Getting a grip on pain.

A model on pain and quality of life in adolescents with chronic pain.

The studies presented in this thesis were performed at the department of Medical Psychology and Psychotherapy (Netherlands Institute for Health Sciences) and the department of General Practice of the Erasmus MC Rotterdam, the Netherlands.
The studies presented in chapter 3 and 7 of this thesis were funded by a grant from the Algesiology foundation and the Pain Expertise Center, Erasmus MC Rotterdam, respectively.

Cover illustration: Carlijne Hagendijk (www.carlijneart.tk)

Printed and bound in the Netherlands by FEBODRUK BV - Enschede

ISBN 90-9018896-7

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A model on pain and quality of life in adolescents with chronic pain.

Greep krijgen op pijn.

Een model voor pijn en kwaliteit van leven van adolescenten met chronische pijnklachten.

Proefschrift

ter verkrijging van de graad van doctor
aan de Erasmus Universiteit Rotterdam
op gezag van de Rector Magnificus

Prof.dr. S.W.J. Lamberts
en volgens besluit van het College voor Promoties.

De openbare verdediging zal plaatsvinden op

woensdag 22 december 2004 om 11.45 uur

door

Vivian Petronella Benerdina Maria Merlijn

geboren te Hoorn.

Promotiecommissie

Promotoren:	Prof. dr. J. Passchier
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Ik heb een steen verlegd
in een rivier op aarde.
Nu weet ik dat ik nooit zal zijn vergeten.
Ik leverde bewijs van mijn bestaan,
omdat door het verleggen
van die ene steen
de stroom nooit meer
dezelfde weg zal gaan.

B. Vermeulen

Voor mijn ouders

Ze zaten allemaal voor het huis, onder een grote boom, en de vader zei: ´Nu zal ik jullie allemaal op de proef stellen, en zien wat je nu eigenlijk geleerd hebt´.

De vier volleerde broers. Sprookjes van Grimm.

Content

Chapter 1

General introduction

Prologue

Chronic pain in children and adolescents is increasingly recognized to occur. It often results in considerable functional disability and is a cause for health services utilisation. Chronic pain is known to have substantial impact on the quality of life of adolescents and their families. The maintenance of chronic pain in the absence of a defined organic diagnosis has led researchers to seek for psychosocial explanations. This thesis evaluates a model on the adjustment of adolescents to chronic pain, in which psychosocial factors are incorporated. Examined are two issues in relation to this model. The first issue concerns the influence of psychosocial factors on chronic pain and quality of life of adolescents. The second issue concerns the evaluation of a cognitive-behavioral intervention program for adolescents with chronic pain. Before presenting studies on these issues, this introductory chapter starts with a description of the background of these studies. After presenting information on chronic pain and quality of life in adolescents, the reader is acquainted with the issue of psychosocial factors related to chronic pain and quality of life. Additionally, the background of the cognitive-behavioral intervention program is addressed. Finally, an overview of the various chapters in this thesis is presented.

The prevalence of chronic pain

The International Association for the Study of Pain (IASP) defines pain as: "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". Pain that exists three months or longer, continuously or recurrently, is in children and adolescents considered as chronic pain.[1,2,3] The problem of chronic pain in adolescents is widespread, and research on specific pain conditions has indicated that there are a variety of chronic pain conditions that occur in adolescence. Most epidemiological studies investigating pediatric pain have addressed a specific pain condition rather than providing a comprehensive analysis of pain problems.[1] Recent research on pediatric pain irrespective of location has focused on longitudinal, population-based studies using standardized assessment techniques. A Dutch community sample of 6,636 children and adolescents (0-18 years) shows that approximately 25% of the respondents experience chronic pain (recurrent or continuous) existing for more than 3 months.[2] Chronic pain increases in adolescence[2,4-8], with girls reporting more frequent pain than boys, irrespective of their pain location.[5,8-13] The most common types of pain in adolescents are limb pain, headache and back pain.[2,14,15] Longitudinal studies showed that chronic pain is persistent at 1-year follow-up in about 50% of the children and adolescents who report chronic pain at baseline.[16,17] After two years still a third of the children and adolescents report persistent pain.[17] In addition, pain remains stable across the assessments, in other words it does generally not deteriorate with time.[4,17,18] Considering the prevalence and natural course of (back) pain in adolescents, Burton and colleagues suggest that much of the symptomatology may be considered as a normal life experience.[4]

The impact of chronic pain

Due to the high prevalence of chronic pain much research has focused on its impact on adolescents. Chronic pain is often associated with complex social and psychological problems and it has been shown to lead to school absenteeism, nuisance in the adolescents' life,[19,20] reduced quality of life[21-23] and considerable medication use.[24-26] Population-based studies showed that substantial proportions of adolescents use medications for headache and stomach-ache.[25] This medication use among adolescents has increased over the years in particular regarding medicine for stomach-ache among 13- and 15 year old girls.[26] A prospective study on children and adolescents with chronic pain showed that more than half (53%) used medications for their pain.[24] Considering these increasing proportions of users, it is suggested that more information about medicine should be built into health education programs.[25,26]

Quality of life

The impact of a chronic condition like chronic pain affects various aspects of a person's well-being, such as physical, psychological and social life. These aspects are covered in the concept Quality of life (QoL). Quality of life is a multidimensional construct including a persons' physical health, psychological state, level of independence, social relationships, personal beliefs and their relationships to features of the environment.[27] Quality of life assessment encompasses subjective evaluations of the effects of the condition and its treatment in relation to personal goals, values and expectations.

Quality of life instruments

In recent years quality of life measures for children and adolescents have been developed, initially for children with specific chronic illnesses, but more recently generic and health-related QoL measures have become available. Validated disease-specific instruments are available for adolescents with asthma, cancer, epilepsy and juvenile arthritis.[28] Generic measures are useful in situations where it is important to be able to measure quality of life across different conditions and across different cultures. The generic Child Health Questionnaire (CHQ) measures quality of life and children's functioning across 14 health and well-being domains. The CHQ has been normed with children experiencing a range of chronic health conditions (e.g. epilepsy, juvenile rheumatoid arthritis). However, generic quality of life may not be sensitive to small changes in a child's functioning or to disease-specific functions. Consequently, some measures are currently being developed which incorporate both a generic and a disease-specific element.[29] The Quality of Life Headache-Youth (QLH-Y), developed by Langeveld and colleagues,[30] is an example. Langeveld and colleagues operationalized QoL in the following domains: psychological functioning, functional status, physical functioning, social functioning, satisfaction of life and satisfaction of health. Functional status contains disease specific items to measure the level of impediment caused by headache and migraine symptoms. These items were based on interviews with headache patients. The other five domains address generic elements of quality of life.

Previous studies

Studies using this (QLH-Y) measure showed that adolescents with chronic headache evaluate their quality of life as being less satisfactory than their healthy peers.[21,30] Compared to healthy controls, headache sufferers report significantly worse psychological functioning, physical functioning, functional status, social functioning and diminished satisfaction with life in general and with their health status.[30,31] The quality of life of adolescents with chronic pain changes when their pain changes.[22,23] Adolescents with intense pain score worse on functional status and psychological functioning than those with milder pain.[30,31] Bandell-Hoekstra and colleagues[21] reported that adolescents with intense pain also experience more problems with physical functioning and social functioning at home.

Different pain locations

Hunfeld and colleagues[22] studied the relative impact of different pain locations on quality of life. In this study headache and back pain were associated with lower quality of life than abdominal pain and limb pain, even though the frequency and intensity of the pain are lower for headache than for the other pain locations. On the other hand, Kashikar-Zuck and colleagues[32] found that children and adolescents with musculoskeletal pain report higher levels of disability and more difficulty coping than the chronic daily headache group. A three-year follow up study showed that QoL and the impact of pain on the adolescents and their family remained stable across assessments.[19]

Depth-interviews showed that adolescents with headache report specific problems with cognitive activities, whereas those with limb pain and back pain report problems with physical activities.[19] Although chronic pain in adolescents has great impact on various aspects of quality of life and daily functioning, few instruments are available to measure the pain-related problems irrespective of pain location. Available measures are the PedMIDAS[33] and the Functional Disability Inventory (FDI)[34] but these measures are only validated for children and adolescents with headache and recurrent abdominal pain, respectively. Moreover, Walker and Green[34] studied children and adolescents from a pediatric outpatient clinic and Hershey[33] and colleagues studied patients from a tertiary referral center. No measures are available for children and adolescents in the general population and with pain other than headache or abdominal pain. Therefore, this thesis also addresses the development of a pain-related problem list suitable for adolescents from the general population with chronic pain irrespective of their pain location.

Psychosocial factors related to chronic pain

To understand underlying psychological mechanisms of chronic pain in adolescents, initiating and perpetuating factors have been studied more closely recently. A number of psychosocial factors have been associated with the onset, exacerbation, and/or maintenance of chronic pain in adolescents. Gragg and colleagues[35] showed that psychosocial factors may be better predictors of children's pain rating than medical factors. Their study showed that disease activity only partly explains the variation in the pain experience and pain perception of children with juvenile idiopathic arthritis.

Adolescent factors

Psychological vulnerability

The most frequently studied factor is psychological vulnerability. This is a trait-like tendency to experience a broad range of negative feelings such as distress, worry, inadequacy, anxiety and depressive symptoms (gloominess). Children and adolescents who present for treatment of chronic pain, independent of medical diagnosis, report anxious[36], and depressive[32,37] symptomatology. Comparisons with controls show that recurrent headache is related to stress and psychological symptoms in schoolchildren,[5] and to psychological distress[38] and anxiety[39] in adolescents. A high level of anxiety was also found for children with abdominal pain.[40,41] Prevalence of depression symptoms is higher among adolescents with recurrent pain[42] and especially among girls with neck and shoulder pain.[43]

Children complaining of headache showed a tendency towards high achievement motivation at school[44] and reacted with somatic symptoms during stress situations.[45] The association of stress in school with headache was strongest in girls.[46] Negative fear of failure (the fear to fail which results in not optimal functioning of the child) can be considered as a specific aspect of psychological vulnerability. Levine et al.[47] found that perceived failure leads to negative affectivity and an increase in pain report. Similarly, Passchier and Orlebeke[48] found that negative fear of failure and headache complaints in children and adolescents are positively correlated.

Although an association between psychological vulnerability and pain has been found in correlational research, the direction of a (causal) relation is still unclear. Do adolescents develop these characteristics as a result of frequent pain or is the experience of chronic pain a consequence of psychological vulnerability? Adult studies suggest that emotional problems (as depression symptoms) are more likely to be a consequence than a cause of chronic pain[49-52] other studies indicate that they may share common underlying etiologic factors[53,54]. Further research is needed to disentangle this relation.

Coping

Coping refers to intentional cognitive and behavioral strategies used to manage stressful situations.[55] Chronic pain can be a stressful experience for children and adolescents that have to be dealt with.[20,56] Gil and colleagues[57,58] found that children's use of coping strategies that involved a high level of negative thinking and self-isolation was associated with more pain intensity, functional disability and more health care use.

Children with chronic pain are more likely to use distraction (e.g. reading, playing, listening to music) and social support (ask a friend for help).[55,59,60] A Dutch study of children and adolescents with chronic headache showed that those with severe headaches seek more social support, are more likely to internalize and externalize, they use less behavioral and cognitive distraction techniques, and seek information less than those with mild pain.[21] Palliative, avoidance, and depressive coping are related to higher pain intensities and longer headache duration in adolescent males.[61] A review study of Jensen and colleagues[62] showed that coping is strongly related to adjustment to chronic pain. Patients who believe they can control their pain, who avoid catastrophizing, and who believe that they are not severely disabled function better than those who do not.[62]

Reid and colleagues found that more approach coping (i.e. direct attempts to deal with pain and the use of active methods to regulate feelings when in pain) was related to less disability in children with headache.[63] Walker and colleagues found that passive coping (i.e. catastrophizing) was associated with higher levels of pain and functional disability across samples of schoolchildren, patients with abdominal pain, and those who formerly had abdominal pain.[64] It is important, however, to note that much of the current data are correlational. Again, cause and effect are not always clear.

