regression weights are presented in Table 2. Graphic displays of the results are presented in Figure 1.

DISCUSSION AND CONCLUSIONS

The findings of this follow-up study seem promising. Most positive short-term changes in migraine-related outcomes were maintained at long-term follow-up, i.e. two to four years after BMT. The short-term improvements in attack frequency and self-efficacy in managing headache episodes post-BMT were maintained at 6-month follow-up and at long-term follow-up. Quality of life and migraine-related disability improved in the period from pre-BMT to long-term follow-up. Only headache-specific internal control decreased from post-BMT to long-term follow-up.

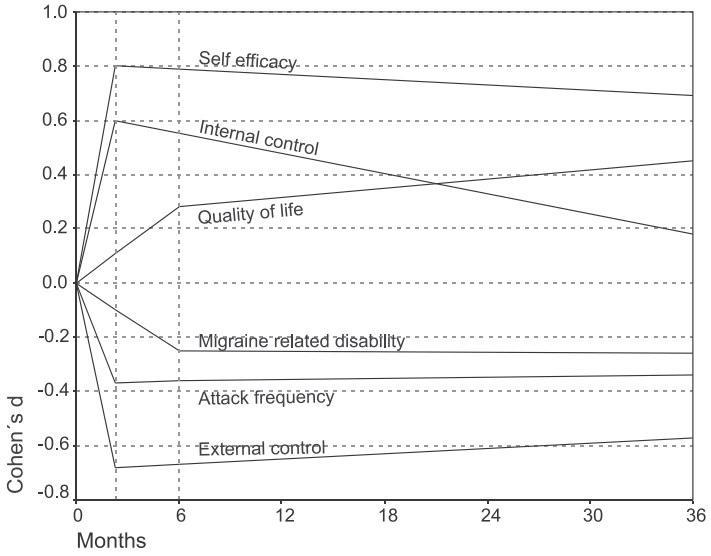

FIGURE 1 Graphic representation of effect sizes of primary and secondary outcomes

Results found at post-BMT and 6-month follow-up slightly differ from the results found by Mérelle and colleagues [117]. Mérelle and colleagues did not find any decreases in migraine-related disability [117] whereas in the present study migraine-related disability significantly decreased during BMT and 6-month follow-up and was maintained at the last follow-up. This difference in results has been caused by the use of different analyzing techniques, i.e. repeated measurements ANOVA versus longitudinal multi-level analysis. With repeated measurements ANOVA only complete cases are analyzed, whereas with longitudinal multi-level analysis all available data is analyzed.

It is an interesting finding that headache-specific internal control returned towards its original level, while the decrease in external control chance was mostly maintained at long-term follow-up. These results supports the notion that internal control and external control are independent constructs instead of being opposite ends of one continuum [104]. Someone can have internal and external beliefs at the same time. Furthermore, previous research suggests that internal control is not always associated with better health outcomes than external locus of control. It is also remarkable that headache specific internal control decreased at follow-up, while the short-term increase in self-efficacy in managing headaches was maintained at follow-up. Self-efficacy refers to the confidence in one's ability to use specific behavioral strategies to prevent and manage migraine attacks [52]. The results might thus imply that three to four years after treatment participants are still confident that they are able to perform the self-management behaviour that they learned during treatment (i.e. self-efficacy), but that they feel less optimistic about the overall degree in which migraines can be influenced by their own actions (i.e. internal control; see Seng and colleagues [149]).

The strength of the present follow-up study is the length of the follow-up period. Research has shown that traditional vis-à-vis behavioral therapies are effective at the long-term [154]. To the best of our knowledge no study has examined the maintenance of effects of a behavioral management training provided by migraine patients at two to four years follow-up. Another strong methodological point is that longitudinal multi-level analysis is used to analyze the data. Longitudinal multi-level analysis is less biased than repeated measurements ANOVA, for all participants of the original study are taken into account, including participants that dropped out at follow-up.

The most important limitation of the present study is that there was no control group anymore at follow-up, because it was unethical to withhold the treatment to the original waiting list controls. Therefore it is important to interpret the results with caution. The found improvements may represent effects of (non) pharmacological treatment received during the follow-up period. In the present study the use of other medications and/or behavioural treatments was not monitored. Additionally, spontaneous recovery might have been occurred, for on average migraine decreases with aging [86]. Furthermore, it is not known if participants still used the behavioural and cognitive strategies they were taught during the BMT intervention. Therefore, no firm conclusions about causality can be drawn from this study. Another important limitation is that the follow-up time was not the same for all participants. Some participants filled out the questionnaires two years after BMT, others filled out the questionnaires four years after BMT. Furthermore, the level of headache specific internal control and quality of life significantly differed for patients who did and patients who did not participate in the long-term follow-up study. Dropouts who did not remain in the study had less headache specific internal control and a higher quality of life at baseline. These differences have resulted in slightly different short-time results reported by Mérelle et al. [116] who had conducted complete cases analyses.

In conclusion, BMT seems to improve the self-management skills of migraine patients, even at long-term follow-up. If the cost-effectiveness has been proven, BMT by lay trainers could be implemented at large scale. This would make treatment for migraine patients more available. However, problems with finding suitable lay trainers may be a threat to large-scale implementation (17, 18). Our results suggest that self-efficacy is an important component of self-management. Therefore, newly to develop therapies for patients with migraine should include strategies to enhance self-efficacy. Future research should compare BMT delivered by lay trainers and BMT delivered by a professional. Additionally, it will be interesting to know if booster interventions using, for example Internet or text messages, intensify the long-term benefits of BMT. Also, future follow-up studies should include qualitative interviews to find out if migraine patients that have participated in BMT keep using learned strategies.

CHAPTER 5

Early signaling, referral, and treatment of adolescent chronic pain: a study protocol

Voerman, J.S., Remerie, S., De Graaf, L.E., Van de Looij-Jansen, P.M, Westendorp, T., I., Van Elderen, De Waart, F.G., Passchier, J., Dommisse-van Berkel, A.A.M., De Klerk, C. (2012) Early signaling, referral, and treatment of adolescent chronic pain: A study protocol. BMC Pediatrics, 12:66.

ABSTRACT

Background

Chronic pain is prevalent among young people and negatively influences their quality of life. Furthermore, chronic pain in adolescence may persist into adulthood. Therefore, it is important early on to promote the self-management skills of adolescents with chronic pain by improving signaling, referral, and treatment of these youngsters. In this study protocol we describe the designs of two complementary studies: a signaling study and an intervention study.

Methods and design

The signaling study evaluates the Pain Barometer, a self-assessed signaling instrument for chronic pain in adolescents. To evaluate the feasibility of the Pain Barometer, the experiences of youth-health care nurses will be evaluated in semi-structured interviews. Also, we will explore the frequencies of referral per health-care provider. The intervention study evaluates Move It Now, a guided self-help intervention via the Internet for teenagers with chronic pain. This intervention uses cognitive behavioural techniques, including relaxation exercises and positive thinking. The objective of the intervention is to improve the ability of adolescents to cope with pain. The efficacy of Move It Now will be examined in a randomized controlled trial, in which 60 adolescents will be randomly assigned to an experimental condition or a waiting list control condition.

Discussion

If the Pain Barometer is proven to be feasible and Move It Now appears to be efficacious, a health care pathway can be created to provide the best tailored treatment promptly to adolescents with chronic pain. Move It Now can be easily implemented throughout the Netherlands, as the intervention is Internet based.

Trial registration

Dutch Trial Register NTR1926.

INTRODUCTION

In 1986 the International Association for the Study of Pain (IASP) formulated the following definition of pain: 'Pain is an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage. Note: pain is always subjective. Each individual learns the application of the word through experiences related to injury in early life'. Most child studies define chronic pain as pain that persists continuously or intermittently for at least three months [105,106,133,136,144,147]. In the general population approximately 25% of children and adolescents (0-18 years) suffer from chronic pain [4]. Limb pain, headache, and abdominal pain are the most reported types of pain. A prevalent combination is headache plus abdominal pain [60]. The prevalence of chronic pain is the highest in girls aged 12 to 14 years [4]. In 30% of the children and adolescents pain seems to persist after two years [136]. Additionally, 59% of the women and 39% of the men who reported pain in childhood also reported pain in early adulthood [16].

Chronic pain negatively influences the quality of life of adolescents [72]. Children and adolescents with pain are often unable to meet friends and to pursue hobbies [60]. Additionally, they may report disturbed sleep, loss of appetite, frequent use of medication, subclinical depression, and school absence. School absenteeism is also burdensome for parents, who often have to take days off from work [50]. Chronic pain also significantly impacts on the health care system [135]. Approximately 43% of children and adolescents with chronic pain utilize health care and 53% use pain medication at least once every three months. The frequency of health care use is positively related to age, pain intensity, and pain duration [144].

Some studies suggest that parents influence the way children and adolescents experience pain. Pain seems to be less intense when parents distract their child from pain, while pain appears to be worse when parents focus on the child's pain [179]. Additionally, parents of children and adolescents with chronic pain often have chronic pain themselves [90]. Pain in adolescents also seems to be predicted by depressive symptoms, somatic complaints (other than pain), reduced leisure time activities, number of friends, and recent parental divorce [89]. Additionally, chronic pain in adolescents is related to coping [24]. Adolescents with an avoidant coping style and adolescents with a dependent coping style, report the most disabled functioning. They have higher levels of depression and anxiety than other adolescents with chronic pain.

In summary, chronic pain in adolescents is influenced by many factors. This is in accordance with the biopsychosocial model [167], which states that pain is caused by a complex interaction between biological (e.g., genetics), psychological (e.g., attention), and social variables (e.g., role models). Differences in the interaction between these variables may explain differences in pain expression between pain patients. For this reason, different types of treatment, i.e. medical and psychological, might be useful for adolescents with chronic pain.

Signaling and referral

To optimally treat adolescents with chronic pain it is important to signal pain complaints at an early stage. The Pain Questionnaire was the first instrument that was developed for indexing pain and pain

parameters in children and adolescents, irrespective of pain localization [133]. The questionnaire consists of questions about the experience, location, frequency, duration, and intensity of pain. The Pain Questionnaire was designed for usage in the general population but is not considered suitable as a signaling instrument as it does not allow for the impact of pain. This means that currently chronic pain is not taken into account in the regular signaling and registration practice of the Preventive Youth Health Care in the Netherlands. To signal chronic pain in the general population a short signaling questionnaire should be developed. Additionally, a referral protocol is needed to offer the most tailored treatment to adolescents with chronic pain.

Psychological treatment

A systematic review of randomized controlled trials (RCTs) has shown that cognitive-behavioral therapy (CBT) and relaxation exercises are effective in reducing pain frequency and pain intensity in children and adolescents [44]. CBT may be effective in providing strategies that help adolescents to cope with their pain. As a result, pain-related symptoms of anxiety and depression may also be reduced and quality of life may improve. However, only few studies have examined if psychological treatment also enhances adolescents' well-being [64,165]. The results of a study by Trautmann and colleagues [165] showed that their self-help training program improved pain catastrophizing, but not depression and quality of life.

More studies on the effect of psychological treatment on coping and well-being are needed. Unfortunately, inclusion is often problematic in face-to-face cognitive behavioral therapy [19]. Adolescents have difficulty combining vis-à-vis therapy sessions with homework and activities with friends. Additionally, youngsters with bodily symptoms are often reluctant to consult a psychologist. Psychological interventions using an Internet format could therefore be more attractive to adolescents. Advantages of internet therapy are that it is available for all geographical districts and adolescents can work through the program at any place and any time.

Internet interventions

Stinson and colleagues concluded in their review that Internet-based self-management interventions seemed to improve health outcomes in children and adolescents with various health conditions, including pain [161]. In 2005, Hicks and colleagues tested the efficacy of an Internet-supported treatment for adolescents with chronic pain in the general population [64]. One month after treatment the adolescents in the experimental group experienced significantly reduced pain compared to adolescents in the waiting list group. More recently, Palermo and colleagues (2009) evaluated an Internet-delivered family CBT intervention [128]. Results showed that pain-related disability and pain intensity were reduced more in the Internet group than in the waiting list group. These effects were maintained at three months follow-up. In summary, these findings suggest that Internet interventions may be a promising alternative to face-to-face therapy in teenagers with chronic pain.

Current study

In the current manuscript we describe the study protocols of two related studies: a referral study and an intervention study. The main aim of these studies is to improve signaling, referral, and treatment of adolescents with chronic pain. By collaborating with various (mental) health-care institutions, a health-care pathway can be created, through which adolescents can receive the best tailored treatment.

Firstly, a signaling instrument for chronic pain was developed. The instrument includes a referral protocol for implementation in the preventive health examinations of the Public Health Services Rotterdam. Secondly, a guided self-help intervention via the Internet was developed for adolescents with chronic pain. The intervention includes cognitive behavioural techniques and relaxation exercises and is one of the referral options in the referral protocol. The primary objective of this intervention is to improve the way adolescents cope with pain.

Our main research questions are: 1. What is the feasibility of the signaling instrument and the accompanying referral protocol in the context of Preventive Youth Health Care? 2. What is the efficacy of the guided self-help intervention via the Internet?

METHODS REFERRAL STUDY

Population

Each year, the Municipal Public Health Services in Rotterdam preventively examine health conditions and developmental problems in approximately 12.000 first-grade adolescents from several secondary schools in the Rotterdam area. The signaling and referral instrument developed for this study will be part of this preventive examination. The instrument was filled out by approximately 6.000 adolescents in the period between October 2008 and July 2011. The Medical Ethical Committee of Erasmus MC University Medical Hospital, Rotterdam, The Netherlands approved the study protocol (MEC-2009-195).

The Pain Barometer

The Pain Barometer is a self-assessed signaling and referral instrument for chronic pain in adolescents aged 12 to 17 years. The instrument is based on the Pain Questionnaire [133], literature research, and expert knowledge (e.g., a medical doctor and researcher from Rijndam rehabilitation centre, psychologists from the Erasmus MC University Medical Hospital and researchers, nurses, and a medical doctor from the Preventive Youth Health Care in Rotterdam). The Pain Barometer consists of 12 questions about several characteristics of pain (e.g., duration, localization, frequency, intensity, and consequences). Chronic pain is defined as pain that lasts for at least three months, recurrently or continuously.