Social environment factors: parents and peers

Modeling

Modeling refers to the mechanism that the behavior a child expresses might be copied from someone else. Significant others with pain (e.g. parents) can serve as models on how to cope with pain. Children might copy the pain behavior of their parents. The role of family pain models in the etiology of pain behaviors is emphasized by various researchers.[65,66]

Parents with chronic pain

Results of several studies suggest that children of parents with chronic pain may be at risk for illness behavior. These children reported to have more frequent pain, to use more medication and to miss more school because of illness than children of healthy parents.[67,68] Further, they showed higher concern about their health status.[69]

Compared to healthy controls children and adolescents with recurrent pain are more likely to have parents with frequent pain.[70-72] Chronic pain is more frequently reported among children of chronic pain patients compared to children of healthy people [68,59,73-75].

Number of pain models

There is also increasing evidence for the positive relation between the number of pain models (people with pain in the environment of the pain patient) in an individuals' environment and frequency of his/her pain reports.[76] Additionally, Osborne[77] found that the number of pain models was higher in families with unexplained pain than in families with medically explained pain complaints. Moreover similar pain locations were found in both the child and the pain model. Maternal modeling of pain behavior (in contrast to paternal modeling) had a particularly strong influence on children's pain[78] and their coping efforts[79]. Pain models have greater impact on females than males.[78] Combined these studies support the clinical notion that pain runs in families, but they do not provide causal evidence for this association.

Laboratory-based studies

Laboratory-based experiments provide more direct evidence that modeling has an impact on what people do when they have pain. Thastum and colleagues[80,81] examined the relation between child pain and parental pain during a cold pressor task. Their studies compared the pain threshold, pain tolerance, mean pain intensity and pain coping strategies reported by 9- to 15-year-old children (healthy and those with juvenile rheumatoid arthritis) and their parent. Moderate correlations were observed between children and their parents on pain intensity, pain tolerance and pain coping strategies. Goodman and McGrath[82] found some support for a causal association with their laboratory study. They assigned 96 healthy children and their mothers to one of three experimental groups. Mothers assigned to the exaggerate condition were instructed, via videotape, to slightly exaggerate their display of pain and indicate a pain threshold within the first 10-20 sec of a 4-min cold pressor task. Mothers in the minimize condition were instructed to minimize their display of pain, while mothers in the control group were given no specific instructions. After observing the mother completing the cold pressor task, the child started the experimental trial. Children assigned to the exaggerate condition reported significantly lower pain thresholds and children in the minimize group showed less pain-related facial behavior compared to children in the control group. This study showed that observing the behavior of their mothers during an experimental pain task directly influences the pain behavior of children.

Reinforcement by parents

Children's pain may be rewarded inadvertently when they are allowed to remain at home instead of attending school, encouraged to withdraw from potentially stressful sports or social situations, and relieved from routine responsibilities. These secondary gains may prolong pain episodes or initiate new episodes. Like modeling, reinforcement is a process often implicated in the development of complex pain behavior patterns, but in chronic pain patients only its outcome can be measured.

Laboratory-based studies

Laboratory studies allow the possibility of using normal subjects who have no pain conditions with established reinforcement contingencies. Recently, Jolliffe and Nicholas[83] conducted a study in which a blood pressure cuff was applied to healthy undergraduate students in order to induce a pain sensation. After each of the fifteen trials the students were asked to mark the experienced pain on a Visual Analogue Scale. Half of the students received positive verbal reinforcement (such as 'very good') if their report of pain intensity following the inflation of a blood pressure cuff exceeded that of the previous trial. If they reported the pain intensity as being lower than the previous trial negative statements (such as 'that's strange' or 'this does not look too good') were made. The other students did not receive any verbal reinforcement after they reported their pain intensity. Mean pain reports of reinforced subjects were greater than those of the non-reinforced subjects both when the pain intensity was stable over trials and when it decreased, suggesting that operant techniques play a role in pain reports. Comparisons between chronic back pain patients and healthy controls show that chronic pain sufferers are more easily influenced by operant conditioning factors which may add to the maintenance of the pain problem.[84] Flor and colleagues[84] recorded EEG, EOG, heart rate, skin conductance and muscle tension levels of chronic back pain patients and healthy controls in an experimental study. Pain threshold and pain tolerance for the arm and back muscles were individually determined in series of electric stimuli (baseline rating). Both groups received electric stimuli of different intensities (pain threshold, 25, 50 and 75% of the distance between pain threshold and tolerance). After shock delivery, subjects rated the intensity of the electric stimulus on a visual analogue scale provided on a computer screen. Subjects were given positive feedback (smiling face at the computer screen and monetary gain) when their actual pain rating was higher than the average rating in the baseline rating, lower ratings were followed by negative feedback (sad face on the computer screen and monetary loss). Neutral feedback was provided when the pain rating was equal to the baseline rating. Both controls and chronic back pain patients showed similar learning rates, i.e. higher pain ratings after reinforcement. Following the operant conditioning of verbal pain reports, chronic back pain sufferers displayed slower extinction of verbal and cortical pain response and displayed prolonged elevated electromyogram levels compared to controls.

Clinical practice and observational studies

In clinical practice, the impact of parental responses on adolescents pain coping was demonstrated by Dunn-Geier and colleagues[85]. Exploring the mother-child interaction of patients who where judged to be coping well and patients who were not (those who missed monthly 3 or more days of school because of pain), they found that mothers of poorly coping adolescents frequently discouraged adaptation with negative statements such as 'Doesn't it hurt?' or 'You must be exhausted'. Simply reframing these questions to neutral interactions 'How are you feeling now' or to verbally encourage coping behavior ('That's good, you're trying very hard') provides a more positive coping climate. Additionally, non-coping adolescents engaged in significantly more negative behavior ('I quit', 'It is too hard') and tended to express more pain and be on-task less often as compared to coping adolescents in a pain-oriented situation. These findings support Fordyce's contention that non-coping chronic pain patients exhibit behaviors that signify that they are experiencing pain to those around them.[86] Other studies showed higher rewarding for illness behavior in children with recurrent abdominal pain than healthy controls.[87] On the other hand, another study indicates that parents of children with chronic pain often neglect the pain of their child because they belief that their child complains of pain in order to avoid aversive or non-preferred activity.[19] Also Engel[88] found that mothers often ignored their children's pain complaints (85% of children between 12 and 17 years). Osterhaus[89] showed that parental rewarding was related to less headache activity of the child. Considering these contradicting results, the process of parental rewarding is complex and difficult to unravel. The results of these prior studies might be explained by another aspect of rewarding: they may be the result of an intermittent reinforcement pattern, as suggested by McGrath.[90] Some parents respond inconsistently to children's pain complaints, either providing excessive emotional and physical support or indicating that they do not have time to assist their children. The implicit message that children receive is that their pain complaints need to be stronger if they are going to convince their parents that they need the same level of support that parents have provided in the past. The children gradually learn to exaggerate their complaints or develop new symptoms to obtain their parents' attention.

Reinforcement by peers

Harris[91] argues that peer relationships are the chief determinants of personality development. In adolescence, peers have a major influence on the social and emotional functioning of adolescents.[92] Socially, adolescence is marked by an increasing independence from parents as they spend more time with peers and turn to peers for social support and identification.[93] Although adolescents tend to become more peer-oriented and less parent-oriented as they get older, this is not the case in every situation. Parents were found to remain more influential than peers in areas such as educational plans, while peers have influence on choice of clothes and use of leisure time.[94] Studies showed that girls exhibited greater emotional self-disclosure to parents and peers than did boys, and that emotional self-disclosure to friends was greatest among older adolescents. In addition, while younger adolescents preferred to disclose information about their emotional state to parents, older adolescents chose friends.[95] These studies support the importance of involving (reactions of) peers in the social context of adolescents with chronic pain. So far, it has been shown that children with headache reported more often being bullied in school, having problems in getting along with other children than children without headache[46] and having fewer peer relations than healthy controls.[96] Adolescents with chronic headache were often found to keep the fact that they had a headache to themselves.[88]

Psychological theories

From a traditional biomedical perspective, pain has been conceptualized as the direct result of tissue damage or injury. Consequently, management of pain consisted of recognizing and localizing the underlying pathology and of medical treatment to remove or cure this pathology.[97] Biomedical findings did not provide sufficient explanations for the varying extent of pain-related disability in daily functioning of chronic pain sufferers. Some patients appear not to be disabled by extensive damage and pain, whereas others respond with extensive disability to seemingly minor damage and pain.[98] In some patients the extent of damage does not refer well to the experience of pain.[99] These people report pain that has no identifiable lesion. In other patients abnormalities are found in the MRI's (magnetic resonance imaging) or CAT-scans while they do not experience back pain symptoms.[100,101] This implies that abnormalities or tissue damage are not always related to the discomfort of pain symptoms. Pain is more complex than tissue damage alone. Therefore, pain researchers developed new experimental approaches that have increased our understanding of basic pain processes and have led to the development of new theoretical models of pain. The bio-psycho-social model considers pain as a complex phenomenon caused or influenced by an interaction of biological, psychological and social factors. Pain can be understood by not only considering underlying tissue damage, but also by taking into account psychological and social factors. The increasing knowledge on psychosocial factors influencing pain experiences, has led to the development of psychological programs for managing chronic pain. Frequently evaluated interventions mostly address principles of cognitive-behavioral therapy. These principles are based on two major psychological theories; the behavioral and social learning theory and cognitive theories.

Behavioral and social learning theory

Behavior learning theory and social learning theory address the notion that pain behaviors develop and persist as a result of learning. The behavioral learning theory discriminates two main forms of learning: operant and respondent conditioning.

Behavioral learning theory

Operant conditioning suggests that the reinforcement of pain behaviours (e.g. moaning, inactivity or medication intake) leads to the maintenance of that behavior. Pain behaviors followed by pleasurable events or by removal of negative events (positive and negative reinforcement) increase while pain behaviors followed by negative events or by the absence of a rewarding (punishment or extinction) decrease.[102]

Respondent conditioning suggests that pain is an end result of a pain-tension cycle: elevated levels of muscle tension (arise through conditioning in stressful events) can lead to pain sensations. Subsequently, these unpleasant feelings evoke tension.
Anticipated fear considering certain movements that are associated with pain can lead to a conditioned reaction. Pain as a result of injury is often associated with fear to expose the painful region to external stimuli. Because of this fear, patients are inclined to avoid certain movements. When the avoidance of movements is maintained after a normal healing time for the injury, avoidance learning is developed. The resulting inactivity often leads to a deterioration of the physical condition and new health problems.

Social learning theory

Social learning theory emphasizes that learning does not require direct experience with reward and punishments. The observation of the consequences of others' behavior can be as powerful. Bandura[103] has demonstrated that observational vicarious learning (modeling) is one of the major modes of acquisition of new behavior. This social modeling of behavior may aid children in learning about pain behaviors and the consequences of such behaviors if they have a parent who also suffers from frequent pain complaints and especially when the child observes the model being reinforced for pain behavior.[104]

Behavioral treatment of chronic pain

The purpose of pain management based on learning theories is changing one's learned pain behavior (e.g. complaining of pain, grimacing or time spent in bed) into healthy behavior. Behavioral methods in pain management include operant techniques (changing reinforcement schemata by verbal reinforcement of healthy behavior) and respondent techniques (systematic desensitisation, relaxation techniques). A way to change reinforcement schemata in daily life of the chronic pain sufferer is to involve their parents or peers in the intervention. In stead of reinforcing pain behaviors parents and peers are encouraged to support healthy behavior. Considering social learning techniques, a therapist or another group member could model healthy behavior (and show positive consequences of this healthy behavior) during role-play or group discussions. As far as we know, no pain management programs are available that ask parents or partners (of pain patients) to change their own pain coping strategies in order to model healthy pain coping strategies to their child or spouse in pain.