Also part of the Pain Barometer is a referral protocol that states several referral options and their criteria for adolescents with chronic pain. The referral options are presented in Table 1. Additionally, adaptations were made to a digital dossier, e.g. Electronic Child Dossier (ECD), to enable registration. The newly developed registration categories are presented in Table 2.

TABLE 1. The Pain Barometer referral protocol

Referral option	Criteria
No referral for pain	- the adolescent does not experience the pain as a problem or - other problems are more urgent
General practitioner	- the adolescent and/or the parents worry about a medical cause of the pain or - the youth nurse suspects a medical cause of the pain
School doctor	- the adolescent is absent from school for more than two days per month or - the adolescent regularly misses gym lessons at school or - the school nurse is not sure if it is necessary to refer to the general practitioner
Move It Now	- the adolescent is aged 12 to 17 years old and - there is no apparent medical cause of pain according to the general practitioner and - there are no serious psychosocial problems and - the adolescent withdraws from activities or becomes overburdened
Youth rehabilitation	- there is no apparent medical cause of pain and - the adolescents' pain is located in the limbs or back and - the adolescent experiences pain as a big problem and - Move It Now does not seem to be sufficient

TABLE 2. The Pain Barometer registration system

Registration categories	Answer categories
Conversation details	- Free text
Chronic pain, but other problems more important	- Yes - No
Referral to the school doctor	- Yes - No
Referral to the GP	- Yes - No
Referral to Youth rehabilitation	- Yes - No
Referral to Move It Now	- Yes - No

Procedure

When the adolescent has filled out the Pain Barometer, a computer calculates whether he or she has chronic pain. Adolescents who are in chronic pain discuss their pain complaints with a youth health-care nurse during the standard consultation procedure with the Public Health Services. The youth health-care nurse uses the referral protocol to determine which treatment option is the best. He or she registers details about the consultation and the referral in the ECD.

To evaluate the feasibility of the Pain Barometer and the referral protocol, the experiences of the youth health-care nurses are evaluated in semi-structured interviews by telephone. Examples of questions are 'Can you describe the adolescents you referred to the school doctor, General Practitioner (GP), rehabilitation center, or Move It Now?' and 'Were the referral options clear?'

Qualitative analyses

The interviews will be audio-recorded and transcribed. The transcripts will be analyzed using the program ATLAS.ti [122]. First, unique themes will be identified. Second, the unique themes will be clustered according to main themes.

Quantitative analyses

The ECD will be used to determine the frequencies of referral per health service, i.e. school doctor, GP, rehabilitation centre, or Move It Now.

METHODS INTERVENTION STUDY

Population

The study sample will consist of 60 adolescents. Adolescents are eligible to participate if they meet the following criteria: age 12 to 17 years; chronic pain for at least three months; abdominal pain, headache, back pain or limb pain; no apparent medical cause of pain (an exception is made for adolescents with migraine); withdrawing from activities or feeling overburdened; access to the Internet at home; fluent in the Dutch language; no current psychological treatment for pain; no severe psychosocial problems.

Recruitment of participants takes place via the referral study, the Internet, the media, and rehabilitation centers and hospitals in the Netherlands. The study started April 2009. The inclusion of patients will end in June 2012. The three-month follow-up period will be finished in September 2012.

Intervention

Move It Now is a translated and adapted version of the intervention program developed by Hicks and colleagues [64]. It is a guided self-help intervention via Internet for adolescents aged 12 to 17 years with chronic pain. The objective of the intervention is to improve the way adolescents cope with pain. The intervention is based on the principles of cognitive-behavioral therapy. Adolescents independently work through seven online modules. The modules aim to teach important life skills, such as problem solving and relaxation. Additionally, the adolescents learn to change dysfunctional thoughts and beliefs. Other topics include deep breathing and improving self-esteem and interpersonal relationships. All topics in the modules are presented in Table 3. Additionally, a therapist from the Rijndam rehabilitation centre contacts the participants by e-mail or telephone each week. This therapeutic contact consists of emotional support and a discussion about the online modules and exercises. Parents work through two online modules and have contact with the therapist three times (at the beginning, in the middle, and at the end of the intervention). In the parent modules, information is given about how parents should handle the pain of their child. Parents are strongly advised to encourage their child to complete Move It Now.

TABLE 3. Overview of Move It Now modules

Module number	Module subject	Module contents
1	Background and goals	Psycho-education about pain and its consequences / treatment goals
2	Pain killers and breathing	Strategies to relieve pain are discussed / deep breathing.
3	Relaxation	Advantages of relaxation / two relaxation exercises, one for the whole body and one in which fantasy is used
4	Thinking and feeling	Strategies to reduce negative thinking patterns
5	Helpful thinking	Reasoning errors / by helpful thoughts.
6	Staying active	Staying active physically (e.g., sports) and socially (e.g., going out with friends)/ a short relaxation exercise, which can be used to relax rather quickly
7	Making a plan	Develop a plan about what to do when in pain at home or at school

Much attention has been given to the design and lay-out of the Move It Now website. For example, the adolescents are led through the online chapters by an animated female guide using a voice-over. Furthermore, several interactive elements were developed to lead the adolescents to the material that fits their response, i.e. tailoring. For example, if the adolescent reports having headaches, only information about headaches will appear.

Instruments

The measures in the intervention study are in accordance with the recommendations of the Pediatric Initiative on Methods, Measurement and Pain Assessment in Clinical Trials (PedIMMPACT) [114].

Demographic data

Demographic data collected includes the adolescents' gender, date of birth, ethnicity, and educational level and was gathered using a self-assessed questionnaire.

The Pain Diary

The participating adolescents keep a daily diary for one week. Pain intensity is measured by several multiple choice questions and a Visual Analogue Scale (VAS), with *no pain* and *worst pain ever* at the respective ends. Prior research has shown that the reliability and validity of this scale is high [76].

Pain Coping Questionnaire

Pain coping is measured by the Pain Coping Questionnaire (PCQ) [142]. The PCQ consists of 39 items, which are evaluated on a five point scale (with 1 = *never* and 5 = *very often*). The PCQ has three dimensions: approach, problem-focused avoidance, and emotion-focused avoidance and eight subscales: information seeking, problem solving, seeking social support, positive self-statements, behavioral distraction,

cognitive distraction, externalizing, and internalizing / catastrophizing. Higher scores indicate greater use of the coping strategy. The Dutch version of the PCQ has good psychometric properties [7].

Pediatric Migraine Disability Assessment Scale

Pain-related disability is measured with an adapted version of the Pediatric Migraine Disability Assessment Scale (PedMIDAS) [62,182]. The PedMIDAS consists of 6 items. Participants report the number of days a specific aspect of functioning was impaired in the previous three months due to pain. A higher score on the PedMIDAS is indicative of more pain-related disability. The reliability and validity of the PedMIDAS are acceptable [62].

Sleep

The following self-assessed questions are used to measure sleeping problems: Do you have sleeping problems?, What do you think causes your sleeping problems?, and How many hours of sleep do you get in a period of 24 hours?.

Child Health Questionnaire

The Child Health Questionnaire (CHQ) [85] is used to measure physical and psychosocial wellbeing of adolescents. The CHQ consist of 87 items, divided over 10 multi-item subscales and two single item questions: physical functioning, role functioning emotional, role functioning behavioral, role functioning physical, bodily pain, general behavior, mental health, self-esteem, general health perceptions, change in health, family activities, and family cohesion. Higher scores indicate better functioning and well-being. The psychometric properties of the Dutch version of the CHQ are good, even when completed via the Internet [139,140].

EQ-5D-5L developed by the EuroQol Group

The EQ-5D-5L developed by the EuroQol Group is also used to measure adolescents' health-related quality of life. The EQ-5D-5L consists of 5 dimensions: mobility, self-care, daily activities, pain/discomfort, and depression/anxiety. Each dimension comprises 5 levels: no problems, slight problems, moderate problems, major problems, and extreme problems. A higher score on the EQ-5D-5L dimensions is indicative of a worse quality of life. The psychometric qualities of the 5 level version of the EQ-5D seem promising [61].

Illness Behaviour Encouragement Scale

The Illness Behaviour Encouragement Scale (IBES) [180] measures the rewarding of pain behavior by parents. The IBES consists of 12 items, which are evaluated on a five point scale (with 0 = *never* and 4 = *always*). The items of the IBES comprise two subscales: rewarding by parents in a pain situation and rewarding by parents in a pain-free situation. Higher scores indicate more rewarding of pain by parents. The reliability and validity of the IBES is satisfactory [11].

Pain Catastrophizing Scale – Child version

Pain catastrophizing is measured by the Pain Catastrophizing Scale Child version (PCS-C) [32], which consists of 13 items. Items are scored on a five point scale (with 0 = *not at all* and 4 = *extremely*). Higher scores indicate higher levels of pain catastrophizing. Research has shown that the Dutch version of the PCS-C is reliable and has satisfactory predictive validity [32,112,173].

Design

The intervention study is a randomized controlled trial. Participants are automatically randomly assigned to the experimental group or the waiting list group by the Move It Now website. The Move It Now website uses a randomization list which has been generated by an independent statistician. Block randomization has been used to keep the group sizes equal. Prior to entry of the participants into the study, both the researchers and the therapist are blinded for randomization. Figure 1 shows the design and the anticipated flow of participants.

Adolescents in the waiting list group wait seven weeks before they are allowed to start with the intervention. Adolescents in the experimental group fill out the same online questionnaires and the online Pain Diary three times in total: at baseline, directly after the intervention, and at three months follow-up. Adolescents in the waiting list group fill out the same online questionnaires and the online Pain Diary four times in total: at baseline, after the waiting period of seven weeks, directly after the intervention, and at three months follow-up. The Medical Ethical Committee of Erasmus MC University Medical Hospital, Rotterdam, The Netherlands has approved the study protocol (MEC-2009-195). The study has been registered at the Netherlands Trial Register, which is part of the Dutch Cochrane Centre (NTRcode 1926).

Procedure

Adolescents are informed about Move It Now by their youth health-care nurse or their medical specialist. When an adolescent decides to participate in Move It Now, he or she contacts the researcher by mail or e-mail. Thereafter, the researcher contacts the adolescent and one of the parents by telephone. In this phone call, the researcher checks the inclusion criteria, gives information, and asks the adolescent and the parent to send their signed informed consent forms.

When the researcher has received the signed informed consent forms, the adolescent and the parent are registered at the Move It Now website. An automatic e-mail with their login codes is sent to them. When the adolescent logs into the website, he or she first fills out the online questionnaires; the PedMIDAS, the self-constructed sleep questionnaire, the CHQ, the PCQ, the IBES, and the PCS-C. For logistic reasons, the EQ-5D-5L and a separate reply form are sent to the adolescent by e-mail. Additionally, the adolescent keeps an online Pain Dairy for one week. After this baseline measurement, the adolescent and the parent receive an automatic e-mail that informs them whether they can start with Move It Now directly in the intervention group or if they have to wait for seven weeks on the waiting list control group.

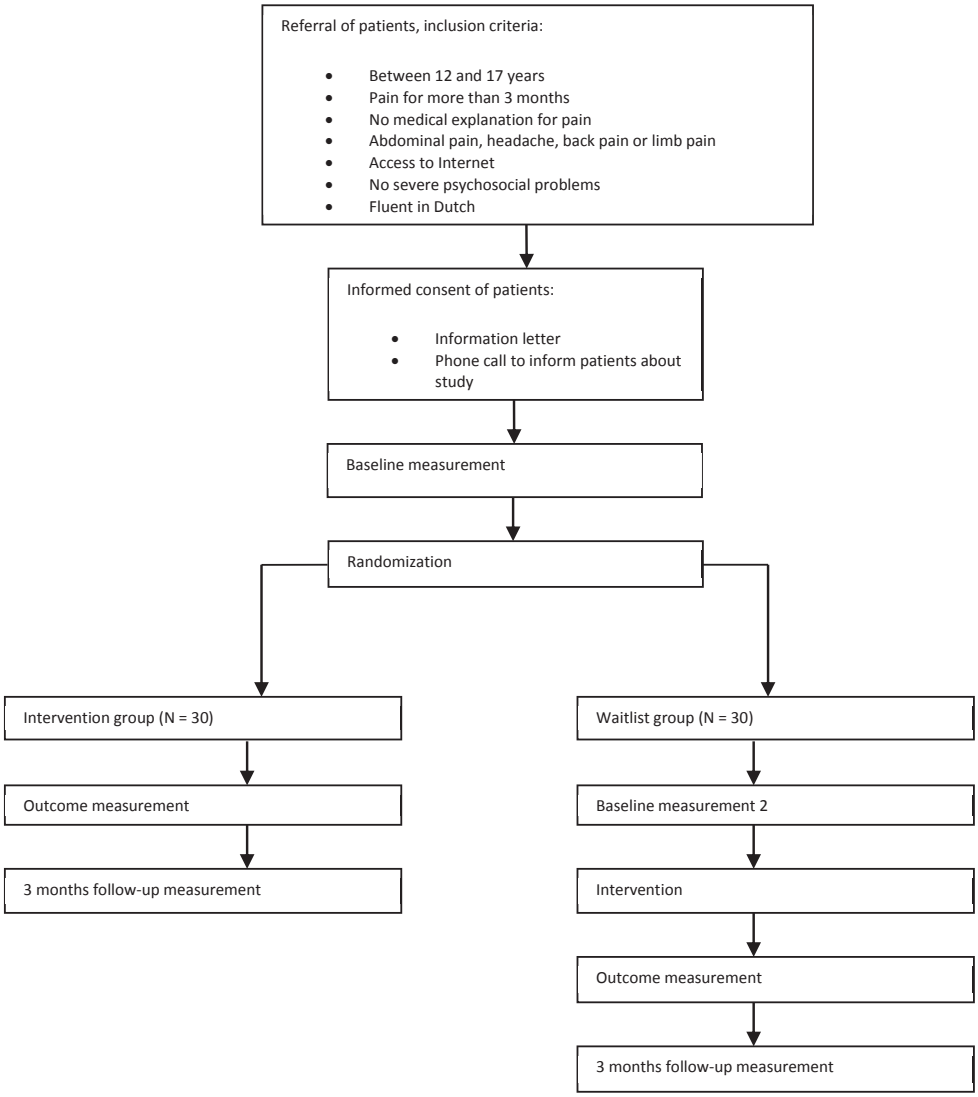

FIGURE 1. Design of the randomized trial

Statistical analyses

Power calculations have shown that for an effect of d = .70 on the questionnaires, a sample size of 25 participants per group is needed (power 80%, alpha 0.05). Adjusting for potential study withdrawal (20%), we estimate that 60 participants should be included.