Cognitive theory

The cognitive theory is concerned with attention processes around pain and the effect of assumptions and beliefs about pain on an individual's coping with pain. The way an individual perceives events depends on the basic assumptions or beliefs about himself and others.[105] Two cognitive processes play a role in the maintenance of chronic pain: attributions and self-efficacy beliefs. Attributions refer to the meaning a person gives to pain and determines the coping strategies of pain patients. Self-efficacy beliefs are mainly determined by consistent factors. Successful coping leads to an increase of self-efficacy beliefs (belief in one's own ability to manage pain). Self-efficacy beliefs are an important determinant of pain behaviors and disability associated with pain.[106]

Cognitive treatment of chronic pain

Cognitive methods in pain management include distraction (taking the attention away from the painful situation), thought-stopping (this interferes with recurrent negative ruminations or obsessions), positive self-talk and transformation (refers to the attempt to change the evaluation of an event or sensation from being awful to being irritating or annoying).

Pain management programs

Table 1 presents the content of available pain management programs for children and adolescents. Most of these programs consist of (a combination of) one of the following cognitive-behavioral techniques:

Respondent conditioning

Relaxation techniques: various relaxation techniques are taught to use at early signs of symptoms to reduce muscle tension and level of arousal. Separate muscles-groups are being systematically tightened and relaxed in progressive relaxation. Cue-controlled relaxation is based on a simple word that patients associate with deep breathing (e.g. children are instructed to engage in deep breathing practice 'when the school bell rings'). Autogenic relaxation techniques require visual perceptions of relaxed situations.

Biofeedback: is most often applied to headache patients. In biofeedback, patients are assisted in relaxing the muscles of neck, face and head by means of registrations of electromyogram. Thermistors are attached to the patient's fingers and/or frontal muscle to register bodily reactions (muscle tension, skin temperature, and heartbeat). If the patient succeeds in reducing body tension, skin temperature goes up.

Operant conditioning

Positive reinforcement: during treatment healthy behavior is positively reinforced. Because of individual differences, it is important to gather insight in reinforcing factors that can be used during treatment for each patient individually. Examples of universal reinforcers are: a pat on the back or nodding approvingly. To generalize the healthy behavior to daily functioning, some programs provide parents with guidelines in order to learn them to recognize and reinforce healthy behavior.[106-108]

Extinction: Concentrating one's attention (positive reinforcement) on healthy behavior brings about less attention for pain behavior. The absence of positive reinforcement for pain behavior causes extinction of that pain behavior.

Shaping: End goals (e.g. walking to the supermarket: 15 minutes) are formulated in terms of functional behaviors that are categorized in sub-goals. Methods of graded activity are used to shape the functional behavior from baseline level (not able to walk) to end goal (walking for 15 minutes).

Cognitive techniques

Psycho-education: irrational beliefs about pain ("an activity that brings more pain is harmful for my body") are corrected by psycho-education.

Rational emotive therapy: the goal is to help individuals to obtain more effective behavior by assisting them to change their irrational beliefs into more rational ones. Pain patients are asked to complete ABC-schemes to become aware of the chain of behavior and any irrational thoughts one might have. This scheme produces a view on A: a pain situation; B: the personal view on this, cognitions and C: following emotional reactions and behavioral consequences. This ABC-scheme helps pain patients learn to recognize and understand that thoughts based on beliefs influence emotions and behavior and patients learn to identify their irrational (pain) beliefs. To change these irrational beliefs they need to be challenged or discussed and patients are requested to formulate more rational beliefs.[105]

Format

Self-management versus therapist guided

To facilitate the applicability of clinically-based treatments, several psychological interventions have a self-administered format[110] but they are limited to a single pain complaint. Self-management programs have been shown to be as effective as psychological treatment guided by a therapist, but less expensive.[111-113]

Patient alone versus including social environment

Most psychological interventions involve the chronic pain sufferer only. From a behavioral point of view, Fordyce and colleagues.[114] have pointed out the necessity of including the family in treatment. Although only a few treatments involve the parents of children and adolescents with chronic pain, the results are promising.[107-109] For example, parents rated treatments in which they were involved as more satisfying and effective.[107,109] These experiences were supported by clinical improvements in pain severity, pain frequency[107,109] and reduction of parental stress[108].

Effectiveness of psychological pain programs

Recently, two systematic reviews showed that psychological treatments based on the principles of cognitive-behavioral therapy are effective in reducing the severity and frequency of chronic headache[1] and recurrent abdominal pain[115] in children and adolescents. Few psychological treatments for adolescents with chronic pain other than headache have been evaluated in a randomized controlled study design. Significant reductions in pain and improvements in functioning were achieved in children and adolescents with recurrent abdominal pain[110] and complex regional pain syndromes[116]. Despite the benefits with use of cognitive-behavioral intervention strategies, no studies are available that have a primary focus on enhancing children's functioning.[20] The fact that in most studies pain relief is the primary outcome for evaluating treatment efficacy might be a result of the continuing influence of the biomedical perception.[1]

Table 1: Effect studies on psychological treatments for children and adolescents with chronic pain.

Authors	Sample source	Design	Intervention
Allen and Shriver[107]	Clinical sample of 27 children (7-18 yr) with migraine headaches	Case-control study	*Thermal biofeedback alone*: clinic and home practice of biofeedback *Thermal biofeedback plus behavior management guidelines*: parent-mediated pain-management guidelines to reduce pain behaviors
Bandell et al.[119]	General population (school) sample of 158 children and adolescents (9-17 yr) with headache	Randomized controlled trial	*Self-help (cognitive behavioral) training (n=76)*: encompassing relaxation training, cognitive techniques, assertiveness and problem solving. 7 weeks with manual, telephone contact once every two weeks. *Control group (n=82)*: self-management placebo training without cognitive and relaxation training. 7 weeks with manual and telephone contact once every two weeks.
Barry and von Baeyer[120]	Clinical sample of 36 children (7-12 yr) with headache	Randomized controlled trial	*Cognitive therapy (n=18)*: two sessions (90 min.) for children and two for their parents. Incorporating relaxation techniques, distraction, visualization, stress-management skills. *Waiting list control (n=18)*: no treatment
Eccleston et al. [108]	Clinical sample of 57 adolescents (11-18 yr) with chronic pain	Case-report	*Interdisciplinary cognitive behavioral therapy (CBT) (n=57)*: incorporating physical therapy, education on pain, activity encouragement and cognitive techniques (total 110 hours). Three week residential programme of group cognitive behavioral therapy (including parents in treatment)
Finney et al. [121]	Clinical sample of 32 children (4-18 yr) with recurrent abdominal pain.	Case-control study	*CBT (n=16)*: encompassing self-monitoring, reinforcement of non-illness behavior, relaxation training, dietary giber, and required school attendance *Control group (n=16):* no treatment to assess changes in health care use
Griffiths and Martin[111]	General population (newspaper) sample of 51 children (10-12 yr) with headache	Randomized controlled trial	*Clinical-based CBT (n=17)*: incorporating education, progressive relaxation, breathing exercises, autogenic relaxation training, mental imagery, cue-controlled relaxation and cognitive skills. Eight sessions of 90 minutes weekly. *Home-based CBT (n=17):* 3 clinical sessions alternated with 5 self-management weeks. Intervention is similar to the clinical-based CBT. *Waiting list control (n=17)*: no treatment
Larsson and Melin[122]	General population (school) of 31 adolescents (16-18 yr) with headache (tension and tension combined with migraine headache)	Randomized controlled trial	*Relaxation training (n=11)*: incorporating progressive relaxation training and cue-controlled relaxation. 9 sessions at school, twice a week for 5 weeks *Information-contact (n=13)*: education on pain-facts; prevalence and causes; no suggestions were made to change the situation. 9 sessions at school, twice a week of 5 weeks *Self-registration (n=7)*: no treatment
Larsson et al. [112]	General population (school) of 41 adolescents (16-18 yr) with tension-type headache	Randomized controlled trial	*Self-help relaxation (n=16)*: progressive relaxation, cue-controlled relaxation techniques. 5 audiotapes of 5-10 minutes. *Therapist assisted relaxation (n=14)*: about 7 sessions of 45 minutes; progressive relaxation and cue-controlled relaxation. *Self-monitoring (n=11)*: no treatment
Larsson and Carlsson[123]	General population (school) of 26 adolescents (10-15 yr) with tension-type headache	Randomized controlled trial	*Relaxation training (n=13)*: progressive relaxation, cue-controlled relaxation. 20 min sessions, twice a week for 5 weeks *Self-monitoring (n=13)*: no treatment

Authors	Sample source	Design	Intervention
McGrath et al. [113]	Clinical sample of 87 adolescents (11-18 yr) with migraine	Randomized controlled trial	*Self-help CBT (n=30)*: cognitive restructuring, relaxation techniques, identify unrealistic beliefs, distraction, imagery, assertiveness, problem solving and mental activities. 8 weeks manual and cassette tapes (weekly telephone contact) *Therapist-assisted CBT (n=29)*: Intervention program is similar to the self-help CBT. 8 sessions once a week on a individual basis *Control group (n=28)*: placebo intervention without cognitive and relaxation training; list of common triggers that can cause migraine (f.i. different food, too much sun). 8 weeks manual and weekly telephone contact to monitor progress
Osterhaus et al. [124]	General population (school) sample of 41(12-19 yr) with headache (TTH/migraine)	Randomized controlled trial	*CBT group (n=32):* education, relaxation training (progressive and autogenic), temp. feedback and cognitive training (RET). Eight weekly sessions (4 group sessions of 90 minutes and 4 individual sessions of 45 minutes) *Waiting list control (n=9)*: no treatment
Osterhaus et al. [125]	General population (local newspaper) sample of 39 adolescents (12-22 yr) with headache (TTH/migraine)	Randomized controlled trial	*CBT group (n=24):* education, relaxation training (progressive and autogenic), temp. feedback and cognitive training (RET). Eight weekly sessions (4 group sessions of 90 minutes and 4 individual sessions of 50 minutes) *Waiting list control (n=15)*: no treatment
Passchier et al. [126]	General population (school) sample of 202 adolescents (Mean = 13.7 yr) with weekly headache	Randomized controlled trial	*Teacher presented training (n=110)*: progressive relaxation training; 10 lessons (10-20 minutes) *Control group (n=92):* placebo intervention incorporating physical concentration exercises: 10 lessons (10-20 minutes).
Sanders et al. [127]	Clinical and general population sample of 16 children (6-12 yr) with recurrent abdominal pain	Randomized controlled trial	*CBT*: 6 sessions (n=8): 4 for the children (encompassing progressive muscular relaxation, cognitive restructuring, positive self-verbalization, distraction, problem solving) and 2 for their parents (encompassing pain-management techniques: i.e. ignoring non-verbal pain behavior and learning to discriminate between sick and well behavior). *Waiting list control (n=8)*: no treatment
Sanders et al. [109]	Clinical sample of 44 children (7-14 yr) with recurrent abdominal pain	Randomized controlled trial	*CBT family intervention (n=22)*: incorporating education about RAP, contingency management training for parents, self-management training for children (progressive muscular relaxation, deep-breathing exercises, positive self-talk, distraction, imagery skills, problem solving techniques and relapse prevention. 6 sessions, 50 minutes including one session for parents. *Standard Paediatric Care (SPC) (n=22)*: incorporating reassurance, encouragement not to overreact, child participation in school. 4-6 sessions.
Walco and Ilowite[128]	Clinical sample of 5 children (8-17 yr) with juvenile primary fibromyalgia syndrome	Case-report	*CBT (n=5)*: 4-9 sessions encompassing progressive muscle relaxation, meditative breathing, cognitive distraction and guided imagery.

Study design

The purpose of our research was to gain insight in the influence of multiple psychosocial factors (vulnerability, reinforcement, modeling and coping mechanisms) on chronic pain and quality of life of adolescents. For this, we have developed a model in which these factors are taken into account (Figure 1).

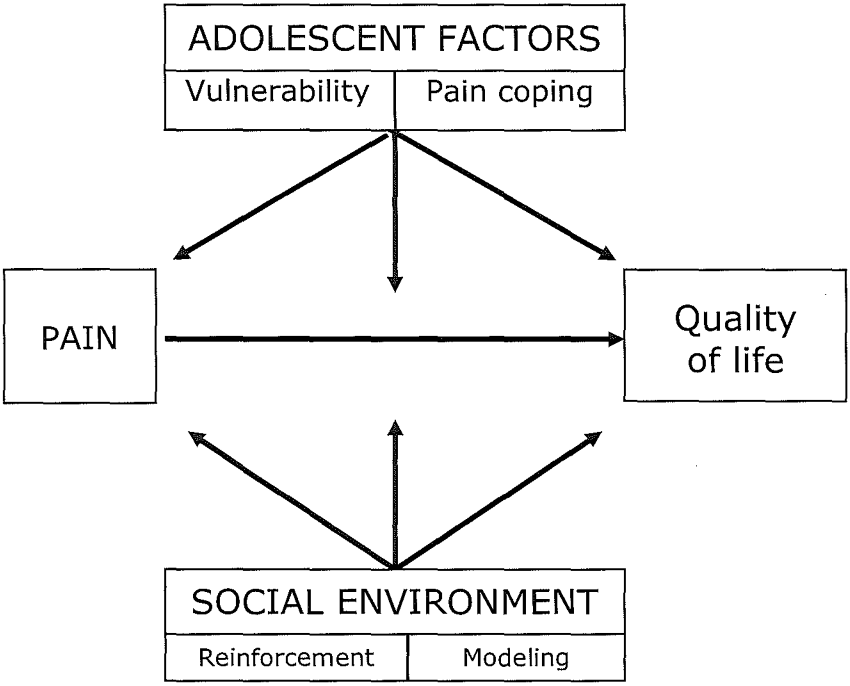

Figure 1: Model of pain and quality of life in adolescents with chronic pain

This model is based on Varni and Wallander[117] and Varni[118]. Their theoretically driven research has focused on a stress and coping model that has been derived to explain the observed variance in adaptation of children with chronic disorders. Risk and resilience factors can be identified which provide guidance for new (bio-) behavioral treatment interventions for children with chronic disorders. Varni[118] has extended this risk and resilience conceptual model to pediatric pain; the multidimensional Biobehavioral Model of Pediatric Pain. In this model, a number of variables are included that may directly or indirectly influence pediatric pain perception and associated functional status outcome parameters. The precipitants include disease, physical injury, and psychological stress.

Intervening factors are biological predispositions, family environment, cognitive appraisal, coping strategies. Outcome measures are functional status variables and pain. Functional status factors are hypothesized to be bi-directionally linked to pain perception, but also to be effected by precipitants and intervening variables. Major drawback of this model is the clustering of large numbers of variables. In our model adaptation to chronic pain is reflected by the quality of life of the adolescents. Focussing on quality of life enables a broad view on an adolescents' daily functioning (i.e. social functioning, psychological functioning, functional status). The extent of adaptation is reflected in the adolescents' view of their quality of life.

The model was evaluated in two designs: a cross-sectional and a randomized controlled design. In the first study quality of life and psychosocial factors were studied in adolescents with chronic pain and healthy controls. Additionally, the model was evaluated by investigating the contribution of variables from our model to chronic pain and quality of life. Based on these results a cognitive behavioral intervention program was developed for adolescents with chronic pain irrespective of their pain location. A randomized controlled trial design was used to explore its effect on quality of life and chronic pain. In this second design, the model was tested by studying changes in pain and quality of life due to changes in the psychosocial factors as a consequence of the intervention program.

Focus of the studies described in this thesis are adolescents (aged 12 through 18 years) with chronic pain with unknown organic etiology. Chronic pain was defined as having recurrent (i.e. pain with pain-free intervals) or continuous pain existing for three months or longer. Adolescents reporting their pain as part of a diagnosed chronic disease (rheumatic arthritis, malignancies) were excluded, because the rareness of these conditions would require a different design and the treatment of the disease and of the pain would be difficult to separate.

Research questions

The following research questions were addressed:

1. Is the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP) a reliable and valid instrument to measure the quality of life of adolescents with chronic pain irrespective of their pain location? (Chapter 2)
2. Is the Pain-related Problem List (PPL) a reliable and valid instrument to measure pain-specific problems in adolescents with chronic pain? (Chapter 3)

3. Are there differences in psychosocial factors (vulnerability, reinforcement, modeling and coping mechanisms) between (parents of) adolescents with chronic pain and (parents of) controls? (Chapter 4)
4. What is the contribution of psychosocial factors to chronic pain? (Chapter 4)
5. Which factors are related to the quality of life of adolescents with chronic pain? (Chapter 5)
6. Is the relationship between pain and quality of life moderated by psychosocial factors? (Chapter 5)
7. Is a cognitive-behavioral intervention that includes parents and peers, feasible for adolescents with chronic pain? (Chapter 6)
8. Is a cognitive-behavioral intervention more effective in enhancing quality of life and reducing pain than no intervention (a control group), in adolescents of the general population with chronic pain irrespective of pain localisation? (Chapter 7)
9. Does the intervention group differ from the control group in psychosocial factors and pain-related problems after the intervention procedure? (Chapter 7)

Outline of the thesis

The studies in this thesis are primarily concerned with testing the model of pain and quality of life in adolescents with chronic pain (figure 1).

Instruments for measuring the impact of chronic pain on the life of adolescents are sparse, especially those based on the personal experiences of these adolescents. *Chapter 2 and* 3 present the shortening of a quality of life questionnaire and the development of a pain-related problem list, respectively. *Chapter 2* presents the shortening of an existing generic quality of life questionnaire for adolescents with chronic headache. We were interested in obtaining a quality of life questionnaire suitable for adolescents with chronic pain irrespective of their pain location. Psychometric qualities of the shortened version, the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP), are presented. To obtain insight into the impact of chronic pain on daily functioning we developed the Pain-related Problem List (PPL) for adolescents. This questionnaire is based on the personal experiences of adolescents with chronic pain. Psychometric qualities of this questionnaire are presented in *Chapter 3.*

Chapter 4 and 5 present the results of the cross-sectional studies. *Chapter 4* describes the comparison between adolescents with chronic pain and healthy controls on the factors of the model. It further addresses the contributions of the psychosocial factors on chronic pain. *Chapter 5* describes the contribution of the psychosocial factors on quality of life and the moderating effect of these factors on the relationship between pain and quality of life.

Chapter 6 and 7 describe the development and results of a cognitive-behavioral training program for adolescents with chronic pain. The CBT program is developed based on results presented in chapter 4 and 5 and on existing cognitive-behavioral programs. *Chapter 6* presents a pilot study on our CBT program. In this study the program is evaluated on its feasibility in daily life of adolescents with chronic pain. Additionally, an impression of the effect of the program on quality of life and pain is presented. In *chapter 7* this program is evaluated in a randomized controlled study.

Chapter 8 concludes on the major findings of the study, reflects on the study methodology and gives recommendations with regard to clinical practice and further research.

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Chapter 2

Shortening and psychometric qualities of a quality of life questionnaire for adolescents with chronic pain (QLA-CP)

Psychological Reports 2002;90:753-759

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Abstract

Chronic pain is a common experience in adolescence. To measure its influence on quality of life in adolescents with chronic headache, Langeveld developed the 71-item scale, Quality of Life Headache-Youth (QLH-Y).[1] On the basis of this questionnaire we developed a shortened list, also suitable for other pain locations to enhance compliance. For this, we tested a sample of 98 adolescents from an open population with chronic pain. This article presents the psychometric qualities of the shortened version, named the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP). The original version could be reduced to 44 items which showed suitable internal consistency and construct validity against COOP/WONCA charts.

Introduction

Little is known about the influence of chronic pain on the quality of life in adolescents, mainly because, until recently, no suitable instruments were available. Recently, Langeveld and colleagues developed a generic quality of life questionnaire for adolescents with headache (Quality of Life Headache-Youth; QLH-Y) which they reported to be reliable, sensitive, and of adequate construct validity.[1,2] Hunfeld and colleagues adapted the original version for adolescents with different types of chronic pain (by replacing 'headache' with 'pain' in the items).[3] Their research showed that the greater the intensity and frequency of pain, the lower the quality of daily life, especially for Psychological Functioning, i.e., more depressive symptoms, tiredness, less harmony and vitality, Physical Functioning, i.e., more somatic symptoms, and Functional Status, i.e., more interference with daily activities.

To facilitate its use in routine clinical practice and to increase the compliance of the respondents, a shortening of the original version (71 items) is recommended.[1] The present article reports on the construction and psychometric qualities of a shortened version of the QLH-Youth, the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP). Tested were hypotheses that the shortened version is reliable in terms of internal consistency and the shortened version has adequate construct validity.

Methods

Sample

The sample of 98 adolescents (aged 12 through 18 years) with chronic pain participated in a previous study.[3] They reported pain existing for 3 months or more. Adolescents who indicated that a physician had diagnosed a chronic

disease, which could explain their pain, e.g., rheumatic arthritis or other malignancies, were excluded.
The 76 girls and 22 boys had a mean age of 14.6 yrs. Their most frequently reported pain complaints were limb pain (35%), headache (25%), back pain (16%) and abdominal pain (14%). The mean duration of the pain was 3 yrs (SD = 3, range 3 mo. to 15 yrs).

Procedure

The adolescents completed questionnaires on quality of life and health status. In addition, they all kept in a pain diary a record of their pain for three weeks.

Instruments

Questionnaires

Quality of life was measured with the Quality of Life for Headache in Youth (QLH-Y).[1] This questionnaire is subdivided into six domains, each measuring a particular aspect of quality of life: (1) Psychological Functioning, (2) Functional Status, (3) Physical Functioning, (4) Social Functioning, (5) Satisfaction with Life in General, and (6) Satisfaction with Health. The domain Psychological Functioning has of 33 items, distributed over seven subscales (a) stress, (b) harmony, (c) tiredness, (d) vitality, (e) depression, (f) cheerfulness, and (g) optimism about the future. The domain Functional Status (14 items) is distributed over two subscales, which record the limitations associated with the pain on daily activities and on leisure activities. The domain Physical Functioning has of one subscale, somatic functioning of 10 items. The three subscales of the domain Social Functioning have 12 items which comprise interactions (a) at home, (b) with brothers and sisters, and (c) with peers. Response categories, ranging from 0 to 3 were assigned to the items. The last two domains consist of two visual analogue scales on which to record Satisfaction with Life in General and Satisfaction with Health. A high score on each domain of the questionnaire represents a better quality of life.

Also, the Dartmouth Primary Care Cooperative Information Project (COOP) Functional Health Assessment Charts/WONCA were used in a Dutch translation to measure health status.[4] This generic questionnaire measures health status in the domains Physical Health, Feelings, Daily Activities, Social Activities, General Health, and Pain. Each domain consisted of one question to be rated on a 5-point scale with categories of very much, rather, moderate, a little bit, and not at all. A high score represents a lower quality of life.

Pain diary

The pain diary contained Visual Analogue Scales on which the adolescents recorded the intensity of their pain on a daily basis. The scale is a 100-mm horizontal line with 'no pain' and 'worst pain ever' at the respective ends. The percentage of recordings of pain represented the frequency of occurrence of pain. The adolescents were asked to mark a position on the Visual Analogue Scales that best matched the intensity of their pain at the moment they had to complete the scale, i.e., breakfast, dinnertime, and bedtime. To obtain valid pain data, adolescents with more than 25% missing pain data (VAS) in the diary were excluded from analyses on diary data. Prior estimates of the reliability and validity of this scale are high.[5]

Data reduction and analysis

Principal component analysis is a multivariate data-analysis technique, which is often used to analyse a large number of variables. The most important coherence within a set of variables can be described using this technique. Considering the many items for the domain of Psychological Functioning (33 items), we conducted a principal component analysis with varimax rotation only for this domain. Aiming to identify a simple structure in the observed data, we used varimax rotation. A selection was made based on the component loading, where initially a value of 0.50 was maintained as a cut-off point. After this principal component analysis, the domains of Psychological Functioning, Functional Status, Physical Functioning, and Social Functioning were taken through the same steps for further shortening. The domains that record Satisfaction with Life in General and Satisfaction with Health were excluded from analysis because they each have only one question (VAS).