Multi-level modelling will be applied for longitudinal analysis. This analysis allows handling incomplete data with a minimal loss of information. Results of the measurements at baseline, post-treatment,

and at three months follow-up will be presented. Linear splines will be used to test the differences in outcomes between time points. For all differences, effect sizes will be reported, with *d* = .2-.5 indicating a weak effect, *d* = .5-.8 indicating a medium effect and *d* = >.8 indicating a strong effect [25].

The EQ-5D-5L, in combination with the CHQ [85], will be used for the economic evaluation of Move It Now. The results will be expressed as cost per Quality Adjusted Life Year (QALY).

DISCUSSION

The primary aim of our study is to improve signaling, referral, and treatment of adolescents with chronic pain. In the referral study we will explore if the Pain Barometer, the signaling and referral instrument, is feasible in the context of preventive Youth Health-Care. In the intervention study we will explore if Move It Now, the guided self-help program via Internet, is efficacious.

Our study is the first in developing an instrument to signal chronic pain in adolescents that directly can be applied in practice. We expect the Pain Barometer to be a feasible instrument. A limitation of the referral study is that it is not possible to explore the validity of the Pain Barometer in this setting.

Move It Now is one the first tailored and guided self-help interventions via the Internet for adolescents with chronic pain. We expect the participants to cope with their pain more actively directly after the Move It Now intervention. We also hypothesize that Move It Now reduces pain frequency, pain duration, pain intensity, and pain disability. A strength of the intervention study is that it includes important but often neglected, outcome variables, i.e. coping with pain and quality of life. Additionally, the intervention is not only directed at the adolescents, but also at the parents. A limitation of the intervention study is that there will be no control group at follow-up, since the intervention is offered to the adolescents after a short waiting period. Furthermore, a waiting list group is used as a control. Therefore it will not be clear if the specific or nonspecific factors, such as attention, influence the intervention.

If the Pain Barometer appears to be feasible, it can be used to signal and refer adolescents with chronic pain at an early stage in various health care settings. If Move It Now is proved to be efficacious it can be easily implemented throughout the Netherlands, as the intervention is internet based. Then it will also be possible to perform a large scale (cost-)effectiveness study to examine the effect of Move It Now in daily practice. Additionally, future research should examine which elements are working for which adolescents. The Move It Now intervention could also easily be adapted to suit the needs of adolescents with other chronic health issues.

CHAPTER 6

Effects of a guided Internet-delivered self-help intervention for adolescents with chronic pain

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Busschbach, J.J.V., Passchier, J., De Klerk, C.
Effects of an Internet-based self-management intervention for adolescents with chronic pain (submitted).

ABSTRACT

Background

Cognitive behavioral therapy (CBT) is effective in reducing the frequency and intensity of chronic pain in adolescents. However, CBT seems not to be considered acceptable by all adolescents. The main aim of our study was therefore to evaluate the effects of a guided Internet-delivered self-help for adolescents with chronic pain.

Method

Adolescents (N=69) were assessed on the outcome measures: pain, coping, disability, catastrophizing, rewarding of pain behavior by parents, and quality of life. Measures were taken seven weeks before treatment, pre-treatment, post-treatment, and at three months follow-up. Multi-level modelling was used for longitudinal analysis of the data.

Results

Pain intensity, interference caused by pain, rewarding of pain behavior by parents, and sleep problems significantly decreased during the intervention. The quality of life scores for pain, general behaviour, mental health, family activities, and health changes did also significantly improve during the intervention. Concerning coping, only problem-focused avoidance behaviour significantly increased. No significant differences were found for pain-related disability and pain catastrophizing.

Conclusions

Contrary to expectations, a guided Internet-delivered self-help for chronic pain is difficult to employ in adolescents, as it suffers from treatment attrition and loss to follow-up.

Trial registration

Dutch Trial Register NTR1926

INTRODUCTION

Chronic pain in children and adolescents is defined as pain that exists for more than three months, either continuously or recurrently [105,106,133,136,144,147]. Approximately 25% of children and adolescents (0-18 years) in the general population report chronic pain [133]. Chronic pain is most frequently reported in girls aged 12 to 14 years. The most commonly reported types of pain are limb pain, headache, and abdominal pain.

Chronic pain frequently interferes with activities associated with daily living [60]. Children and adolescents are often unable to pursue their hobbies and to meet their friends. Additionally, they may report school absenteeism, disturbed sleep, frequent use of medication, and depressive feelings. Pain-related disability in adolescents appears to be related to certain pain coping profiles [24]. Adolescents with an avoidant or dependent coping style report more pain-related disability and more depressive and anxiety symptoms than adolescents with more active coping profiles. This knowledge could be used to develop pain management interventions that stimulate the use of active coping strategies in adolescents with pain.

Research has shown that cognitive behavioral therapy (CBT) and relaxation exercises are effective in reducing pain frequency and pain intensity in children and adolescents [44], by providing strategies that help adolescents to better cope with their pain [79]. As a result, depressive symptoms and functional disability may decrease [78,79]. However, CBT seems not to be considered acceptable by all adolescents [120]. Like adults, adolescents with chronic pain might not be inclined to consult a psychologist since they have a biomedical perspective on their symptoms [151]. Additionally, they often have difficulty combining therapy sessions with their busy lives [121]. Psychological interventions using an Internet format might be more appealing to adolescents. The Internet is the most frequently used health information resource in this age group and is therefore in line with their frame of reference [16]. Another advantage of CBT via the Internet is that it will increase adolescents' access to CBT [1]. Both adolescents and therapists can work through the intervention at any place and any time. Adolescents do not have to miss school to visit their therapist.

Indeed several studies have suggested that Internet therapy may be a promising alternative to face-to-face therapy in adolescents with chronic pain [64,128,161]. In a systematic review, Stinson and colleagues reported that Internet-based self-management interventions seemed to improve health outcomes in children and adolescents with various health complaints, including chronic pain [161]. Hicks and colleagues examined the effectiveness of an Internet-based self-management intervention for adolescents with chronic pain in the general population [64]. Treatment consisted of a Web-based manual for children (aged 9-16 years of age; N = 47) and their parents with weekly therapist contact by e-mail or telephone. One month after treatment the adolescents in the experimental group experienced significantly less pain than adolescents in the waiting list group. No differences in quality of life were found. Palermo and colleagues also tested an Internet-based self-management intervention for adolescents with chronic pain and their parents [128]. Treatment consisted of four child modules (11-17 years of age; N = 48) and four parent modules. The modules were tailored to individual needs by personalizing instructions and

assignments (fitting to reported stressors). The adolescents and their parents were supported by a therapist via online messages (in total 1 hour per family). Results showed a greater reduction in pain-related disability and pain intensity after treatment in the Internet group than in the waiting list group. No significant group differences in parental protectiveness or depressive symptoms were found.

A meta-analysis of Spek [158] and colleagues has shown that Internet-based therapy with support is more efficacious than Internet-based therapy without support. Additionally, tailored content seems to increase the effectiveness of Internet-based therapy, probably by increasing personal relevance [103].

The main objective of the present study was to examine the effects of a seven-week guided interactive CBT Internet intervention, Move It Now, for adolescents with chronic pain as compared to a waiting list trajectory. Measurements were taken seven weeks before treatment (T0), pre-treatment (T1), post-treatment (T2), and at three-months follow-up (T3). The innovative aspect of Move It Now is that elements of previous studies are combined: the content of the intervention was tailored to the individual pain characteristics of the participants, and both adolescents and parents received emotional support by e-mail and telephone from a therapist. Move It Now is the first Internet intervention with telephonic therapeutic support according to a specific therapist protocol. The primary outcome of interest in the present study was pain intensity as measured with a VAS score. We hypothesized that pain intensity would decrease during the treatment trajectory, but not during the waiting list trajectory. Secondary outcomes included pain-related disability, quality of life, rewarding of pain behavior by parents, pain coping, and pain catastrophizing. We explored whether adolescents improved on these outcomes during treatment. The second objective of the present study was to examine adolescents' satisfaction with treatment.

METHODS

Participants

The study sample consisted of 69 adolescents. Adolescents were eligible to participate if they met the following criteria: age 12 to 17; self-reported continuous or recurrent pain for at least three months, with minimally one pain episode per month; abdominal pain, headache, back pain or limb pain; no apparent medical cause of pain (an exception is made for adolescents with migraine); self-reported interference with daily activity (e.g., missing school, withdrawing from sports, and withdrawing from activities with friends) or feeling overloaded (e.g., not taking any rest); Internet-access at home; fluent in the Dutch language; no current psychological treatment for pain; no current participation in other research trials; no severe psychosocial problems. During the first months of our trial we changed the age criterion of 12 to 14 years of age to 12 to 17 years of age to increase inclusion. Adolescents did not receive any reward for their participation.

Recruitment of participants took place via youth health-care nurses of the Municipal Health Services Rotterdam-Rijnmond, medical specialists in hospitals and rehabilitation centers all over the Netherlands,

and the media (e.g. Internet, newspapers, and radio). The study started in May 2010. The inclusion of patients ended in September 2013. The three-month follow-up period was completed in December 2013. The full trial protocol can be accessed in Voerman and colleagues [174].

Study design

The present study was originally meant to be a randomized controlled trial. Participants were automatically randomly assigned (ratio 1:1) to an experimental group or a waiting list group by the Move It Now website. The Move It Now website used a randomization list which had been generated by an independent statistician. Block randomization was used to keep the group sizes equal. Randomization was concealed from both the researcher and the therapist.

Adolescents assigned to the waiting list group waited seven weeks before they were allowed to start with the intervention. Adolescents in the experimental group filled out the online questionnaires and the online Pain Dairy three times: before randomization, directly after the intervention, and at three months follow-up. Adolescents in the waiting list group filled out the same questionnaires and the Pain Dairy four times: before randomization, after the waiting period of seven weeks, directly after the intervention, and at three months follow-up. Satisfaction with treatment was only measured directly after the intervention.

We encountered unexpectedly high levels of treatment attrition (52%) and loss to follow-up post-treatment (65%). For this reason we had to adjust our analyses. We did not apply the originally intended between group design [174] but instead used a within group design, comparing the waiting list trajectory with the treatment trajectory. Figure 1 illustrates the progression of the adolescents through the trial. Reasons for treatment attrition were: reduction of pain ($n = 6$), intervention too time consuming ($n = 2$), crashed computer ($n = 1$), worsening psychological complaints ($n = 2$), not satisfied with the content of the intervention (n = 2), and not satisfied with the therapeutic support ($n = 2$). A reported reason for loss to follow-up was: filling out the questionnaires was too confronting ($n = 1$). Twenty-one adolescents gave no reason for treatment attrition and 44 adolescents gave no reason for loss to follow-up.

The study protocol was approved by the Medical Ethical Committee of Erasmus MC University Medical Hospital, Rotterdam, the Netherlands (MEC-2009-195). The study has been registered at the Netherlands Trial Register, which is part of the Dutch Cochrane Centre (NTRcode 1926).

Intervention

Move It Now is a guided interactive CBT Internet intervention for adolescents with chronic pain. The objective of the intervention is to help adolescents improve their coping strategies for pain. The intervention is grounded in the theory of cognitive-behavioral therapy. Adolescents independently work through seven online modules in a fixed order. Adolescents only gained access to the next module by finishing the previous module. The modules aim to teach important self-management skills, including distraction and relaxation. The topics of all modules are presented in table 1. Adolescents interact with online modules by completing fillable responses to queries. Adolescents are asked to login once per

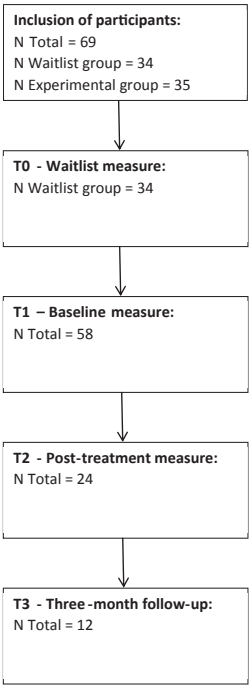

FIGURE 1. Flowchart

TABLE 1. Overview of Move It Now modules

Module number	Module subject	Module contents
1	Background and goals	Psycho-education about pain and its consequences / goal setting
2	Pain killers and breathing	Pain management strategies / information about medication use / deep breathing exercise
3	Relaxation	Education about relaxation / two relaxation exercises
4	Thinking and feeling	ABCDE method for challenging maladaptive thoughts
5	Helpful thinking	Challenging cognitive distortions / thought stopping exercise / distraction exercise
6	Staying active	Behavioural activation / brief relaxation exercise
7	Making a plan	Relapse prevention

week to complete a module, taking about 30 minutes to complete. At the end of each module the adolescents are instructed to practice their skills every day. For example, to do their deep breathing exercise, using the audio files on the website. Participants are contacted weekly by a therapist by e-mail and every other week by telephone. The therapist gives emotional support, checks participants'

understanding concerning the online modules and exercises, and answers questions about the content of the intervention. The content of the e-mails and phone calls is standardized by using a therapist protocol. Parents work through two online modules and have contact with the therapist three times (at the beginning, halfway, and at the end of the intervention). The parent modules are separate from the adolescent modules; parents receive their own login codes. In the parent modules, information is given about how parents should handle the pain of their child. Parents are strongly advised to encourage their child to complete Move It Now.

Much attention has been given to the design and lay-out of the Move It Now website. The adolescents are led through the online chapters by an animated female guide using a voice-over. Several interactive elements were developed to lead adolescents to the material that fits their response, i.e. tailoring. For example, if the adolescent reports having headaches, specific information about headaches is shown.

Measures

The outcome domains in the present study were in accordance with the recommendations of the Pediatric Initiative on Methods, Measurement and Pain Assessment in Clinical Trials (PedIMMPACT)[114]. Our final choice of instruments was based on their availability in Dutch and previous experience with the instruments within our research group.