The contribution of the items to the internal consistency of the specific subscale was considered for each domain. Items that did not contribute much to the value of alpha were potential candidates for elimination. Three psychologists evaluated items in terms of their relevance for the scale for which they were intended to contribute. Items were excluded when their content was judged by consensus as not measuring an aspect of the intended concept.

Next, Pearson correlation coefficients were calculated between the domain scores of the shortened version and those of the COOP/WONCA charts. We expected a high negative correlation between similar scales (convergent validity) and a low correlation between dissimilar scales (divergent validity).

Furthermore, subgroups were made to assess the sensitivity of the shortened version for the frequency and intensity of pain. Adolescents with a mean pain frequency of at least 50 % of the time during the three weeks of diary recording were considered as having high frequency of pain. Adolescents with a mean pain intensity (measured on a Visual Analogue Scale) of at least 50 mm were labeled as having high intensity of pain. A t-test was used to assess the differences between subgroups on quality of life.

Results

Subjects

Eighty-one diaries and 98 questionnaires completed by adolescents with chronic pain were included in the analysis. The 17 adolescents who were excluded from analyses based on diary data had more than 25% missing data in their diaries.

Item reduction

In accordance with the original seven subscales of the domain of Psychological Functioning, the principal component analysis was set to extract seven components. The principal component analysis showed that the first five components explained 58% of the total variance. The last two components explained a further 4 and 3%. Study of the items of each component enabled the maintenance of the original subscale names. The items of the subscales for harmony and vitality in the original version showed great coherence on one subscale in the principal component analysis; therefore, these two subscales were merged into one new subscale. Considering these, the domain of Psychological Functioning was reduced to five subscales: (1) Harmony and Vitality, (2) Stress, (3) Tiredness, (4) Cheerfulness, and (5) Depression.

Tabel 1: Number of items and internal consistency of domains from Quality of Life Headache-Youth and Quality of Life questionnaire for Adolescents with Chronic Pain.

Domain and Scales QLA-CP	**Number of items**		**Cronbach alpha**	
	Original version	Shortened version	Original version	Shortened version
Psychological Functioning	33	17	0.93	0.87
1. Harmony and vitality	8	4		
2. Stress	8	4		
3. Fatigue	4	3		
4. Depression	6	3		
5. Cheerful mood	5	3		
6. Optimism about the future	3	-		
Functional Status	14	11	0.86	0.81
7. Impact on daily activities	8	7		
8. Impact on leisure activities	6	4		
Physical Functioning	9	5	0.76	0.71
9. Somatic symptoms	9	5		
Social Functioning	12	9	0.66	0.66
10.Interactions at home	6	5		
11.Interactions with peers	3	2		
12.Interactions with brothers / sisters	3	2		
Satisfaction with Life	1	1		
Satisfaction with Health	1	1		

Reliability and validity

Table 1 shows the reliability of each domain of the shortened version after reduction. The internal consistency of these domains showed little reduction after dropping items.

Table 2 gives an overview of the Pearson correlations between the domains of the shortened version and the domains of the COOP/WONCA charts. There were moderate to high negative correlations between the domains of Psychological Functioning, Functional Status and Social Functioning of the shortened version and similar domains of the COOP/WONCA charts. These correlations were higher than those between dissimilar domains. Physical Functioning of the shortened version, however, had a high correlation with Feelings and a low correlation with Physical Fitness of the COOP/WONCA charts domains.

Tabel 2: Correlations for domains from Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP) and domains from the COOP/WONCA (n=98)

	COOP / WONCA CHARTS				
Quality of life for adolescents with chronic pain (QLA-CP) **M (SD)**	Physical Fitness M (SD)	Emotional Feelings M (SD)	School Work M (SD)	Social Support M (SD)	Family M (SD)
	3.73 (1.02)	3.42 (1.08)	3.73 (1.04)	4.27 (1.14)	2.77 (1.08)
Psychological Functioning 1.88 (.50)	- .05	**- .71****	- .24*	- .28**	- .03
Functional Status 2.27 (.52)	.03	- .26**	**- .31****	- .30**	- .06
Physical Functioning 2.03 (.55)	**- .15**	- .55**	- .08	- .21*	- .03
Social Functioning 1.71 (.51)	.05	- .29**	- .30**	**- .42****	- .34**

Note Associations between similar domains are in boldface. * $P < 0.05$ ** $P < 0.01$

T-tests showed that a high frequency of pain (≥ 50% pain during the three weeks of diary entries) corresponds to a significantly lower quality of life on all domains of the shortened version, with the exception of the domain of Social Functioning (Table 3). Adolescents with high pain intensity had significantly lower quality of life as represented by scores on the domains of Psychological Functioning and Functional Status of the shortened version. Adolescents with mean pain intensity higher than 50 mm had a significantly more favorable mean on the domain of Social Functioning of the shortened version.

Tabel 3: Relationship between pain parameters (intensity and frequency) and domains of the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP).

Quality of life for adolescents with chronic pain (QLA-CP)	*Frequency*		t_{81}	**P**	*Intensity*		t_{79}	**P**
	<50% *n=22*	*≥50%* *n=59*			*<50mm* *n=72*	*≥50mm* *n=9*		
Psychological Functioning	2.15	1.78	3.41	<.01	1.92	1.53	2.34	.01
Functional Status	2.44	2.18	2.00	.03	2.29	1.98	1.64	.05
Physical Functioning	2.19	1.96	1.81	.04	2.03	1.95	0.41	ns
Social Functioning	1.66	1.68	1.66	ns	1.65	1.95	-1.66	.05

Note Student *t*-test; one –tailed; n = 81. * Visual Analogue Scale

Discussion

The original 71 items of the Quality of Life Headache-Youth were reduced to 44 items in the new version of the Quality of Life questionnaire for Adolescents with chronic pain (QLA-CP). The internal consistency of the domains and subscales were sufficiently preserved.

The high negative correlation for Psychological Functioning, Functional Status, Social Functioning with the corresponding domains of the COOP/WONCA charts suggests that the shortened version has adequate construct validity. A possible explanation for the low correlation between Physical Functioning of the shortened version and Physical Fitness of the COOP/WONCA charts is that items of the domain Physical Functioning asked about psychosomatic symptoms. This explanation is supported by the relatively high significant correlation between Physical Functioning of the shortened version and Feelings on the COOP/WONCA charts.

This study showed a negative relationship for pain parameters (intensity and frequency of pain) with all domains of the shortened version with the exception of Social Functioning (for frequency) and Social Functioning and Physical Functioning (for intensity). The positive correlation between pain intensity and the domain of Social Functioning could be explained by the perception that this domain refers to social support instead of actual functioning in social situations. It is conceivable that adolescents with intense pain refer more to the social support of significant others. This explanation is supported by the moderate correlation between Social Functioning of the shortened version and social support of the COOP/WONCA charts. Notwithstanding this observation, it may be stated that the shortened version discriminates between adolescents with pain on both intensity and frequency.

To summarize, our analysis of the psychometric qualities indicates that the shortened version is internal consistent and valid. Test-retest reliability and research on the responsivity to change of this questionnaire is recommended. Combining this generic type of questionnaire with a method of measurement for pain-specific problems is advisable for use in multidisciplinary interventions. This enables the assessment of pain related quality of life of adolescents with different types of chronic pain.

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Chapter 3

Development and psychometric qualities of a pain-related problem list for adolescents with chronic pain (PPL)

Patient Education and Counseling, in press

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Abstract

Instruments for measuring pain-related problems in adolescents with chronic pain are sparse, especially those based on the personal experiences of these adolescents. This study aimed to develop and test such an instrument, the Pain-related Problem List for adolescents (PPL). A sample of 129 adolescents with chronic pain with unknown organic etiology completed the 57 item problem list, which was based on interviews with a similar group of adolescents with chronic pain. Principal components analysis yielded four domains: problems related to 1) concentration, 2) mobility, 3) adaptability and 4) mood. The questionnaire was shortened to 18 items and has good reliability (total $\alpha = 0.82$, concentration $\alpha = 0.86$, mobility $\alpha = 0.77$, adaptability $\alpha = 0.71$, mood $\alpha = 0.78$); the validity also proved to be adequate, especially in the general population sample. The PPL provides a tool to assess the impact of chronic pain in adolescents. Future research should focus on further validation of the PPL in a large clinical population and establishing its test-retest reliability.

Introduction

More than one third of Dutch adolescents aged of 12 tthrough 18 years suffers from chronic pain.[1] The IASP defines chronic pain as pain that persists beyond the normal time of healing.[2] In children and adolescents chronic pain has been defined as recurrent or continuous pain existing for three months or longer[3] and should be considered a stressful event for the child[4] and their family[5]. In adolescents the most frequently occurring chronic pains are headache (19%), limb pain (16%), abdominal pain (13%) and back pain (11%).[1] Due to the high prevalence of chronic pain much research has focused on its impact on adolescents, and it has been shown that adolescents with chronic pain report lower quality of life scores than those without pain.[6] A positive relation between chronic pain and depression[7,8] and anxiety[8] has been found. Of all chronic pain types in adolescence, headache is the most extensively studied and has been shown to have a negative impact on quality of life.[9-11] Adolescents with frequent headaches show higher levels of anxiety and depressive symptoms,[12-15] functional disability,[12] and somatic complaints.[15]

The impact of other types of chronic pain have also been studied. For example, in a sample of schoolchildren with low back pain, 94% reported some disability, mostly difficulty carrying school bags.[16] Recurrent abdominal pain is also reported to be associated with depression[17] and anxiety[18]. Although chronic pain in adolescents has great impact on various aspects of quality of life and functioning, few instruments are available to measure the pain-related problems irrespective of pain localisation. Moreover, to measure specific burden associated with chronic pain, specific items based on experiences of adolescents with pain are needed. Such a questionnaire enables treatment planning and evaluation in clinical practice. An useful theoretical starting-point and a well written procedure to develop our questionnaire is postulated by McKenna and colleagues we followed their procedure.[19-20] They assume the needs-based model of Maslow in which quality of life is considered the extent to which an individual is able to meet his or her needs. The development of disease-specific instruments should therefore focus on the extent to which needs are hampered by the disease in question. The problems that patients encounter when they try to fulfil their needs can determine the reduction in their quality of life. These problems differ between patients with different diseases. For instance, walking is impeded in patients with claudicatio intermittens, while thinking is obstructed in those with a migraine attack. This theoretical basis for the development of a disease-specific instrument requires the involvement of patients in the item construction both as experienced experts and as providers of the right wording of the items. The purpose of this study was to develop an interview-based list of pain-related problems that hamper the needs of adolescents with chronic pain, the Pain-related Problem List for adolescents (PPL), and to assess its psychometric properties.

Methods

Subjects

Three different samples were used for this study. The first sample was used for item construction and is described under the following heading *'Item construction'*. The second sample was used for item reduction and validation; the third sample was used for validation in a clinical sample. Both samples are described in the following heading *'Item reduction and validation'*.

Item construction

Twenty-four adolescents (aged 12-18 years) with chronic pain at different locations who had participated in a prevalence study[1] were followed-up three years later (aged 15-21 years) and interviewed about the impact of the pain on their everyday life.[21] The interviews were performed by a psychologist at the subject's home: The questions addressed pain and pain-related consequences for everyday life, particularly in terms of how the pain hampered them in their emotional, social and physical functioning, home and school activities, hobbies, going out and self-esteem. The semi-structured interviews were audiotaped and later transcribed. The content of the interviews was analysed by comparing all the consequences reported by the subjects and clustering them if they referred to the same topic. In order to maintain the patient-based nature of the instrument the items were formulated in the actual words of the respondents, as much as possible. The interviews were examined for phrases that might be suitable for inclusion in a pain-related problems measure. According to the needs model, phrases were selected if they described the negative impact of pain on the adolescents' ability to meet their needs. This was done by a team consisting of three psychologists, a pedagogue and a paediatrician, leading to the formulation of 57 items. Before distributing the pain-related problem items at school, the items were presented to five adolescent girls without chronic pain to test their face validity.

Item reduction and validation

After determining the face validity, 447 students of a secondary school in Rotterdam were asked to complete the pain-related problem items and additional questionnaires (see method section) during class. The questionnaires were administered to students of all years and educational levels.

Students were included in the study sample if they were 12 through 18 years, and experienced recurrent or continuous pain (with unknown organic etiology), existing for three months or longer. As the PPL addresses the impact of the pain in the previous week (the week before administration), students were excluded if they had no pain in the previous week, or if the pain of the previous week was different from the location of their chronic pain.

The goal of the study (i.e. to develop a pain-related problem list for adolescents with chronic pain) was explained and instructions on how to fill out the questionnaires were given by one of the researchers. The researcher and the teacher remained in the class during completion.

The questionnaires were also administered in a clinical sample of 31 adolescents (12-18 years) with chronic pain with unknown organic etiology. These adolescent had reported chronic pain and were enrolled for a psychosocial intervention on learning how to cope with chronic pain in daily life in the Erasmus University Medical Center.

Measures

Pain-related problems

The 57 items of the Pain-related Problems List (PPL) have response categories ranging from 0 (seldom or never) to 3 (very often or always).

Demographic data

Demographic data included the adolescent's date of birth, gender, nationality, educational level and current school year.

Pain

The Pain Questionnaire collected information about the location, frequency, duration of the pain episodes and history (i.e. number of months in pain), using a retrospective format.[1] From a list of possible locations (head, abdomen, back, limb, neck, ear, throat, chest and elsewhere) subjects were asked to indicate all locations where they had experienced recurrent or continuous chronic pain in the previous three months. Pain intensity was measured with the Chronic Pain Disability Index (CPDI). Adolescents were asked whether they had consulted a physician or general practitioner about their pain and, if so, the physician's diagnosis.

Additional questionnaires

CPDI

The Chronic Pain Disability Inventory (CPDI) is a seven-item questionnaire which measures the intensity of the pain and disability caused by pain in the previous month. It consists of three pain intensity items and four disability items. Response categories range from 0 ("no pain" or "no disability") to 10 ("worst imaginable pain" or "complete disability"). The CPDI was adapted from the chronic pain grading scale, which is a reliable and valid instrument.[23]

PedMIDAS

The Pediatric Migraine Disability Assessment (PedMIDAS) is a six-item questionnaire that assesses disability caused by headache in children and adolescents.[22] For each item subjects report the number of days a particular aspect of functioning was impaired in the previous three months due to headache. It has shown to be a sensitive, reliable and valid instrument for this group of patients.[22] The items in PedMIDAS have been translated by us into Dutch and adapted to suit all pain locations by replacing the word "headache" with the word "pain".

QLA-CP

Two domains of the QLA-CP (Quality of life questionnaire for Adolescents with Chronic Pain; see chapter 2), measuring satisfaction with life in general and satisfaction with health, were included for validation in the general population sample. Both domains were measured with a visual analogue scale (VAS). This is a 100-mm line with the anchors "completely dissatisfied" (0 mm) to "completely satisfied" (100 mm). In addition, the four remaining domains (Psychological Functioning, Functional Status, Physical Functioning and Social Functioning) were administered for validation in the clinical sample. The QLA-CP is adapted from the QLH-Y[24] and has shown suitable consistency and construct validity against COOP/WONCA charts (see chapter 2). A higher score on each domain of the QLA-CP represents a better quality of life.

Data analysis

Item reduction

To identify the internal structure of the PPL and to reduce the number of items a principal components analysis was conducted with varimax rotation. It was first set to extract the components with an eigenvalue ≥ 1. Subsequently a scree plot of the eigenvalues was used to determine the number of components to retain. Items with a component loading < 0.40 were excluded.[25] Additional reduction was done by excluding items that contributed little to the internal consistency of that specific component.

Domain scores were computed by averaging the items loading on each component. A total score was obtained by summing the domain scores.

Internal consistency and validity

Internal consistency of the PPL total score and its domains were evaluated by Cronbach's coefficient alpha. Pearson's correlations were carried out between the PPL with the disability items of the PedMIDAS items and the CPDI, the two visual analogue scales of the QLA-CP, and the pain parameters (i.e. frequency and intensity of the pain). To assess the validity of the PPL in a clinical practice, Pearson's correlations were also calculated with the CPDI, all domains of the QLA-CP and with pain parameters. Correlations between similar domains and items were expected to be strong, thereby indicating convergent validity. Correlations between dissimilar domains and items were expected to be weaker and reflect divergent validity. Convergent validity was also tested in both the general and clinical sample by comparing the pain-related problem scores of adolescents with high pain intensity with those with low intensity using independent t-tests. A score higher than the median (for both samples 5 on a scale of 0 'no pain', to 10 'worst imaginable pain') was considered to be high intensity, low intensity was a score of 5 or lower.

Results

Subjects

Item construction

The mean age of the 24 subjects was 17.5 years, four of whom were male (16.7%) and 20 (83.3%) female. Pain was reported in the head (54.2%), limbs and/or back (41.2%) as well as multiple pains (abdomen and limbs, 4.2%). The five adolescent girls without chronic pain participating in the face validity study had a mean age of 15 years (range 13 to 18 years).

Item reduction and validation

Table 1 gives the characteristics of the 129 subjects (aged 12-18 years) included in the present study. The majority were girls (71%) and the most frequently reported pain was headache (30%). The mean pain intensity was 5.2 (SD=2.2), on a scale ranging from 0 to 10. On average pain was experienced once a week. The pain existed on average for 29.2 months (SD = 29.9).

Table 1: Characteristics of the study population

	General Population (n=129)*	Clinical sample (n=31)
Demographic factors		
Age: Mean in years (SD)	15.1 (1.6)	15.2 (2.0)
Gender: Number (%)		
Boys	37 (28.7)	2 (0.7)
Girls	90 (69.8)	29 (93.0)
Pain Characteristics		
Intensity: Mean (SD)**	5.2 (2.2)	5.3 (2.2)
Location: Number (%) ***		
Head	39 (30.2)	14 (45.0)
Limb	25 (19.4)	5 (16.0)
Back	17 (13.2)	4 (13.0)
Abdomen	15 (11.6)	4 (13.0)
Neck	6 (4.7)	-
Multiple	20 (15.5)	-
Other	7 (5.4)	4 (13.0)

* Gender of 2 subjects missing ** Range of scores 0-10 *** Pain location that troubled the adolescents the most

Table 1 also shows the characteristics of the 31 subjects (aged 12-18 years) in the clinical sample. The majority were girls (93%) and the most frequently reported pain was headache (45%). The mean pain intensity was 5.3 (SD=2.2), on a scale ranging from 0 to 10. The majority (66%) of the adolescents experienced their pain every day. The pain existed on average for 47.5 months (SD = 23.3).

Item construction

After content analysis of the interviews, 57 items concerning the impact of pain were formulated. Response categories for the 57 items ranged from 0 (seldom or never) to 3 (very often or always). Considering the face validity of the 57 items none of the five adolescents had difficulty understanding the items; all attributed the same meaning to the items as intended by the researchers. The only change made to the items was the addition of an example ('a schoolbag') in the item "the pain troubled me when lifting".

Item reduction

Principal components analysis extracted 16 components with an eigenvalue > 1. Examination of the scree plot suggested four components, this was also the solution that was best interpretable. The four components together explained 42% of the variance; these components were designated: problems related to concentration, mobility, adaptability, and mood.

Of the 57 items, five had a loading < 0.40 on each of the four components, and these were excluded from further analyses. Supplementary item reduction was done by examining the internal consistency of each of the components. Items contributing little or nothing to the internal consistency were also excluded. Our purpose was to retain a Cronbach's alpha of at least 0.70 for each component.[25] In this way, the original 57 items were reduced to 18 items.

Internal consistency

The internal consistency (Cronbach's alpha) was adequate for the domains and the total score. Table 2 lists the 18 remaining items the factor loadings, Cronbach's alpha and Pain-related problem scores. These items together formed the PPL (Pain-related Problem List for adolescents).

Validity

Table 3 gives the Pearson product-moment correlations of the PPL total score and domain scores, with pain intensity (CPDI items) and pain frequency. Problems in *concentration, mobility* and *adaptability,* and the total scale show significant positive correlations with pain intensity, indicating that adolescents with higher pain intensity experienced more problems in these areas. This was also shown by t-tests comparing adolescents with low pain intensity (score ≤ 5) to adolescents with high pain intensity (score > 5). Adolescents with high pain intensity scored higher on problems in *concentration* (t (112.9) = - 2.33, $P < 0.05$), *mobility* (t (126) = - 6.19, $P < 0.001$), *adaptability* (t (125) = - 2.89, $P < 0.01$) and the total scale (t (123) = - 4.42, $P < 0.001$) than adolescents with low pain intensity. *Mood* was the only domain that was less affected by the pain intensity, i.e. adolescents with high and low pain intensity did not report much differences in problems related to their mood. *Mobility* is the only domain significantly correlated with pain frequency, indicating that adolescents with more frequent pain are less mobile.

Table 2: Component loadings and internal consistency of the Pain-related Problem List

Components and items	Component Loadings	Cronbach's alpha	Scores* M	(SD)
Concentration		0.86	2.14	(0.82)
1. drowsiness	0.71			
2. trouble with reading	0.71			
3. prolonged mental activity	0.75			
4. trouble with thinking	0.71			
Mobility		0.77	2.40	(0.58)
5. unable to play sports	0.69			
6. trouble with walking	0.46			
7. trouble with pushing	0.56			
8. trouble with lifting	0.62			
9. trouble with carrying	0.60			
Adaptability		0.71	2.72	(0.44)
10. must wear flat shoes	0.68			
11. use of aids at school/work	0.70			
12. frequent toilet use	0.49			
13. can not eat much	0.63			
14. must sit bent over	0.59			
Mood		0.78	1.83	(0.69)
15. feel peevish	0.54			
16. have bad moods	0.56			
17. react angrily to others	0.59			
18. feel like doing nothing	0.59			
Total		0.82	9.10	(1.69)

Higher score indicates more pain-related problems

Note: Range of subscale scores: 0-3, range of total: 0-12

Table 3 also shows the correlations between the PPL and satisfaction with life and health, CPDI disability items and PedMIDAS items. The problems related to *concentration* showed significant positive correlations with items concerning schoolwork, daily functioning and negative correlations with quality of life. Problems related to the *mobility* correlated with all disability items of the CPDI and items on diminished functioning of the PedMIDAS. Problems related to *adaptability* showed significant positive correlations with items on normal and fun activities of the CPDI and satisfaction with health, and *mood* had a moderate correlation with the two satisfaction scales, items related to school and normal activity of the CPDI. Finally, the total scale showed significant correlations with most items.