Pain location was measured with a question with 10 predefined answer options, namely: head, arms, hands, abdomen, back, neck, shoulders, hips, legs, and feet. Adolescents were allowed to give more than one answer. To index time with pain a question with four predefined options was used, namely: 3-6 months, 6-12 months, 1-5 years, and >5 years. Pain intensity was measured with a daily pain diary filled out by the participants once per day for one week, consistent with the study by Palermo and colleagues [128]. Three Visual Analogue Scales (VAS) were recorded and averaged over seven days: 1. Pain intensity at this moment (with *no pain* and *worst pain* ever at the respective endpoints; range 0-100) 2. Pain intensity of the worst pain today (with *no pain* and *worst pain* ever at the respective endpoints; range 0-100) 3. Level of interference caused by the worst pain today (with *I was able to perform all activities of daily living* and *I could not do anything* at the respective endpoints; range 0-100). Prior research has shown that the reliability and validity of VAS scales measuring pain intensity is high [76]. Completion time for the diary is approximately five minutes per day.

Pain-related disability was measured with an adapted version of the Pediatric Migraine Disability Assessment Scale (PedMIDAS) [62,182]. We changed the wording of the questions of the PedMIDAS by replacing the word "headaches" with the word "pain" and "three months" by "one month". The PedMIDAS consists of 6 items. Completion time is approximately 5 minutes. A higher score on the PedMIDAS (range 0-50) is indicative of more pain-related disability. The reliability and validity of the PedMIDAS of the English version are acceptable [62]. However, no Dutch validation study is available. In our sample, the internal consistency varied between .61 and .74, which is moderate to acceptable.

The Child Health Questionnaire - Child Form (CHQ-CF87) [85] was used to measure adolescents' quality of life. The CHQ-CF consists of 87 items, divided over 10 subscales: physical functioning, role functioning

emotional, role functioning behavioral, role functioning physical, bodily pain, general behavior, mental health, self-esteem, general health perceptions, change in health, family activities, and family cohesion. After recoding and transformation, all scale scores ranged from 0 to 100, with 0 indicating worst health and 100 best health. Completion time is approximately 20 minutes. The psychometric properties of the Dutch version of the CHQ are good, even when completed via the Internet [139,140].

Rewarding of pain behavior by parents was measured with the Illness Behavior Encouragement Scale - Child Form (IBES-CF) [180]. The IBES-CF consists of 12 items and measures rewarding by parents in painful situations, as perceived by the child. Higher scores indicate more rewarding of pain by parents (range 0-48). Completion time is approximately 7 minutes. The reliability and validity of the Dutch version of the IBES is satisfactory [11].

Pain coping was measured with the Pain Coping Questionnaire (PCQ) [142]. The PCQ consists of 39 items and has three dimensions: approach (range 14-70), problem-focused avoidance (range 15-75), and emotion-focused avoidance (range 10-50) and eight subscales: information seeking (range 4-20), problem solving (range 5-25), seeking social support (range 5-25), positive self-statements (range 5-25), behavioral distraction (range 5-25), cognitive distraction (range 5-25), externalizing (range 5-25), and internalizing/catastrophizing (range 5-25). Completion time is approximately 20 minutes. Higher scores indicate greater use of the concerning coping strategy. The Dutch version of the PCQ has good psychometric properties [7].

Pain catastrophizing was measured by the Pain Catastrophizing Scale Child version (PCS-C; range 0-52) [32]. The PCS-C consists of 13 items. Completion time is approximately 7 minutes. Higher scores indicate higher levels of pain catastrophizing. Research has shown that the Dutch version of the PCS-C is reliable and has satisfactory predictive validity [32,112,173].

Satisfaction with treatment was measured with six self-constructed questions. The questions "How satisfied are you with the Move It Now intervention?" and "How satisfied are you with the results of Move It Now?" were answered on 5-point scale (with 1 = *very satisfied* and 5 = *very dissatisfied*). The question "Would you recommend Move It Now to other adolescents with chronic pain?" was answered on a two-point scale (with 1 = *yes* and 2 = *no*). The question "To what extent have you obtained the goals you set at the beginning of Move It Now?" was answered on a VAS scale from 0 to 100 (with 0 = *no goals obtained* and 100 = *all goals obtained*). The questions "How much did you improve in dealing with problems since the start of Move It Now?" and "How much did you improve in your daily activities since the start of Move It Now?" were answered on a 5-point scale (with 1= *clearly improved* and 5 = *clearly worsened*). Completion time is approximately 5 minutes.

Additional measures included demographic data and self-developed questions about sleep: "Do you experience any problems with sleeping?" (with 0 = *no* and 1 = *yes*).

Procedure

Adolescents were informed about Move It Now by a youth health-care nurse of the Municipal Health services, a medical specialist in a hospital or rehabilitation center in the Netherlands, or the media (e.g.

Internet, newspapers, radio). When an adolescent decided to participate in Move It Now, he or she contacted the researcher by phone, mail or e-mail. Information about the intervention and the trial was send by mail. After a week the researcher contacted the adolescent and one of the parents by phone. During this phone call, the researcher checked the inclusion criteria, gave information about the study, and asked the adolescent and the parent to send their signed informed consent forms.

When the researcher had received the signed informed consent forms, the adolescent and the parent were registered at the Move It Now website. An automatic e-mail was sent with their personal login codes. When the adolescent logged into the website, he or she first filled out the online questionnaires. Additionally, the adolescent kept an online Pain Dairy for one week. After completing the baseline measurement, the adolescent and the parent received an automatic e-mail that informed them whether they could start with Move It Now directly in the intervention group or if they had to wait for seven weeks in the waiting list control group. After seven weeks adolescents in the waiting list control group had to complete the questionnaires and the Pain Dairy for a second time to be able to access the intervention.

Data analysis

Multi-level modelling (SPSS 21, MIXED module) was applied for longitudinal analysis. This analysis allows handling incomplete data with a minimal loss of information, as incomplete cases can be included in the analyses. Results of the measurements seven weeks before treatment (T0), pre-treatment (T1), post-treatment (T2), and at three-months follow-up (T3) are presented. At three-months follow-up there were too many missing cases to validly calculate mean scores for the CHQ scale family activities. Linear splines were used to test the differences in outcomes between time points, in other words, the trajectories T0-T, T1-T2, and T2-T3 were analysed. For all differences, effect sizes are reported, with d = .2-.5 indicating a weak effect, d = .5-.8 indicating a medium effect and d = >.8 indicating a strong effect [25]. It seems that drop-out has not been selective, since drop-out analysis (T1-T2) showed no significant differences for the primary outcome (pain intensity) at T1 (p = .64 and p = .33 respectively).

RESULTS

Inclusion numbers

According the randomization scheme (Figure 1), 34 adolescents were selected for the waiting list group and 35 for the experimental group. All adolescents in the waiting list group completed the questionnaires 7 weeks before baseline. The baseline measures were filled out by 58 adolescents, in both the waiting list group and the experimental group. Treatment protocol was completed by 33 adolescents. The post-treatment measures were filled out by 24 adolescents and the three-month follow-up measures by 12 adolescents.

Patient characteristics

Table 2 summarizes the characteristics of the sample both overall and by group. Participants consisted of 69 adolescents (77% female), aged between 12-17 years (M = 14.88, SD = 1.11). The most reported pain types were headaches (68%), back pain (40%), and pain in the legs (42%). Most of the adolescents had been in pain since one to five years. The majority of adolescents participated in pre-vocational secondary education (38%). Almost half of the adolescents (49%) had a family member who also experienced chronic pain. Almost half of the adolescents had visited their GP (46%) and 86% had used pain medication. Twelve percent had visited a psychologist because of their pain. Forty-one percent of the adolescents reported other somatic complaints besides pain. Patient characteristics did not differ

TABLE 2. Characteristics of the sample at baseline

	Randomized sample (N=66)	Treatment group (N=33)	Waitlist control (N=33)	Overall *p*-value
Mean age in years (SD)	14.88 (1.11)	14.67 (1.47)	15.09 (1.40)	.24
Gender (female)	77%	79%	76%	.77
Pain location				
Head	68%	72%	64%	.53
Arms	32%	28%	36%	.53
Hands	26%	25%	28%	.80
Abdomen	21%	22%	20%	.86
Back	40%	31%	52%	.11
Neck	32%	22%	44%	.08
Shoulders	35%	25%	48%	.07
Hips	18%	19%	16%	.79
Legs	42%	38%	48%	.43
Feet	39%	41%	36%	.72
Pain duration				.69
3-6 months	4%	3%	4%	
6-12 months	23%	28%	16%	
1-5 years	56%	50%	64%	
>5 years	18%	19%	16%	
Education				.52
Pre-vocational secondary education	38%	33%	42%	
Higher general secondary education	23%	30%	15%	
Pre-university education	27%	24%	30%	
Other education	12%	12%	12%	
Other family member with pain	49%	39%	58%	.14
Care use last month				
GP consultation	46%	49%	42%	.62
Used medication	86%	88%	85%	.72
Psychologist or social worker consultation	12%	12%	12%	1

significantly from participants in the treatment trajectory and adolescents starting in the waiting list trajectory. Patient characteristics also did not significantly differ for adolescents who completed treatment and adolescents who did not complete treatment. However, compared with adolescents who dropped out between pre-treatment (T1) and post-treatment (T2), adolescents who completed treatment did report a significantly lower level of physical quality of life (t = 2.34, p = .02) and significantly more use of cognitive distraction as a coping strategy (t = -2.32, p = .02) at baseline.

Changes in primary outcomes

For our primary outcome pain intensity no significant changes were found during the waiting list period (T0-T1). From pre-treatment to post-treatment (T1-T2) *pain intensity at this moment* and *pain intensity of the worst pain today* significantly decreased from pre-treatment to post-treatment (T1-T2) (d_{T1-T2}=-.42 and d_{T1-T2}=-.43 respectively), indicating a small improvement. From post-treatment to follow-up (T2-T3) no significant changes were found for pain intensity (Tables 3 and 4).

Changes in secondary outcomes

Waiting list period (T0-T1)

For our secondary outcomes, only one significant change was found during the waiting list period. Encouragement of pain behavior by parents significantly increased during this period (d=.57).

Treatment period (T1-T2)

Interference caused by pain significantly decreased from pre-treatment to post-treatment (d=-.46). Also, pain measured with the CHQ-CF significantly decreased from pre-treatment to post-treatment (d=-.55). The CHQ scores for general behavior, mental health, family activities, and health changes did significantly improve from pre-treatment to post-treatment (d=.34, d=.40, d=.40, and d=-.87 respectively). The other CHQ scores did not significantly differ from pre- to post-treatment. Encouragement of pain behavior by parents significantly decreased during the treatment period (d=.91). No significant differences were found for the approach dimension and the emotion focused avoidance dimension of the PCQ. Problem-focused avoidance did significantly increase from pre-treatment to post-treatment (d=.45). Sleep problems significantly decreased from pre-treatment to post-treatment (d=-.60). No significant differences were found for pain-related disability and pain catastrophizing.

Follow-up period (T2-T3)

Almost all effects were maintained at three-months follow-up, except the improvement in mental health. Mental health significantly decreased during follow-up (d=-.71). Although no changes were found for catastrophizing between T1 and T2, catastrophizing was reduced at T3 (d=-.52). Details about the mixed models and their regression weights are presented in table 3. Estimated means and effect sizes of the results are presented in table 4.

TABLE 3. Parsimonious mixed models of outcome variables

Outcome	Intercept		T0-T1 Waiting list period		T1-T2 Experimental period		T2-T3 Follow-up period	
	estimate	*p*-value	estimate	*p*-value	estimate	*p*-value	estimate	*p*-value
Pain intensity (VAS)								
At this moment	44.22	<.01	.34	.93	-10.70	.03	5.29	.40
Today	58.90	<.01	-1.52	.71	-10.70	.04	.28	.97
Pain interference (VAS)	42.49	<.01	-.62	.89	-11.17	.03	.25	.97
Pain-related disability (PedMIDAS)	26.43	<.01	-.07	.99	-7.62	.14	5.32	.55
Quality of life (CHQ)								
Physical functioning	70.88	<.01	2.88	.36	2.70	.42	1.72	.77
Role functioning emotional	68.16	<.01	5.07	.35	5.07	.39	7.43	.47
Role functioning behavior	84.64	<.01	.60	.88	-1.25	.76	10.09	.18
Role functioning physical	63.52	<.01	-1.65	.77	10.80	.08	-5.01	.65
Bodily pain	57.64	<.01	-3.23	.43	-13.34	.<01	9.21	.36
General behavior	80.59	<.01	-.65	.69	3.66	.04	-.90	.77
Mental health	64.45	<.01	-.41	.86	6.21	.01	-11.08	.01
Self-esteem	43.19	<.01	-.88	.62	-2.91	.13	-3.36	.31
General health perceptions	51.76	<.01	2.51	.32	-2.61	.33	5.02	.28
Change in health	63.63	<.01	4.71	.35	-25.11	<.01	-6.01	.59
Family activities	74.61	<.01	4.32	.05	6.56	<.01		
Family cohesion	32.52	<.01	.63	.84	-4.51	.17	-1.60	.81
Rewarding of pain behavior by parents (IBES)	20.54	<.01	2.55	<.01	-4.08	<.01	.95	.58
Pain coping (PCQ)								
Approach	49.04	<.01	1.05	.49	1.62	.33	-2.63	.37
Problem focused avoidance	38.02	<.01	.56	.68	4.42	<.01	-.08	.98
Emotion focused avoidance	21.10	<.01	.36	.64	-1.32	.12	-.84	.56
Information seeking	7.80	<.01	.51	.31	-.37	.51	-.31	.75
Problem solving	14.36	<.01	.05	.94	.64	.39	-1.49	.25
Seeking social support	13.28	<.01	.63	.35	-.39	.59	-.22	.86
Positive self-statements	13.60	<.01	-.12	.85	1.60	.03	-.50	.69
Behavioral distraction	11.66	<.01	.50	.26	1.46	<.01	.55	.51
Cognitive distraction	12.88	<.01	.03	.97	1.52	.03	-.10	.93
Externalizing	7.72	<.01	.12	.75	-.10	.81	.93	.21
Internalizing/ Catastrophizing	13.35	<.01	.29	.64	-1.28	.06	-1.66	.16
Pain catastrophizing (PCS)	23.81	<.01	-.39	.76	-1.99	.17	-5.64	.02
Sleep problems	.63	<.01	.02	.83	-.28	<.01	-.11	.42

TABLE 4. Estimated means and effect sizes of outcomes

Outcome	Estimates				Effect sizes		
	T0	T1	T2	T3	*d* T0-T1 Waiting list period	*d* T1-T2 Experimental period	*d* T2-T3 Follow-up period
Pain intensity (VAS)							
At this moment	44.22	44.56	33.86	39.79	.01	-.42	.23
Today	58.88	57.36	46.69	46.98	-.06	-.43	.01
Pain interference (VAS)	42.49	41.86	30.69	30.94	-.03	-.46	.01
Pain-related disability (PedMIDAS)	26.43	26.36	18.74	24.05	.00	-.36	.25
Quality of life (CHQ)							
Physical functioning	70.88	73.76	6.46	78.18	.14	.13	.08
Role functioning emotional	68.16	73.22	78.29	85.63	.17	.17	.25
Role functioning behavior	84.64	85.23	83.98	94.07	.03	-.05	.43
Role functioning physical	63.42	61.76	72.56	67.56	-.05	.35	-.16
Bodily pain	57.64	54.41	41.07	50.29	-.13	-.55	.38
General behavior	80.59	79.93	83.60	82.70	-.06	.34	-.08
Mental health	64.45	64.04	70.25	59.17	-.03	.40	-.71
Self-esteem	34.19	33.31	30.39	27.04	-.06	-.21	-.24
General health perceptions	51.76	54.26	51.65	56.67	.13	-.14	.26
Change in health	63.63	68.34	43.23	37.22	.16	-.87	-.21
Family activities	74.61	78.93	85.49		.26	.40	
Family cohesion	32.52	33.14	28.63	27.03	.03	-.20	-.07
Rewarding of pain behavior by parents (IBES)	20.54	23.08	19.00	19.95	.57	-.91	.21
Pain coping (PCQ)							
Approach	49.04	50.09	51.71	49.08	.09	.14	-.23
Problem focused avoidance	38.02	38.57	43.00	42.91	.06	.45	-.01
Emotion focused avoidance	21.10	21.46	20.14	19.30	.05	-.19	-.12
Information seeking	7.80	8.31	7.94	7.63	.17	-.12	-.10
Problem solving	14.36	14.41	15.05	13.56	.01	.15	-.35
Seeking social support	13.28	13.90	13.51	13.29	.13	-.08	-.05
Positive self-statements	13.60	13.47	15.08	14.57	-.03	.34	-.11
Behavioral distraction	11.66	12.15	13.61	14.16	.15	.43	.16
Cognitive distraction	12.88	12.91	14.43	14.33	.01	.41	-.03
Externalizing	7.72	7.84	7.74	8.67	.04	-.03	.29
Internalizing/ Catastrophizing	13.35	13.64	12.36	10.70	.06	-.25	-.33
Pain catastrophizing	23.81	23.42	21.43	15.79	-.04	-.18	-.52
Sleep problems	.63	.65	.37	.25	.04	-.60	-.24

Treatment satisfaction

The treatment satisfaction questionnaire was completed by 21 adolescents, who completed treatment. Most of these adolescents reported that they were satisfied with the Move It Now intervention (57%). Only 5% of the participants were unsatisfied and 5% reported that they were very unsatisfied. Almost half of the adolescents were neutral about their improvements since the start of Move It Now (43%); 33% was satisfied. Seventy-six percent of the adolescents said they would recommend Move It Now to other adolescents with pain. Participating adolescents on average achieved half of their goals set at the beginning of Move It Now ($M = 50.1$, $SD = 30.0$). A slight majority of the adolescents (57%) reported that they felt more able to deal with their problems since the start of Move It Now. Furthermore, 38% of the adolescents reported that they had improved in carrying out their daily activities. Graphic displays of the satisfaction results are presented in Figure 2.

CONCLUSION AND DISCUSSION

The aim of the present study was to evaluate the effects of the first Dutch guided Internet-delivered self-help intervention for adolescents with chronic pain. Contrary to expectations based on the literature, a guided Internet-delivered self-help intervention for chronic pain seemed difficult to employ in Dutch adolescents, as it suffered from treatment attrition and loss to follow-up. Therefore, the results might be biased and not generalizable to the general population of adolescents with chronic pain. Nevertheless, we found indications that the adolescents in our trial perceived a reduction in pain after treatment. Additionally, interference caused by pain, rewarding of pain behavior by parents and sleep problems significantly decreased. The quality of life scores for pain, general behaviour, mental health, family activities, and health changes did also significantly improve during the intervention. Concerning coping, only problem-focused avoidance behaviour significantly increased. No significant differences were found for pain-related disability and catastrophizing. Overall, the adolescents reported to be satisfied with Move It Now. However, some of the adolescents dropped-out because they experienced the intervention to be too time consuming, they were not satisfied with the content, or were not satisfied with the therapeutic support. Therefore, our satisfaction measure may reflect an overestimation of the satisfaction with Move It Now.

Concerning the reduction in pain intensity, our results are in line with previous studies [64,128]. Furthermore, in line with the study by Hicks and colleagues [64] we did not find changes in overall measures of quality of life. Concerning pain-related disability, our results differ from the results of Palermo and colleagues [128]. Palermo and colleagues found adolescents were more able to perform activities after treatment. In contrast, we did not find any changes in pain-related disability. This difference in results might be partly explained by the fact that Palermo and colleagues used another instrument to index pain-related disability, i.e. the Child Activity Limitation Interview (CALI) [129]. On the CALI, adolescents select the eight activities, out of a list of 21 activities, that are most difficult for them due to their pain. Difficulty is assessed on a 5-point scale, with 0 = *not very difficult* and 4 = *extremely difficult*. On the

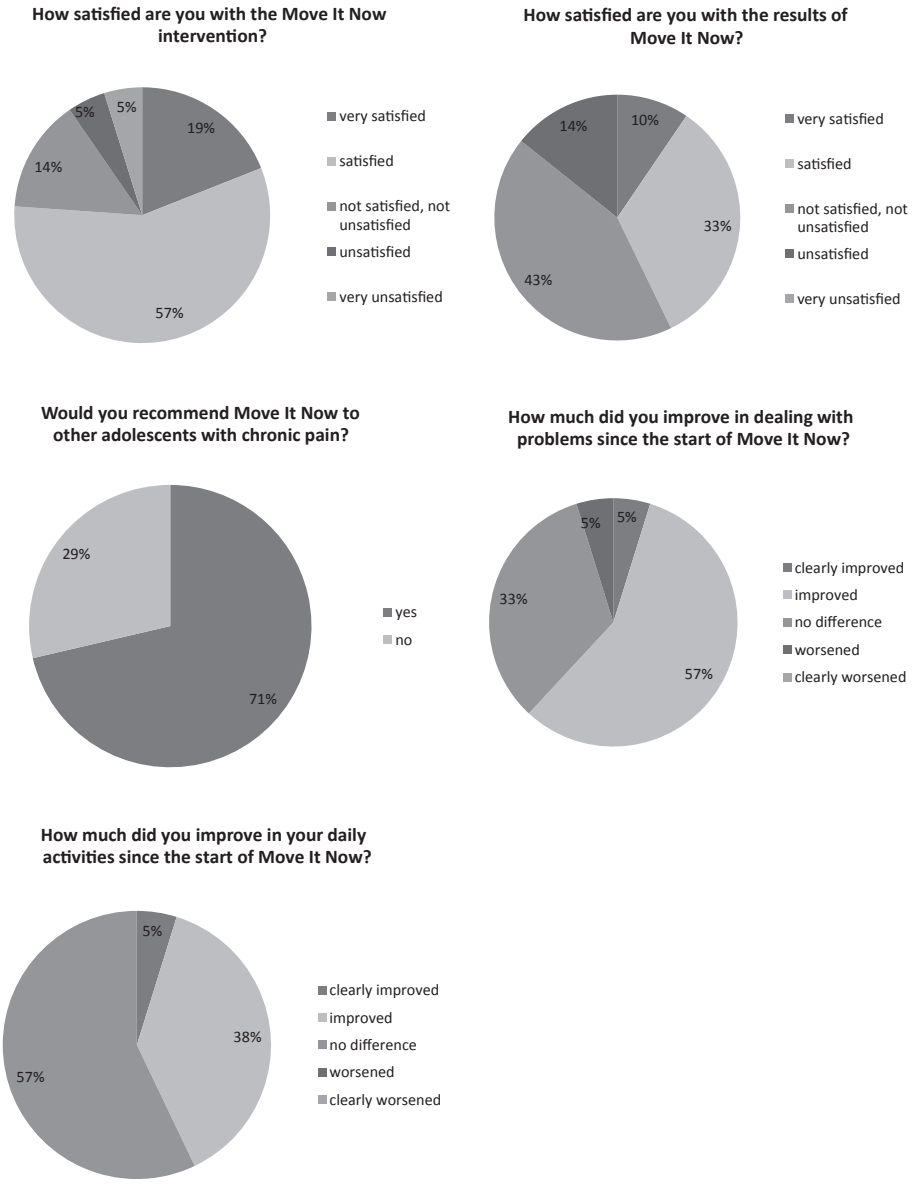

FIGURE 2 Satisfacture with treatment

adapted PedMIDAS adolescents report the number of days a specific aspect of functioning (school, chores at home, and leisure activities) was impaired in the previous month due to pain. The CALI is more tailored to the individual and able to measure smaller differences. Therefore, the CALI seems to measure

a somewhat different concept than the adapted PedMIDAS. Besides differences in the instruments used, the rather low internal consistency of the PedMIDAS in our sample (α = .61-.74) might explain why we did not find any differences in pain-related disability. Furthermore, differences in the intervention and differences in the study protocol may have also contributed to differences in this outcome measure. In the study of Palermo [128] and colleagues, participants did not receive telephonic support by a therapist. Their intervention consisted of eight modules for both adolescents and parents, whereas our intervention consisted of seven adolescent modules and two parent modules. Concerning study protocol, Move It Now was the first online intervention study in adolescents with chronic pain in which almost all data was collected online. There might be differences in how adolescents respond to paper-and-pencil questionnaires versus online measures.

Pain coping was not used as an outcome measure in previous child and adolescent studies. It is an interesting finding that the approach dimension and the emotion focused avoidance dimension of the PCQ did not significantly differ from pre-treatment to post-treatment, but the problem focused avoidance dimension did. Problem focused avoidance increased during treatment, which was not expected. However, the problem focused avoidance dimension consists of the subscales positive self-statements, behavioral distraction, and cognitive distraction. These coping strategies seem to match with the cognitive-behavioral techniques included in the intervention. In the intervention, adolescents were encouraged to divert their attention away from their pain to pleasant thoughts and activities. Although positive self-statements, behavioral distraction, and cognitive distraction have been categorized as avoiding, these behaviors might be helpful when finding a solution for the pain (active coping) is no longer possible [168].

An important limitation of the present study is the small sample size, due to treatment attrition and loss to follow-up. This sample size reduction had substantial consequences for statistical power and analyses; it was no longer possible to compare the experimental group with the waiting list group. A true control group was no longer available. Although we did compare the treatment trajectory with the waiting list trajectory, we are not completely able to rule out spontaneous recovery (or regression to the mean).

A second limitation is the self-selection bias of the participants. Adolescents could register themselves for treatment via the Internet. Adolescents who prefer medical treatment or face-to-face psychological therapy did not respond to our invitation for online treatment. Therefore, results might not be generalizable to all adolescents with chronic pain. However, the results are generalizable to the group of adolescents who will use online self-management in practice, namely those adolescents who feel attracted to online psychological guidance. A final limitation is that we measured pain for only one week instead of three weeks. In our trial, we included adolescents with continuous, but also recurrent pain (e.g., migraine). Adolescents with recurrent pain could have had a week without pain when they filled out the pain dairy. However, we did find a significant difference when measuring pain for only one week. Given the mentioned concerns, it is important to interpret the present findings with caution.

An important strength of the present study is that the outcome domains were in accordance with the recommendations of the PedIMMPACT [114] and therefore were multidimensional. In most studies, pain intensity and physical disability are used as the only outcome measures. However, chronic pain is rather complex and is influenced by biopsychosocial factors. Therefore, it is important that also functional and emotional measures are included in research trials.

Contrary to the expectation that for adolescents with chronic pain Internet therapy would be more acceptable than face-to-face therapy, Move It Now was difficult to employ. In the early stage of our trial we had great difficulty recruiting participants. The reasons are unclear. However, potential referrers mentioned it was hard to motivate adolescents for non-medical treatment On the other side, referrers (e.g. physicians, nurses, and psychologists) were hesitant to refer adolescents to Move It Now. They had doubts on the effectiveness of online treatment as a sole intervention to help adolescents to improve their chronic pain coping.

Besides inclusion problems, our trial suffered from substantial treatment attrition. The most reported reason for treatment attrition was pain reduction. Unfortunately, we were not able to motivate adolescents with reduced pain to complete the treatment protocol. Also, adolescents frequently reported that they forgot to complete their modules. Similarly, it was hard to motivate adolescents to complete the questionnaires after treatment, even though we frequently reminded them by phone and e-mail. The Internet format we used might have given the adolescents a feeling of 'laisser faire'. Considering the number of outcome measures we used, study burden might have been another reason for treatment attrition and loss to follow-up. Completing the questionnaires may have felt like doing homework to the participants. Treatment attrition and loss to follow-up are commonly reported problems in online trials [125], being even more common than in conventional trials. The lowest follow-up rates have been found in studies using only e-mail reminders, whereas the highest follow-up rates have been found in trials combining postal reminders with cash incentives. Therefore, we could recommend minimizing data collection as well as using postal reminders and cash incentives in future studies. Using the so-called "blended care" approach may minimize the feeling of 'laisser faire'. In blended care, face-to-face therapy sessions are combined with online support and online exercises. This approach appears to combine the best of both; i.e. the strong therapist-patient relationship and the lower drop-out of face-to-face therapy and the higher efficiency (in time and money) of online therapy.

The treatment attrition in our study (52%) was larger than in the studies of Palermo and colleagues [128] and Hicks and colleagues [64] (respectively 8% and 32%). The interventions are quite comparable in content and in the age group evaluated. However, there might have been a difference in the frequency of the (telephonic) reminders during the completion of the measures and the intervention. We choose to only minimally remind the adolescents during the intervention itself, because in clinical practice it would not be achievable to constantly remind adolescents.