Table 3: Pearson correlations between the Pain-related Problem List PPL total and domains, and between pain parameters, satisfaction with life and health, CPDI and PedMIDAS

	Concentr	**Mobility**	**Adaptability**	**Mood**	**Total**
Pain :					
Pain frequency	0.03	0.28**	- 0.03	- 0.03	0.08
Pain items CPDI:					
1. How worse is the pain now?	0.37**	0.43**	0.24**	0.21*	0.48**
2. How worse is the pain usually in the past month?	0.31**	0.47**	0.33**	0.14	0.48**
3. How was the most awful pain in the past month?	0.27**	0.37**	0.24**	0.15^{T}	0.41**
Disability items CPDI:					
4. Days unable to do (school) work	0.37**	0.20*	0.19*	0.29**	0.41**
5. (school)work burdened by pain	0.43**	0.33**	0.17	0.21*	0.45**
6. Normal activities burdened by pain	0.32**	0.53**	0.37**	0.20*	0.54**
7. Fun activities burdened by pain	0.29**	0.47**	0.23*	0.13	0.43**
PedMIDAS items:					
1. Schooldays missed	0.30**	0.06	0.04	0.16	0.24**
2. Partial schooldays missed	0.15	0.23**	0.06	0.08	0.21**
3. Days functioned less than normal	0.34**	0.19*	0.16	0.10	0.31**
4. Days not able to do anything at home	0.25**	.02	.06	.14	.20*
5. Days not able to join activities after school	.06	.07	.01	-0.02	.05

PedMIDAS items: continued	**Concentr**	**Mobility**	**Adaptability**	**Mood**	**Total**
6. Days joined in activities while functioning less than normal	0.16	0.30**	0.15	0.01	0.23*
Quality of life:					
Satisfaction with life	- 0.35**	- 0.06	- 0.17	- 0.46**	- 0.42**
Satisfaction with health	- 0.49**	- 0.09	- 0.19*	- 0.41**	- 0.48**

*p<0.05 **p<0.01

Table 4 gives the Pearson product-moment correlations of the PPL total score and domain scores, with the pain intensity items of the CPDI and pain frequency. Problems related to *mobility and adaptability* show significant positive correlations with pain intensity, indicating that adolescents with higher pain intensity experienced more problems in these areas. This was also shown by t-tests comparing adolescents with low pain intensity (score ≤ 5) to adolescents with high pain intensity (score > 5). Adolescents with high pain intensity scored higher on the total scale (t (28) = - 2.15, P < 0.05) and on problems related to the *adaptability* (t (28) = -2.36, P < 0.05) than adolescents with low pain intensity. Although not significant, adolescents with a higher pain intensity tended to report more problems related to *mobility* (t (29) = - 1.77, P = 0.08). As for the general population, problems related to *mobility* are significantly correlated to pain frequency. Table 4 also shows the correlations between the PPL with the disability items of the CPDI and the QLA-CP. The problems related to *concentration* showed moderate correlations with items concerning schoolwork and normal activities while, as expected, *mobility* correlated strongly with all CPDI items. Problems in *adaptability* only correlated with items on fun activities, but *mood*-related problems did not correlate with any of the CPDI-items. The total score correlated highly with items related to schoolwork and normal activities. The problems related to *concentration* showed negative correlations with psychological functioning, satisfaction with life and functional status of the adolescents, indicating that more pain-related problems is related to a lower quality of life. The problems related to *mobility* correlated high but negatively with satisfaction with health and the functional status, while *mood*-related problems only correlated negatively with the psychological functioning of the adolescents. Finally, the total scale showed strong and negative correlations with the functional status of the adolescent (in terms of impact of the pain on daily activities and leisure activities).

Table 4: Pearson correlations between the Pain-related Problem List (PPL), including the total score and the domains with the pain parameters, the domains of quality of life (QLA-CP) and disability (CPDI) in the clinical sample.

	Concentr	**Mobility**	**Adaptability**	**Mood**	**Total**
Pain:					
Pain frequency	0.05	0.47**	0.19	0.03	0.29
Pain items CPDI:					
1. How worse is the pain now?	0.09	0.22	0.05	0.05	0.16
2. How worse is the pain usually in the past month?	0.02	0.36*	0.42*	0.17	0.32^{T}
3. How was the most awful pain in the past month?	0.22	0.48**	0.21	0.28	0.45*
Disability items CPDI:					
4. Days unable to do (school) work	0.02	0.36*	0.25	- 0.14	0.09
5. (school)work burdened by pain	0.41*	0.57**	0.30	0.29	0.59**
6. Normal activities burdened by pain	0.35*	0.45*	0.25	0.22	0.49**
7. Fun activities burdened by pain	0.04	0.46**	0.46**	0.01	0.29
Quality of life:					
Satisfaction with life	- 0.38*	0.15	0.04	- 0.18	- 0.15
Satisfaction with health	- 0.15	- 0.52**	- 0.08	0.08	- 0.27
Psychological Functioning	- 0.42*	- 0.05	- 0.03	-0.41*	- 0.35*
Functional Status	- 0.49**	- 0.50**	- 0.12	- 0.13	- 0.50**
Physical Functioning	- 0.27	0.15	- 0.24	- 0.23	- 0.21
Social Functioning	- 0.22	0.05	- 0.01	- 0.17	- 0.09

*p<0.05 **p<0.01

Discussion and conclusions

This study was designed to develop a pain-related problem list for adolescents with chronic pain. A principal component analysis was performed and the internal consistency and validity of the instrument were established. This resulted in the Pain-related Problem List, an 18-item questionnaire consisting of four domains: problems with (1) concentration, (2) mobility, (3) adaptability, and (4) mood. The internal consistency of the domains and the total scale proved to be adequate. We also found evidence for the validity of the total scale and the four domains in both the general population and the clinical sample.

In the general population sample, the total scale and all the domains except problems related to mood correlated negatively and significantly with pain intensity. In the clinical sample, problems related to concentration and mood did not correlate with pain intensity. T-tests also showed significantly less problems in adolescents with low pain intensity than in adolescents with high pain intensity on the total scale and all the domains except mood in the general population and for problems related to concentration and mood in the clinical sample. These results might indicate that negative mood in adolescents is related more to other factors (e.g. developmental tasks[26] or vulnerability[27]) than to pain. However, this suggestion needs further research. Problems related to mobility correlated positively and significantly with pain frequency in both samples. More frequent pain was associated with more problems in terms of mobility.

The total scale and the four domains all showed convergent and divergent validity in both samples. They had the strongest correlations with the items we expected, and the other items had either a weaker correlations or were not significant. This confirms our hypothesis that the PPL has adequate validity. Compared to the general population, we found less but stronger correlations in the clinical sample, which might be caused by the small number of patients. Therefore, a further validation of the PPL in a larger clinical sample is needed.

Limitations and strengths

In this study, we used the CPDI and the PedMIDAS to examine the PPL in a general population. One limitation of our study could be that the PedMIDAS may not have been an ideal instrument with which to validate our scale. The PedMIDAS originally was validated in patients from a tertiary referral center for pediatric headaches, whereas the PPL is based on experiences of adolescents in the general population and with chronic pain in various locations. However, at present, few validated instruments are available which are short and easy to administer, and measure pain-related problems of adolescents with chronic pain.

One of the main methodological strengths of the present study is the use of interviews to construct the items. The items thus reflect the experiences of adolescents with chronic pain and their related problems, rather than the opinion of professionals or exclusively theoretical constructs. Another strength is the applicability of our questionnaire to pain in different locations as opposed to only one pain location, which is the case with most other pain-related questionnaires on daily functioning.[24,28,29]

Practice and research implications

This relatively short questionnaire (18 items) can easily be implemented in routine clinical practice and enables the assessment of the impact of chronic pain on adolescents with different types of chronic pain. Because the items are based on the experiences of adolescents with chronic pain, the PPL reflects pain-related problems that hamper the needs of these adolescents. When complemented with a generic quality of life instrument, like the QLA-CP or Child Health Questionnaire (CHQ), health care providers are able to obtain a complete picture on the impact of chronic pain on adolescents in terms of problems and quality of life. In addition, the PPL can be used to evaluate the provided (health) care in the course of time. Consequently, the PPL is a useful supplement for assessment in clinical practice and research.

Further study is necessary on the test-retest reliability and responsiveness of this instrument. Because the PPL is a Dutch-language questionnaire, cross-cultural validation should be established and the PPL should also be further validated for a larger clinical population.

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Chapter 4

Psychosocial factors associated with chronic pain in adolescents

Pain 2003;101:33-43

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Abstract

A number of psychosocial factors have been associated with the onset, exacerbation and/or maintenance of chronic pain in adolescents. The present study was conducted to evaluate the relative importance of vulnerability, reinforcement, modeling and coping. We compared 222 adolescents with chronic pain with unknown organic etiology (headache, back, limb and abdominal pain) with 148 controls and their (respectively 183 versus 127) parents. Analyses showed that adolescents with chronic pain are more vulnerable in terms of neuroticism, negative fear of failure and (less) experienced social acceptance. Contrary to our expectations, the chronic pain group experienced less reinforcement for their pain behavior by both parents and peers than the control group. While the number of pain models was higher in the chronic pain group, no differences were found between their parents and those of the adolescents without chronic pain in pain experience, pain parameters, and pain coping. Regression analyses on the contribution of psychosocial factors to chronic pain and its parameters sustained the positive relation between vulnerability, (less) pain reinforcement, pain models and coping with pain. Furthermore, we also found evidence that gender differences have to be taken into account.

Introduction

An earlier prevalence study from our group showed that one-third of the adolescents have chronic pain.[1] The experience of pain not caused by a somatic disease in adolescence seems to have a negative influence on the quality of life.[2] The maintenance of chronic pain in the absence of a defined organic diagnosis has led researchers to seek psychosocial explanations. The most frequently studied factor related to chronic pain experience is psychological vulnerability. Comparisons with controls show that recurrent headache is related to stress and psychological symptoms in schoolchildren,[3] and to psychological distress[4] and anxiety[5] in adolescents. A high level of anxiety was also found for children with abdominal pain.[6,7]

Negative fear of failure (the fear to fail which results in not optimal functioning of the child) can be considered as a specific aspect of psychological vulnerability. Levine and colleagues found that perceived failure leads to negative affectivity and an increase in pain report.[8] Similarly, Passchier and Orlebeke found that negative fear of failure and headache complaints in children and adolescents are positively correlated.[9]

Another factor often associated with chronic pain is the parental response to pain. Behavioral observations of rheumatic adolescents and their mothers show that stimulation to refrain from painful physical activity leads to maladaptation (as revealed by school absence).[10] However, findings are inconsistent because

Osterhaus and colleagues showed that parental rewarding of pain behavior in migraine children was negatively related to pain.[11]

Apart from parental response, the presence of a parent with pain is another psychosocial factor that may influence the child's pain experience; the parent who suffers from chronic pain seems to act as a model. Edwards and colleagues showed a higher risk for abdominal pain in children who have a mother suffering from this type of pain;[12] the same results were found in families with headaches.[13] Besides the presence of pain models, Osborne found that the number of pain models was higher in families with unexplained pain than in families with medically explained pain complaints; moreover, similar pain locations were found in both the child and the pain model.[14] Additionally, maternal modeling of pain behavior (in contrast to paternal modeling) had a particularly strong influence on children's pain.[15] These results suggest that the way mothers cope with pain does influence the way children cope with their pain.

Although considerable research has focused on the association between psychosocial factors and pain, few studies have taken them side-by-side to report on their contribution to pain. Therefore, this study aims to determine the contribution of multiple psychosocial factors (vulnerability, reinforcement, modeling and coping) to the pain experience. In view of reported differences between boys and girls with chronic pain,[1,2] this study analyses both groups separately.

Further, to assess any differences in impact of psychosocial factors between adolescents with chronic pain and controls, these two groups are compared. The following hypotheses were tested for adolescents with chronic pain: a) they are more vulnerable in terms of neuroticism, fear of failure, and perceived social acceptance, b) experience more rewarding for their pain behavior, c) have more significant others with pain than adolescents without chronic pain, and d) are more likely to use emotion-focused avoiding strategies to cope with pain.

Finally, in parents of adolescents with chronic pain, the following hypotheses were tested: e) they report more pain complaints with higher scores on pain parameters (frequency, duration and intensity), f) use more passive coping strategies when in pain, and g) are more vulnerable, in terms of neuroticism, than parents of adolescents without chronic pain.

Methods

Subjects

A total of 1607 adolescents aged 12 through 18 years (48% boys and 52% girls) from the general Dutch population completed a short structured pain questionnaire in the classroom. Of these, 631 adolescents (39%) reported having chronic pain. Of the 1607 adolescents, 330 with and 301 without chronic pain were willing to participate in the present study on determinants of adaptation to chronic pain in adolescents. From these, all (n = 330) adolescents with and 218 adolescents without chronic pain (total 548) and one of their parents received a pain booklet at home. The 218 adolescents without chronic pain matched with the chronic pain group for gender, age (12-14 or 15-18 years) and educational level. The adolescents with chronic pain who were willing to participate (n=330) reported more intense (t(580)=4.47, P<0.001) and more frequent pain (Kruskal-Wallis, χ^2=35057.00, P=0.005), compared to the total sample of adolescents with chronic pain (n=631).

Chronic pain with unknown organic etiology was defined as having continuous or recurrent pain existing for three months or longer, in the absence of actual tissue damage or somatic disease (further referred to as chronic pain). Adolescents reporting their pain as part of a specific chronic disease (rheumatic arthritis, malignancies) were excluded.