In future, large scale effectiveness and cost-effectiveness studies should be performed to prove guided Internet-delivered self-help interventions are effective in reducing pain and improving pain coping and quality of life in adolescents. More research is also needed to improve implementation of

Internet-based self-management. For example, it would be interesting to know what types of adolescents with chronic pain benefit most from online help. Additionally, treatment adherence might be enhanced by an initial face-to-face session or perhaps even a combination of face-to-face therapy and Internet therapy (i.e. blended care). Furthermore, little is known about the experiences of adolescents with computerized interventions for chronic pain. Both qualitative and quantitative studies could be used to further improve the satisfaction with Internet therapy.

In conclusion, Internet-based self-management seems to reduce pain in adolescents with chronic pain, but contrary to expectations, a guided Internet-delivered self-help intervention for chronic pain is difficult to employ in adolescents, as it suffers from treatment attrition and loss to follow-up. Adolescents with chronic pain seem nevertheless satisfied with this mode of treatment.

CHAPTER 7

General discussion

1. INTRODUCTION

The main objectives of this thesis were to find ways to improve, first, the early signaling and assessment of chronic pain in children and adults, and second, the self-management of chronic pain. In this chapter the results of the separate studies are integrated and discussed. The chapter ends with recommendations for future research and practical implementation.

2. DISCUSSION OF THE RESULTS

2.1 Prevalence and psychosocial assessment of chronic pain

2.1.1 Prevalence of chronic pain in adolescents

In **chapters 2** and **3** we estimated the prevalence of chronic pain in adolescents at 6% and 9%, respectively. We may conclude that these two estimates do not essentially differ. However, other studies arrive at quite divergent figures [82]. For example, Perquin and colleagues [133] concluded that 25% of the children and adolescents (0-18 years) in the open population suffer from chronic pain. Of a sample of children and adolescents (4-18 years) studied by Roth-Isigkeit and colleagues [144], 9.5% reported to have had pain for more than three months, and 8.7% reported to have had pain for more than six months. These discrepancies in findings may be explained by the use of different definitions of chronic pain in the used definition of chronic pain [82], as well as differences in characteristics of the study populations. Variations in gender, age, and psychosocial variables may influence prevalence estimates. For example, it is known that pain prevalence is higher in girls than in boys and increases with age. Furthermore, higher pain prevalence is correlated with the presence of other chronic health problems and low socio-economic status. A higher estimate precision could be obtained if consensus should have been reached about a standard measurement for chronic pain in children and adolescents. Additionally, consensus about the population sampling method would help to get consistent estimates over time and place.

2.1.2 Signaling of chronic pain in adolescents

In **chapter 2** we addressed the research question: Are negative life events, e.g. bullying, abuse, and family conflict risk factors for chronic pain in adolescents. The results of our study show that the following negative life events are more common in adolescents with chronic pain than in adolescents without chronic pain: physical abuse by adults who were not the adolescents' parents, sexual abuse, family conflict, and being bullied. Physical abuse by parents and parental divorce were not significantly related to chronic pain. Bullying, abuse, and family conflict may be risk factors for chronic pain. Early signaling of these stressors and providing counselling might perhaps prevent the development of chronic pain. A systematic review of Hahn and colleagues [59] shows that school-based programs intended to prevent violent behaviour (i.e., bullying, physical abuse, and sexual abuse) are indeed effective in reducing violence among children and adolescents. The evaluated interventions are targeted at improving skills like

emotional self-awareness, emotional control, self-esteem, positive social skills, social problem solving, conflict resolution, and team work. Possibly these programs might have a favourable effect on pain too.

2.1.3 Chronic pain and academic performance

In **chapter 3** we addressed the research question: Is disabling pain associated with lower grades, reduced emotional well-being, and more attention problems? We found that disabling pain in adolescents indeed was associated with lower grades, reduced emotional well-being, and reduced self-reported attention. The association between disabling pain and lower grades was no longer significant, however, when either emotional well-being or attention was entered as a covariate. The association between disabling pain and grades can partially be explained by both reduced emotional well-being and attention problems. The influence of pain on emotional-wellbeing has been extensively studied. Only few studies have examined the influence of pain on attention in adolescents. Since our study is only a first cross-sectional study, it would be worthwhile to further explore the relation between chronic pain and attention problems in longitudinal studies.

The results of these studies could be used to develop appropriate psycho-educational techniques that can help these adolescents to cope with chronic pain and related emotional problems. A multidimensional model for stress reactions in adolescents proposes three types of voluntary coping: primary control, secondary control, and disengagement [29]. Primary control is defined as attempts to alter one's emotions or the stressor itself. Secondary control coping is characterized by modifying cognitions or regulating attention. Disengagement coping is defined as removing oneself from the stressor or the emotions related to the stressor. Disengagement coping is associated with higher levels of pain; and primary and secondary control are associated with lower levels of pain [28,164]. Since the development of primary and secondary control coping is largely influenced by higher order executive functions [27], psycho-educational techniques might be aimed at improving executive functions (e.g. planning and problem solving). For example, metacognitive therapy (MTC) is a relatively new kind of psychological treatment, targeted at executive functioning, including attentional biases. MCT includes mindfulness training and an auditory attention task to practice selective and sustained attention and attention switching. A study of Groves and colleagues [57] shows that MTC indeed has an advantage over cognitive-behavioral therapy (CBT) in improving aspects of executive functioning, including attention, in adults with depression. To our knowledge, so far no studies have evaluated the effects of MTC among patients with chronic pain.

2.2 Efficacy of self-management interventions of chronic pain

2.2.1 Use of lay trainers in treatment for chronic pain

In **chapter 4** we answered the research question: Was improvement in migraine frequency, self-efficacy in managing headache episodes, and headache specific internal control resulting from BMT sustained or even more marked two to four years after BMT? BMT provided by lay trainers was addressed in a previous study [116]. From our own study we conclude that the short-term improvements in attack frequency and

self-efficacy found post-BMT were indeed sustained at two to four years. The level of internal control, which had increased during BMT, had decreased from post-BMT to long-term follow-up. Quality of life and migraine-related disability improved gradually over time. The findings of this first long-term follow-up study suggest that BMT for migraine carried out by lay trainers may be beneficial on the long term. Still, caution is needed: a follow-up control group was lacking, as well as information about additional treatments patients may have received during the follow-up period. Therefore, a closer look at the treatment by lay-trainers for patients with migraine is recommended.

An important advantage of involving lay trainers is that it provides better access to psychological therapy [3,31,145,163]. And what is more, lay trainers might improve patients' internal motivation to adhere to treatment [150]. One of the barriers to self-management training is that therapists generally are focused at information provision and verbal persuasion. This might have adverse effects on patients' self-efficacy. Lay trainers will be more inclined to encourage patients' autonomy, thereby increasing their patients' perceived confidence in their own skills [150]. Still, previous studies that compared lay trainer with health care professional led therapy did not find any differences in the outcomes [6,26,37,100]. For example, a study of Baksi [6] and colleagues suggests that peer advisors are as effective as specialist health professionals in helping patients with diabetes to self-manage their disease. Sharma and colleagues [150] hypothesized that co-delivery of self-management programs might be more effective than therapy delivered by either health care professionals or lay trainers alone. They assumed that while health care professionals would be better suited to provide structure and engage patients, lay trainers would more support patients' autonomy. However, their first results failed to confirm the hypothesis; health care professionals and lay trainers used the same teaching styles and hardly supported patients' autonomy.

2.2.2 Internet interventions for adolescents with chronic pain

In **chapter 6** we addressed the research question: What is the efficacy of a guided interactive internet intervention, Move It Now, for adolescents with chronic pain? It appeared that pain intensity (our primary outcome), rewarding of pain behavior by parents, and sleep problems had significantly decreased from pre-intervention to post-intervention. No significant changes were found for general physical and psychosocial quality of life. General behavior, mental health, and family activities did significantly improve from pre-treatment to post-treatment. Concerning coping, no significant differences were found for approach and emotion-focused avoidance. Problem-focused avoidance behavior, on the other hand, had significantly increased from pre-intervention to post-intervention. No significant differences were found for pain-related disability and pain catastrophizing.

Contrary to the expectation that for these adolescents Internet therapy would be more acceptable than face-to-face therapy, Move It Now was not a great success. In the early stage of our trial we had great difficulty recruiting participants. The reasons are unclear. Perhaps these adolescents just found it irrelevant or boring, and did not like to spend time on it. On the other side, potential referrers (e.g. physicians, nurses, and psychologists) were hesitant to refer adolescents to Move It Now, because online treatment was relatively unknown to them. They had doubts on the effectiveness of online treatment

as a sole intervention to help adolescents to improve their chronic pain coping. Additionally, referrers had trouble motivating adolescents for online CBT. They admitted they found it hard to explain to the adolescents how they could benefit from an online self-management program.

Besides inclusion problems, our trial suffered from substantial treatment attrition. A possible reason is the online therapy is noncommittal nature of therapy: there is no therapist waiting for you at a specific place, at a specific time. At the moment we are analyzing the results of a qualitative interview study examining how adolescents experienced Move It Now. Some of the interviewees told us they had trouble reminding themselves to work through the online modules. Another disadvantage they mentioned was that they did not immediately receive answers to any questions. They had to email questions and then wait for a response from the therapist. Others told us they tended to rush through the modules instead of taking the time for it. With no therapist motivating them to rehearse the difficult parts, they just went on to the next module.

Yet, benefits of Move It Now were pointed out as well. One of the benefits participants mentioned was that they did not have to travel to therapy and that they could choose themselves when to work through the modules. This made it easier to combine therapy with daily activities, e.g. school and sports. Second, it was convenient that they go back to the information in the modules as often as they liked. At times of relapse, for example, they could work through the strategies that helped them before.

Previous studies have shown that professionals' attitudes towards online treatment are more negative than patients' attitudes [77,181]. Professionals do not believe that online therapy can be as effective as seeing a patient face-to-face. In our study the therapists reported several disadvantages of online therapy. First, in spite of the short training they had received, online consults were relatively new to them. It was no part of their basic education. Sometimes they found it hard to adhere to their study protocol, because in face-to-face therapy they were used to discuss more subjects than they had to discuss in Move It Now. This problem could probably partly be overcome by greater exposure to and more intense training about online support. Second, the therapists found it hard to combine the online and telephonic support with their face-to-face sessions with other patients. They were of the opinion that this type of support can never be so urgent as support to a patient sitting in front of you. As a result the necessary calls were sometimes forgotten. Third, adolescents were hard to reach by phone between 9 in the morning and 5 in the afternoon. One of the therapists changed her working hours, so she could phone the adolescents between 6 p.m. and 7 p.m. After the study, the therapists admitted they did not like online consults as much as they did face-to-face consults. They missed the personal contact. Many studies have shown that the therapist-patient relationship is a major factor in therapy success [33,130,158]. In online therapy this relationship is rather weak, since there is only minimal contact.

In summary, both adolescents and therapists had severe reservations as to the effectiveness of online therapy. Should we stop using online therapy and go back to face-to-face therapy then? We do not think so. Online therapy definitely can have a place in psychological treatment. Besides our study, the studies of Hicks and colleagues [64] and Palermo and colleagues [128] found indications that online therapy is promising in reducing pain intensity among adolescents with chronic pain.

Furthermore, the so-called "blended care" approach is upcoming. In blended care, face-to-face therapy sessions are combined with online support and online exercises. This approach therefore seems to combine the best of both; i.e. the strong therapist-patient relationship and the lower drop-out of face-to-face therapy and the higher efficiency (in time and money) of online therapy. A few studies have shown that blended care for depression in adults is effective in reducing depressive symptoms [63,67,81]. It should be noted, however, that in these studies online cognitive-behavioral therapy (CBT) was added to care-as-usual, without diminishing the amount of face-to-face sessions. In an uncontrolled study in which the online and face-to-face sessions were combined into only an eight-week protocol, Månsson and colleagues [108] found that this blended care approach had diminished patients' anxiety and depression levels. In view of these findings it is not unlikely that blended care could also be a better solution for adolescents with chronic pain, instead of online sessions alone.

3. RECOMMENDATIONS FOR FUTURE RESEARCH

In **chapter 2** we introduced the Pain Barometer, a self-constructed questionnaire to detect chronic pain among adolescents at an early stage. The psychometric properties of the Pain Barometer have not yet been established. In future research the Pain Barometer should be validated for this aim, for example by comparing it with the gold standard so far, i.e. a semi-structured interview.

Although BMT for migraine was effective in adults (**chapter 4**), to our knowledge the effectiveness of BMT by lay trainers has not yet been studied in adolescents with migraine. Adolescents who suffer from migraine themselves and are able to control this might be able to help their peers with the same complaints, though under supervision of a therapist. Furthermore, BMT by lay trainers might also be effective in reducing other types of chronic pain, e.g. low back pain and fibromyalgia. Future studies should explore the efficacy of BMT by lay trainers among subgroups of patients with these other types of chronic pain.

In **chapter 6** the efficacy of Move It Now was evaluated with quantitative statistics. However, quantitative research has some limitations concerning the evaluation of online interventions. For example, it reduces effects to numeric values and does not examine the experiences of participants. Qualitative research can overcome these limitations. Future research should study adolescents' perspectives on computerized interventions. Adolescents can be expected to prefer online therapy over face-to-face therapy, because they are so familiar with online technology. But is this really true? And if the answer is "yes", what aspects of online therapy do they like and what aspects not?

Additionally, future studies should try to get a grip on predictors of non-adherence, for adherence to Internet therapy is very low. Predictors of non-adherence might be found in demographic factors, like gender and age, but also pain characteristics, such as duration and intensity of the pain before the start of therapy. Information about the reasons for and predictors of non-adherence might give solutions to better implement e-health in the care for chronic pain in adolescents.

In addition, it would be worthwhile to study possible strategies to improve adherence. Previous research suggests that self-monitoring and feedback of professionals might improve adherence [14,21,74]. Moreover, tailored feedback seems more effective than generic feedback. A study of De Niet and colleagues [36] showed that a simple adherence intervention delivered by short message service (SMS) reduced drop-out in obesity treatment in children. The children weekly self-monitored themselves by sending information to the researcher about the time spent on physical activity, the number of days they adhered to their diet, and the number of days they felt happy. Within two days a message tailored to the child's individual pattern of change in lifestyle behaviour was sent in return. These messages were aimed to promote social support, to encourage and motivate, to reinforce positive changes, and to suggest changes in behaviour in cases of negative development. A study of Cafazzo and colleagues [22] found that adolescents with diabetes improved their blood glucose monitoring using an app with gamification incentives. Besides SMS and apps, serious gaming could further improve adherence. Serious gaming is gaming with the intention to gain some information or to develop some skills. Patients motivation for treatment could be improved by serious gaming, because the gaming environment is found attractive by being challenging [87]. Jansen-Kosterink and colleagues [75] used a serious game to motivate their adult patients with chronic musculoskeletal back and neck pain to do their exercises. Their pilot study showed that the game indeed seemed to motivate patients, which resulted in increased motor skills and physical condition. The use of SMS, apps or serious gaming could probably also be effective for adolescents with chronic pain.

4. PRACTICAL IMPLICATIONS

As found in **chapters 2** and **3**, chronic pain in adolescents seems prevalent, and is almost as prevalent as depression (10%) [15]. As it can interfere with adolescents' daily activities, signaling of chronic pain should be standard procedure in the surveillance of children and adolescents' health. This could be achieved, for example, by administering the Pain Barometer, which was part of the Rotterdam Youth Monitor (RYM), a set of self-report questionnaires to early identify physical and psychosocial problems. Adolescents filled out these questionnaires three times in the course of their secondary school education. If they reported in the Pain Barometer to be in chronic pain, the youth nurses could refer them for appropriate treatment (e.g. G.P., psychologist, or rehabilitation center). This seemed a feasible and effective strategy to early signal chronic pain in adolescents.

As described in **chapter 4**, home-based BMT by lay trainers is effective at the long term. Once its cost-effectiveness has been proven, it could be implemented in primary care on a large scale. However, availability of lay trainers might be an obstacle to implementation. The high mental load of providing BMT might be a reason to choose to deliver BMT at a low frequency. Additionally, the travel distance to the trainers' house should be relatively short, because a too large distance would make trainers more vulnerable to migraine attacks. Despite these obstacles, Mérelle and colleagues [118] reported that the lay trainers experienced the trainer role as emotionally rewarding.

5. CONCLUSION

In conclusion, this thesis shows that early signaling of chronic pain is important. Chronic pain is prevalent among adolescents and does not only negatively influence their quality of life, but also their academic performance. Adolescents who have experienced early adversities seem to be vulnerable to experience chronic pain. Signaling for these early adversities might prevent the development of chronic pain. Online self-management could well help to reduce these adolescents' pain, but so far has not got off to a good start. More research should be done on methods to improve adherence to online therapy. Since the patient-therapist relation seems important, strategies to strengthen this relationship should be studied. Home-based BMT provided by lay trainers seems to improve the self-management skills of adult migraine patients, which improvement is even sustained during long-term follow-up. Once its cost-effectiveness has been proven, BMT by lay trainers could be implemented at a large scale. This would give migraine patients greater access to therapy.

SUMMARY

Chronic pain is prevalent in both children and adults and has major negative consequences for their daily life, e.g. reduced participation in activities and depressive and anxious feelings. Therefore, it is important to early signal and treat chronic pain. This thesis aimed to provide answers to two important questions: 1. How to improve early signaling and assessment of chronic pain in adolescents? and 2. How to improve self-management of chronic pain in both adolescents and adults?

Psychosocial stress seems to serve as an important risk factor for the occurrence of pain. In **chapter 2** we examined if early adversities, e.g. bullying, abuse, and family conflict are risk factors for chronic pain in adolescents. Additionally, we described the pain characteristics of chronic pain in adolescents in a community sample of Dutch adolescents. Participants in the present study were 15,220 adolescents, attending schools (grade 7 and 8) in Rotterdam, the Netherlands. Chronic pain was measured with a newly developed questionnaire; the Pain Barometer. Early adversities were measured using single item questions from the Rotterdam Youth Monitor, a longitudinal youth health surveillance system. Cross-sectional associations between early adversities and chronic pain were investigated using logistic multilevel analysis, adjusted for potential confounding. In school year 2010-2011, 9.2% of the 15,220 adolescents reported chronic pain. Physical abuse by others (OR=1.51, 95% CI=1.07-2.14), sexual abuse (OR=1.46, 95% CI=1.05-2.05), family conflict (OR=1.79, 95% C=1.61-1.99), and being bullied (OR=1.23, 95% CI=1.17-1.29) are more common in adolescents with chronic pain. Physical abuse (OR=1.28, 95% CI=.95-1.71) by parents and parental divorce (OR=1.07, 95% CI=.93-1.22) were not significantly related to chronic pain. The results of the present study suggest that bullying, abuse, and family conflict may be risk factors for chronic pain. Early signaling these stressors might, prevent chronic pain.

In **chapter 3** we determined whether pain is associated with specific aspects of academic performance, i.e. lower grades, and with factors critical to an adolescent's academic performance, i.e. decreased emotional well-being and attention problems. We hypothesized that the association between pain and school grades is mediated by emotional well-being and attention problems. In a large cross-sectional study, we collected data from 2215 pupils, aged 12-13 years old. Pain, emotional well-being, and attention problems were reported by a self-rating scale. Dutch, English, and math grades were taken as an index of academic performance. Frequent pain in adolescents was associated with lower grades (Dutch p = .02 and math p = .01). Both occasional and frequent pain were associated with reduced emotional well-being (p =< .01) and reduced self-reported attention (p = <.01). Furthermore, the association between pain and lower grades was partially mediated by both emotional well-being and reduced self-reported attention (p-values = <.01). The results of the present study show that the association between pain and grades can be partially explained by both reduced emotional well-being and attention problems. The results suggest that an intervention targeted at subjective pain in adolescents could have a positive effect on emotional well-being and school performance.

Behavioral migraine approaches are effective in reducing headache attacks. Availability of treatment might be increased by using migraine patients as trainers. Therefore, Mérelle and colleagues

developed and evaluated a home-based behavioral management training (BMT) by lay trainers. The maintenance of effects at long-term follow-up are reported in **chapter 4**. Measurements were taken pre-BMT (T0), post-BMT (T1), at six-month follow-up (T2), and at long-term follow-up, i.e. two to four years after BMT (T3). Data of 127 participants were analyzed with longitudinal multi-level analyses. Short-term improvements in attack frequency and self-efficacy post-BMT were maintained at long-term follow-up (DT0T3 = -.34 and DT0T3 = .69, respectively). The level of internal control that increased during BMT decreased from post-BMT to long-term follow-up (DT0T3 = .18). Quality of life and migraine-related disability improved gradually over time (DT0T3 = .45 and DT0T3 = -.26, respectively). Although the results should be interpreted with caution because of the lack of a follow-up control group and the inability to gather information about additional treatments patients may have received during the follow-up period, the findings suggest that lay BMT for migraine may be beneficial over the long term. If so, this could make migraine treatments more widely available.

Chapter 5 describes a study protocol to early promote the self-management skills of adolescents with chronic pain, using an internet-based self-management program: Move It Now. This program includes cognitive behavioral techniques, including relaxation exercises and positive thinking. The objective of the intervention was to improve the ability of adolescents to cope with pain.

In **chapter 6** the effects of Move It Now were evaluated. Because of treatment attrition and loss to follow-up we could not apply the originally proposed between-group design but had to employ a within-group design, comparing the waiting list trajectory with the treatment trajectory. The Internet intervention consisted of seven weekly interactive Internet modules, which adolescents worked through independently. Additionally, a therapist weekly contacted them by e-mail or telephone. Adolescents (N = 69) were assessed on the outcome measures pain, coping, disability, catastrophizing, rewarding of pain behavior by parents, and quality of life. Measures were taken seven weeks before treatment, pre-treatment, post-treatment, and at three months follow-up. Multi-level modelling was used for longitudinal analysis of the data. Pain intensity, rewarding of pain behavior by parents, and sleep problems significantly decreased from pre-treatment to post-treatment in adolescents with chronic pain. Results for quality of life and pain coping were mixed. No significant differences were found for pain-related disability and pain catastrophizing. Contrary to expectations, an Internet-based self-management intervention for chronic pain is difficult to employ in adolescents, as it suffers from treatment attrition and loss to follow-up. We found indications that an Internet intervention has potential in decreasing pain, rewarding of pain behavior by parents, and sleep problems.

In conclusion, this thesis shows that early signaling of chronic pain is very important. Chronic pain is highly prevalent among adolescents and not only negatively influences their quality of life, but also their grades. Adolescents who have experienced early adversities seem to be more vulnerable to experience chronic pain. Signaling for these early adversities might prevent chronic pain. Online self-management may reduce pain in adolescent with chronic pain, but is difficult to employ in adolescents. In future, more research should be done to improve adherence to online therapy. Since the patient-therapist relation seems so important, strategies to strengthen this relationship should be studied. In adults,

home-based BMT provided by lay trainers seems to improve the self-management skills of migraine patients, even at long-term follow-up. If the cost-effectiveness has been proven, BMT by lay trainers could be implemented at large scale. This would make treatment for migraine patients more available.

SAMENVATTING

Chronische pijn komt veel voor onder zowel kinderen als volwassenen en heeft grote consequenties voor hun dagelijks leven, bijvoorbeeld een verminderde deelname aan activiteiten en depressieve en angstige gevoelens. Daarom is het belangrijk om chronische pijn vroegtijdig te signaleren en te behandelen. Deze thesis was erop gericht om een antwoord te formuleren op twee belangrijke vragen: 1. Hoe kan de vroegtijdig signalering en beoordeling van chronische pijn onder adolescenten verbeterd worden? 2. Hoe kan de zelfredzaamheid van adolescenten en volwassenen met chronische pijn verbeterd worden?

Chronische pijn komt veel voor onder adolescenten en heeft een negatieve invloed op hun kwaliteit van leven. Psychosociale stress lijkt een belangrijke risicofactor voor het ontwikkelen van pijn. In **hoofdstuk 2** hebben we onderzocht of vroege stressvolle levensgebeurtenissen, d.w.z. pesten, misbruik en conflicten binnen het gezin, risicofactoren zijn voor het ontstaan van chronische pijn. Daarnaast hebben we de prevalentie van chronische pijn geschat in een steekproef van Nederlandse adolescenten. Deelnemers aan deze studie waren 15.220 in Rotterdam naar school gaande adolescenten (eerste- en tweede klas middelbare school). Chronische pijn is gemeten met een nieuw ontwikkelde vragenlijst, genaamd de Pijn Barometer. Vroege stressvolle levensgebeurtenissen zijn gemeten met enkele vragen in de Jeugdmonitor Rotterdam, een longitudinaal volgsysteem voor de lichamelijke en psychische gezondheid van kinderen en adolescenten. Cross-sectionele verbanden tussen vroege stressvolle levensgebeurtenissen en chronische pijn zijn onderzocht met een multi-level analyse, waarbij gecorrigeerd is voor mogelijke confounding. In het schooljaar 2010-2011 rapporteerde 9,2% van de 15.220 adolescenten chronische pijn. Fysiek misbruik door anderen (OR = 1,51, 95% CI = 1,07-2,14), seksueel misbruik (OR = 1,46, 95% CI = 1,05-2,05), regelmatige ruzies tussen ouders (OR = 1,79, 95% CI = 1,61-1,99) en gepest worden (OR = 1,23, 95% CI = 1,17-1,29) komen meer voor onder adolescenten met chronische pijn. Fysiek misbruik door ouders (OR = 1,28, 95% CI = 0,95-1,71) en echtscheiding van ouders (OR = 1,07, 95% CI = 0,93-1,22) zijn niet significant gerelateerd aan chronische pijn. De resultaten van de huidige studie suggereren dat pesten, seksueel misbruik en ruzies tussen ouders risicofactoren kunnen zijn voor het ontstaan van chronische pijn. Het vroeg signaleren van deze stressvolle levensgebeurtenissen kan helpen om chronische pijn te voorkomen.

In **hoofdstuk 3** hebben we geprobeerd te bepalen of pijn gerelateerd is aan specifieke aspecten van academisch functioneren, d.w.z. lagere cijfers, en met factoren die belangrijk zijn voor het academisch functioneren van adolescenten, d.w.z. verminderd emotioneel welbevinden en aandachtsproblemen. Wij veronderstellen dat de relatie tussen pijn en school gemedieerd wordt door emotioneel welbevinden en aandachtsproblemen. In een grootschalige cross-sectionele studie hebben we data verzameld van 2.215 leerlingen, in de leeftijd van 12-13 jaar. Pijn, emotioneel welbevinden en aandachtsproblemen zijn in kaart gebracht door middel van zelfrapportage. Als een index voor het academisch functioneren is gekozen voor de cijfers Nederlands, Engels en wiskunde. Frequente pijn onder adolescenten was gerelateerd aan lagere cijfers (Nederlands $p = .02$ en wiskunde $p = .01$). Zowel de pijn die zich af en toe

voordoet en de frequente pijn waren gerelateerd aan een verminderd emotioneel welbevinden (p = <.01) en verminderde aandacht (p = <.01). De relatie tussen pijn en cijfers wordt gedeeltelijk gemedieerd door zowel emotioneel welbevinden als verminderde aandacht (p *waarden* = <.01). De resultaten van deze studie laten zien dat de relatie tussen pijn en cijfers gedeeltelijk verklaard kan worden door zowel verminderd emotioneel welbevinden en aandachtsproblemen. Hiermee kan gesuggereerd worden dat een interventie gericht op de subjectieve pijnbeleving van adolescenten een positief effect kan hebben op het emotioneel welbevinden en schoolprestatie.

Gedragstherapie is bewezen effectief in het verminderen van hoofpijnaanvallen bij patiënten met migraine. Beschikbaarheid van deze behandeling kan vergroot worden door patiënten met migraine te gebruiken als trainers. Daarom hebben Mérelle en collega's een vorm van gedragstraining ontwikkeld en geëvalueerd waarbij de training gegeven wordt door lekentrainers. De handhaving van de effecten hiervan op lange termijn worden gerapporteerd in **hoofdstuk 4**. Metingen vonden plaats voor de training (T0), na de training (T1), zes maanden na de training (T2) en op lange termijn, d.w.z. twee tot vier jaar na de training. Data van 127 deelnemers werd geanalyseerd met behulp van longitudinale multi-level analyse. Korte termijn verbeteringen in aanval frequentie en zelfeffectiviteit na de training werden gehandhaafd op lange termijn (DT0T3 = -.34 en DT0T3 = .69, respectievelijk). Het niveau van interne controle dat toenam tijdens de training nam af tussen het moment direct na de training en de lange termijn (DT0T3 = .18). Kwaliteit van leven en migraine gerelateerde beperkingen verbeterden geleidelijk over de tijd (DT0T3 = .45 en DT0T3 = -.26, respectievelijk). Hoewel de resultaten voorzichtig geïnterpreteerd moeten worden, omdat een controlegroep op lange termijn ontbrak en het niet mogelijk was informatie te verzamelen over aanvullende behandelingen die patiënten doorlopen hebben tijdens de follow-up periode, suggereren de bevindingen dat gedragstraining gegeven door lekentrainers positieve effecten heeft op de lange termijn. Als dit zo is, zou dit migraine behandeling breder beschikbaar kunnen maken.

Hoofdstuk 5 beschrijft een studie protocol om zelfmanagement vaardigheden van adolescenten met chronische pijn te bevorderen, door gebruik te maken van een zelfmanagement programma via het internet: Move It Now. De programma technieken vanuit de cognitieve gedragstherapie, waaronder ontspanningsoefeningen en positief denken. Het doel van de interventie was om het vermogen van adolescenten om om te gaan met de pijn te verbeteren.

In **hoofdstuk 6** zijn de effecten van Move It Now geëvalueerd. Vanwege uitval uit de behandeling en de metingen konden we het origineel voorgestelde between-group design niet toepassen, maar moesten we gebruik maken van een within-group design, waarbij het wachtlijst traject vergeleken is met het behandel traject. De internet interventie bestond uit zeven wekelijkse internet modules, die de adolescenten zelfstandig doorliepen. Daarbij, hadden ze wekelijks contact met een therapeut via e-mail of telefoon. Adolescenten (N = 69) werden onderzocht op de uitkomstmaten pijn, coping, beperkingen, catastroferen, beloning van pijngedrag door ouders en kwaliteit van leven. De metingen vonden plaats zeven weken voor behandeling, direct voor aanvang van de behandeling, direct na afloop van de behandeling en na drie maanden. Multi-level modelling is gebruikt voor een longitudinale analyse van

de data. Pijn intensiteit, beloning van pijngedrag door ouders en slaapproblemen namen significant af tijdens behandeling. De resultaten voor kwaliteit van leven en pijncoping waren niet eenduidig. Voor pijn gerelateerde beperkingen en catastroferen werden geen significante verschillen gevonden. In tegenstelling tot de verwachtingen, is een internet interventie niet zo bruikbaar onder adolescenten met chronische pijn, omdat er sprake is van veel uitval uit de behandeling en de metingen. We vonden wel aanwijzingen dat de internet interventie potentie heeft om pijn, belonend gedrag door ouders en slaapproblemen te verminderen.

Concluderend laat dit proefschrift zien dat het vroeg signaleren van chronische pijn erg belangrijk is. Chronische pijn komt veel voor onder adolescenten en heeft niet alleen een negatieve invloed op hun kwaliteit van leven, maar ook op hun schoolcijfers. Adolescenten die al op jonge leeftijd stressvolle gebeurtenissen meemaken zijn meer kwetsbaar voor het ontwikkelen van chronische pijn. Het vroeg signaleren van deze stressvolle gebeurtenissen kan helpen chronische pijn te voorkomen. Move It Now, een online zelfmanagement programma zou pijn reducerend werken, maar is moeilijk in praktijk te brengen onder jongeren. In de toekomst moet er meer onderzoek gedaan worden naar strategieën die de therapietrouw ten opzichte van online therapie kunnen vergroten. Gezien de relatie tussen patiënt en therapeut zo belangrijk is, moet gezocht worden naar strategieën die deze relatie kunnen versterken binnen online therapie. Voor volwassenen met migraine lijkt een zelfmanagement programma, aangeboden door leken trainers, effectief te zijn op de lange termijn. Als de kosten-effectiviteit van dit programma is aangetoond, kan het op grote schaal geïmplementeerd worden. Hiermee wordt het zelfmanagement programma voor migraine patiënten makkelijker beschikbaar.

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Streefkerk, juli 2015

PhD PORTFOLIO

Summary of PhD training and teaching

Name PhD student: Jessica Voerman	PhD period:	2010-2014
Erasmus MC Department: Psychiatry	Promotor(s):	Prof.dr. J. Passchier
Research School: NIHES		Prof.dr. J.J. van Busschbach
	Supervisor:	Dr. C. de Klerk

1. PhD training

	Year	Workload (Hours/ECTS)
General courses		
- Basiscursus didactiek, Erasmus MC, Rotterdam	2010	0.7 ECTS
- Masterclass Cost-efficient and optimal designs, Amsterdam, Nederland	2011	0.3 ECTS
- BROK ('Basiscursus Regelgeving Klinisch Onderzoek'), Erasmus MC, Rotterdam	2011	1.0 ECTS
- Minicursus Methodologie van patiëntgebonden onderzoek en voorbereiding van subsidieaanvragen, Erasmus MC, Rotterdam	2011	0.3 ECTS
- English Biomedical Writing Short course, Erasmus MC, Rotterdam	2012	1.0 ECTS
- Basistraining SCID I en SCID II	2013	2.0 ECTS
- Omgaan met lichamelijk onverklaarde klachten	2014-2015	2.0 ECTS
Specific courses (e.g. Research school, Medical Training)		
- Master Epidemiology, Netherlands Institute for Health Sciences (NIHES)	2011-2013	70 ECTS
Seminars and workshops		
- Workshop Grant Application, PhD Day, Erasmus MC, Rotterdam	2011	0.1 ECTS
- Workshop Expedition to your future, PhD Day, Erasmus MC, Rotterdam	2011	0.1 ECTS
- Workshop Feedback geven, Erasmus MC, Rotterdam	2013	0.1 ECTS
- Workshop Individuele begeleiding, Erasmus MC, Rotterdam	2013	0.1 ECTS
- Workshop Omgaan met groepen, Erasmus MC, Rotterdam	2013	0.1 ECTS
Presentations		
- Move It Now: Een internetbehandeling voor jongeren met onverklaarde chronische pijn (poster), Nederlands Congres Volksgezondheid, Amsterdam, Nederland	2011	1.0 ECTS
- Guided self-help via Internet for Adolescents with Chronic Pain: A study protocol (poster), 2de symposium Pediatrische Psychologie, Nijmegen, Nederland	2011	1.0 ECTS
- Long-term follow-up of behavioural attack prevention provided by lay trainers with migraine (poster), 7th Congress of the European Federation of IASP Chapters (EFIC), Hamburg, Germany	2011	1.5 ECTS
- Long-term follow-up of home-based behavioural management training provided by migraine patients (oral), First ARPH (Association for Researchers in Psychology and Health) conference, 2011, Lunteren, Nederland	2011	2.0 ECTS
- Guided self-help via Internet for Adolescents with Chronic Pain: A study protocol (poster), First ARPH conference, 2011, Lunteren, Nederland	2011	1.0 ECTS
- Disabling pain is associated with lower grades, attention problems, and reduced emotional well-being in adolescents (poster), 14th World congress on pain, Milan, Italy	2012	1.0 ECTS

- Disabling pain is associated with lower grades, attention problems, and reduced emotional well-being in adolescents (oral), Second ARPH conference, Enschede, Nederland	2013	2.0 ECTS
- Measurement of psychosocial factors in chronic pain patients referred to an academic pain center (poster), Congres Dutch Pain Society: Van multi naar meer, Ede, Nederland	2013	1.0 ECTS
- Disabling pain in adolescents is associated with lower grades (poster), Congres Dutch Pain Society: Van multi naar meer, Ede, Nederland	2013	1.0 ECTS
- Negative life events as predictors of chronic pain among Dutch adolescents (poster prize), Congres Dutch Pain Society: Van multi naar meer, Ede, Nederland	2013	1.0 ECTS
- Negative life events as predictors of chronic pain among Dutch adolescents (poster), 9th International Symposium on Pediatric Pain, Stockholm, Sweden	2013	1.0 ECTS
- Disabling pain in adolescents is associated with lower grades (poster), 9th International Symposium on Pediatric Pain, Stockholm, Sweden	2013	1.0 ECTS
(Inter)national conferences		
- Symposium Neuroimaging, Genetics, and Endophenotypes, Rotterdam, Nederland	2010	0.3 ECTS
- Wetenschappelijke dag Utrecht: Internettoepassingen voor de medische psychologie, Utrecht, Nederland	2010	0.3 ECTS
- Nederlands Congres Volksgezondheid, Amsterdam, Nederland	2011	0.7 ECTS
- Symposium Dutch working group eMental Health for Somatic Conditions (eHBM)	2011	0.1 ECTS
- 2de symposium Pediatrische Psychologie, Nijmegen, Nederland	2011	0.3 ECTS
- 1ste symposium NSRII (Netherlands Society for Research and Internet Interventions), Amsterdam, Nederland	2011	0.1 ECTS
- 7th Congress of the European Federation of IASP Chapters (EFIC), Hamburg, Germany	2011	1.0 ECTS
- First ARPH (Association for Researchers in Psychology and Health) conference, 2011, Lunteren, Nederland	2011	0.7 ECTS
- 2de symposium NSRII, Utrecht Nederland	2012	0.1 ECTS
- COLK- congres: Vallen en opstaan: de conversiestoornis te lijf, Alblasserdam, Nederland	2012	0.3 ECTS
- Second ARPH conference, Enschede, Nederland	2013	0.7 ECTS
- Congres Dutch Pain Society: Van multi naar meer, Ede, Nederland	2013	0.3 ECTS
- 9th International Symposium on Pediatric Pain, Stockholm, Sweden	2013	1.0 ECTS
- 8th Congress of the European Federation of IASP Chapters (EFIC), Florence, Italy	2013	1.0 ECTS
Other		
- Basiskwalificatie onderwijs (BKO)	2013	5 ECTS

2. Teaching

	Year	Workload (Hours/ECTS)
Lecturing		
- Communicatie en Attitude Onderwijs in het medische curriculum	2010-2014	17 ECTS
- Vaardigheidsonderwijs Onbegrepen Pijn, Rouwverwerking en Cognitieve Gedragstherapie	2011-2015	1.0 ECTS
- College Online hulpverlening tijdens keuzevak Effectieve bestanddelen psychotherapie	2011-2014	1.0 ECTS

- Tutor Minor Medische Psychologie	2013	2.0 ECTS
- Workshop tijdens Minor Medische Psychologie	2013-2014	0.5 ECTS
Supervising practicals and excursions, Tutoring		
- Begeleiding klinische stagiaires opleiding Neuropsychologie	2014	2.0 ECTS
Supervising Master's theses		
- Supervisor van de onderzoeksstage van Jeanine d'Anjou, student aan de Vrije Universiteit, Klinische Psychologie, in Amsterdam	2012-2013	2.0 ECTS
Other		
- Coördinatie en organisatie research bijeenkomsten sectie Medische Psychologie & Psychotherapie	2011-2014	5 ECTS

CURRICULUM VITAE

Jessica Stefanie Voerman was born on August 12th, 1987 in Dordrecht, the Netherlands. She graduated from secondary school (VWO, Wartburg College, location Guido de Brès, Rotterdam) in 2005. In 2009 she received her Master's degree in Clinical and Health Psychology at the University of Rotterdam, after completing her internship in a mental healthcare institution for addicted adults (GGZ). For her Master's thesis, she conducted a study on attentional biases and emotion recognition in anxious children, compared to non-anxious children. In 2010 she completed the Advanced Research program at the University of Rotterdam. During her study Psychology and the Advanced Research program she worked as a tutor at the Institute for Psychology, Erasmus University Rotterdam.

In July 2010, she began her PhD study as described in this thesis at the Department of Medical Psychology and Psychotherapy at the Erasmus University Medical Centre in Rotterdam. The main focus of her research is on the epidemiology of chronic pain in adolescents and adults and on psychological self-management programs for children and adults with complex chronic pain. During her PhD study, in 2013, she received her Master's degree in Epidemiology at the Netherlands Institute for Health Sciences, Rotterdam. In addition to research, she is involved in teaching medical psychology and communication skills in the medical curriculum at the Erasmus MC. In 2014 she received her Basic Qualification Education. Since 2012, she is working as a psychologist at the Department of Anesthesiology and Pain Management and since the summer of 2014 at the department of Neuropsychology

LIST OF PUBLICATIONS

Broeren, S., Muris, P., Bouwmeester, S., Field, A., **Voerman, J. S.** (2011) Processing biases for emotional faces in children: An exploratory study of developmental patterns and relationships with social anxiety and behavioral inhibition. Journal of Experimental Psychopathology, 2, 454-474.

Dommisse-van Berkel, A.A.M., van de Looij-Jansen, P.M., de Waart, F.G., **Voerman, J.S**., van Elderen, L.J, Passchier, J., de Graaf, L.E., Remerie, S., de Klerk, C. (2012) Signalering en doorverwijzing van adolescenten met chronische pijn door de jeugdgezondheidszorg: een pilotstudie. Tijdschrift voor Jeugdgezondheidszorg, 3, 56-59.

Voerman, J.S., Remerie, S., De Graaf, L.E., Van de Looij-Jansen, P.M, Westendorp, T., I., Van Elderen, De Waart, F.G., Passchier, J., Dommisse-van Berkel, A.A.M., De Klerk, C. (2012) Early signaling, referral, and treatment of adolescent chronic pain: A study protocol. BMC Pediatrics, 12:66.

Voerman, J.S., De Klerk, C., Mérelle, S.Y.M., Aartsen, E., Timman, R., Sorbi, M., Passchier, J. (2014) Long-term follow-up of home based behavioural management training provided by migraine patients. Cephalalgia, 34, 357-64.

Voerman, J.S., Van der Feen, B., Stronks, D.L. Diagnostiek van psychosociale aspecten in pijnbeleving en -gedrag van patiënten met chronische benigne pijn. In: Huygen, F.J.P.M., Van Kleef, M. Vissers, K.C.P. Zuurmond, W.W.A. (redactie), Handboek Pijngeneeskunde. De Tijdstroom uitgeverij BV, Utrecht.