Procedure

A booklet of questionnaires was mailed to the adolescents and to their parents (only one parent to respond). The adolescents were asked to complete the questionnaires concerning the psychological factors and their pain. Adolescents with more than one chronic pain location were asked to refer only to the pain that generally troubled them most. Adolescents without chronic pain were asked to refer to a situation in which they had experienced pain.

To increase the response rate, the adolescents were telephoned 1 week later to remind them to return the booklet; when this was done, they received a compact disc voucher (value 5 euros).

The responding parents were asked to complete questionnaires concerning their own pain and psychological functioning.

Questionnaires for the adolescent

Pain

The pain questionnaire collected demographic data on the adolescent's date of birth, gender, nationality, educational level and school year (Perquin et al., 2000). When the adolescents reported chronic pain, additional information about the pain was requested concerning location, frequency, duration, and intensity. From a list of possible locations (head, abdomen, back, limb, neck, ear, throat, chest and elsewhere), subjects were asked to indicate all locations where they had experienced chronic pain that existed at least three months. In case of more than one location, respondents were asked to answer questions about the pain they suffered the most. Pre-coded categories were used to assess the frequency of occurrence and the duration of a pain episode. The intensity of pain was assessed with a visual analogue scale (VAS; a 100-mm long horizontal line with the anchors 'no pain' and 'the worst pain you can imagine') by asking "How bad is the pain usually?" (Perquin et al., 2000).

Vulnerability

Three different aspects of vulnerability (neuroticism, fear of failure, and perceived social acceptance by others) were measured.

Neuroticism Neuroticism was measured with the inadequacy scale of the Dutch Personality Questionnaire - Junior.[16] The items on this scale address vague physical complaints, depressed mood, vague fears and feelings of insufficiency. Adolescents with a high score are more likely to describe themselves as tense and fearful and report more feelings of insufficiency; they are frequently characterized as having difficulty in making contact with others. The internal consistency of the inadequacy scale is $\alpha = 0.87$.[16] As to validity, the scale has a high correlation with other questionnaires which measure the same construct.[17] In the present study, the internal consistency for this scale was also high ($\alpha = 0.88$).

Fear of failure To assess fear of failure the scales 'negative fear of failure' and 'positive fear of failure' of the Achievement Motivation Test for Children were used.[18] Negative fear of failure refers to the fear of failing which results in not-optimal functioning of the child in a relatively unstructured stressful-achievement situation. Positive fear of failure is a fear which brings the child in an optimally tense condition that leads to a better achievement than under normal conditions. The higher the score, the more experienced fear of failure in an achievement situation. For the scales 'negative fear of failure' and 'positive fear of failure', the scale homogeneity was 0.79 and 0.82, respectively. The scales have been validated and have proven to be reliable in test-retest situations; 0.66 and 0.61, respectively. As to validity, the scales have shown suitable content and construct validity.[17] In the present study, the internal consistency for both scales was also high ($\alpha = 0.77$ and 0.81, respectively).

Social acceptance The scale 'social acceptance by others' of the School Questionnaire was used to measure the extent to which adolescents feel they are socially accepted by others.[19] A high score indicates a higher level of experienced social acceptance. The internal consistency and the test-retest reliability of this scale are good ($\alpha = 0.82$ and $r = 0.62$ at 4 months).[19] The scale 'social acceptance by others' is part of a higher-order scale Social-emotional attitude at school life. As to validity, this higher-order scale has a high correlation with questionnaires which measure the same construct.[17] In the present study, the internal consistency for this scale was also high ($\alpha = 0.82$).

Reinforcement

We literally translated the Illness Behavior Encouragement Scale (IBES) to measure the extent to which pain behavior is rewarded.[20] There is a version for children and one for parents. The children's version measures the rewarding of pain behavior by the parents, as perceived by the child. Because of the importance of peers for adolescents, we generated another version for use with peers. This latter version measures the child's perception of the rewarding behavior from peers when he/she is and is not in pain.

The rewarding behaviors covered by the items are e.g. giving small gifts, spending more time than usual with the child, giving special privileges, pampering or spoiling, excusing the child from regular chores. A higher score indicates more rewarding of (pain) behavior. The internal consistency for the child-report versions proved to be high (Cronbach's alpha = 0.81 for maternal rewarding and 0.83 for paternal rewarding). Test-retest reliability scores for the child-report versions yielded Pearson correlation coefficients of $r = 0.77$ and $r = 0.72$ for maternal and paternal rewarding of illness behavior, respectively.[20] The internal consistency for the four IBES-scales (rewarding by parents in a pain situation, rewarding by peers in a pain situation, rewarding by parents in a pain-free situation and rewarding by peers in a pain-free situation) in the present study was acceptable ($\alpha = 0.79$, $\alpha = 0.57$, $\alpha = 0.61$, $\alpha = 0.67$ respectively).

Modeling

To assess modeling, adolescents were asked whether they had significant others with pain complaints. When the answer was positive, additional information was requested about their relationship and the location of the pain complaints.

Coping

Pain coping strategies of adolescents were assessed with the Pain Coping Questionnaire.[21] This questionnaire assesses three higher-order strategies: approach (with the subscales: information seeking, problem solving, and seeking social support); problem-focused avoidance (subscales: positive self-statements, behavioral distraction, and cognitive distraction); and emotion-focused avoidance (subscales: externalizing, and internalizing / catastrophizing). Approach-coping strategies are directed toward the stressor while avoidance strategies are directed away from the stressor.

The adolescents were asked to indicate how often they used each of the 39 coping behaviors. A higher score indicates the likelihood to use the coping strategies in pain situations. The subscales and higher-order scales are internally consistent.[21] In the present study, the internal consistency for the higher- order scales was also high (Cronbach's alpha = 0.89 for approach; $\alpha = 0.89$ for problem focused avoidance; $\alpha = 0.78$ for emotion-focused avoidance).

Quality of life

To measure the quality of life of adolescents with and without chronic pain, we used the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP; see chapter 2). This is a shortened version of the Quality of Life Headache – Youth (QLH-Y) from Langeveld and colleagues.[22] The QLH-Y is reported to be reliable and valid.[22] The scale has been found to differentiate between adolescents with headache and healthy adolescents, with the former showing significantly poorer quality of life.[23] Hunfeld and colleagues showed that adolescents with all types of chronic pain reported poorer quality of life when the pain was more intense.[2] The QLA-CP is subdivided into six domains, each measuring a particular aspect of quality of life: (1) Psychological Functioning, (2) Functional Status, (3) Physical Functioning, (4) Social Functioning, (5) Satisfaction with Life in General, and (6) Satisfaction with Health. The last two domains were measured with a VAS (a 100-mm long horizontal line with the anchors 'completely dissatisfied' and 'completely satisfied'). A higher score on each domain of the QLA-CP represents a better quality of life. The QLA-CP has shown suitable internal consistency and construct validity against COOP/WONCA charts (see chapter 2). In the present study the QLA-CP showed an overall internal consistency of $\alpha = 0.74$.

Questionnaires for the parent

Vulnerability

Neuroticism To measure neuroticism of the parent, the inadequacy scale of the Dutch Personality Questionnaire was used.[24] Participants with a high score are often characterized as emotional, tense, despondent, insecure, and gloomy. They are quickly taken aback, discouraged, unbalanced and unstable. The internal consistency and the test-retest reliability of this scale are good ($\alpha = 0.86$ and $r = 0.77$).[24] The Dutch Personality Questionnaire has shown suitable content and construct validity.[17] In the present study the internal consistency for this scale was also high (Cronbach's alpha = 0.83).

Reinforcement

The parent version of the Illness Behavior Encouragement Scale (IBES)[20] measures the parent's rewarding of the child's pain behavior as perceived by the parent. An additional version was used to measure parental rewarding behavior in situations without pain to determine whether the rewarding of pain behavior reflected a general rewarding or attentive attitude.

The rewarding behaviors covered by the items are, e.g. giving small gift, spending more time than usual with the child, giving special privileges, pampering or spoiling, and excusing the child from regular chores. A higher score indicates more (pain) rewarding. The internal consistency for the parent-report version proved to be high (Cronbach's alpha = 0.78 for mother report and

0.80 for father report). Test-retest reliability scores for the parent-report version of their own rewarding of illness behavior yielded a Pearson correlation coefficient of $r = 0.73$.[20] The internal consistency for the two IBES-scales (parent report about encouragement in a pain situation and parent report about encouragement in a pain-free situation) in the present study was acceptable ($\alpha = 0.76$ and $\alpha = 0.51$, respectively).

Modeling

Pain The pain questionnaire collected demographic data on the parent's date of birth, gender, nationality and educational level.[1] When the parent reported having chronic pain, the same additional information about the pain (intensity, frequency, pain episode duration) was requested as for the adolescents.

Coping Pain coping strategies of the parent were measured with the Pain Coping Inventory (PCI).[25] The parent was asked to indicate how often they used each of the 33 coping behaviors. This questionnaire assesses two higher-order strategies: passive coping (retreating, worrying, and resting) and active coping (pain transformation, distraction, and reducing demands). A higher score indicates the likelihood to use that coping strategy in pain situations. The six scales of the PCI have shown suitable internal consistency for patients with headache ($\alpha = 0.78$, $\alpha = 0.75$, $\alpha = 0.68$, $\alpha = 0.67$, $\alpha = 0.64$, and $\alpha = 0.77$, respectively). As to the convergent validity, the PCI scales were found to correlate with other measures for coping, physical activity and psychological unwell-being. Further, the scales have been found to differentiate between different pain patients in the predicted direction.[25] In the present study, the internal consistency of the PCI was high ($\alpha = 0.85$).

Data analysis

Data were analyzed by frequencies and cross-tabulations. Chi-Square tests and Mann-Whitney U (M-W) tests were used to test differences for categorical variables. Student's t-tests were used to compare the quality of life and psychosocial factors in adolescents with and without chronic pain. Cohen's δ was used to assess effect sizes of our results. Cohen's δ: $0.20 \leq \delta < 0.50$ resembles a small effect size; $0.50 \leq \delta < 0.80$ resembles a medium effect size and $\delta \geq 0.80$ resembles a large effect size.26 Data on reinforcement were analyzed with repeated measures of analysis of variance (ANOVAs). Group (chronic pain versus control) was considered as a between-subject factor. Within-subject factors were the reinforcer (parents versus peers) and situation (pain versus pain-free). Post-hoc paired and independent-sample t-tests were used for further analysis of the significant main and interactions effects. Additionally, Student's t-tests were used to compare differences in psychosocial factors and pain between parents of adolescents with and without chronic pain.

Univariate analyses were performed to determine potential predictor variables. If a given variable had an association with a significance level of $P < 0.15$, the variable was included in the multiple regression analysis. This significance level was chosen to minimize the risk of excluding variables which might show, in combination with other variables, a relation with the dependent variable. Logistic regressions were fit to predict the categorical variable of chronic pain (yes/no) for boys and girls separately. For the adolescents with chronic pain, multiple linear regression models were fit to successively predict pain intensity, pain frequency and pain episode duration for boys and girls separately. Final models contained only those variables with a significance level of $P < 0.10$. We used this significance level to minimize the risk of excluding a variable that contributes significantly to the dependent variable but might be repressed by the other variables.

Results

Response

From the 548 adolescents (and their parents) that were initially approached, 372 adolescents and 323 parents returned completed questionnaires; this gave a response rate of 67.9% and 58.9%, respectively. Two adolescents who reported chronic pain as the result of a diagnosed chronic disease were excluded, leaving a study sample of 370 adolescents. Table 1 shows the demographic characteristics of the adolescents and their parents.

Table 1: Demographic characteristics of the study sample

	Adolescents with chronic pain % (n)	Adolescents without chronic pain % (n)	Parents of adolescents with chronic pain % (n)	Parents of adolescents without chronic pain % (n)
Sex				
Male	26 (58)	32 (47)	7.1 (13)	5.8 (7)
Female	74 (164)	68 (101)	92.9 (170)	94.2 (120)
Age[a] (yrs)				
Male	15.2 (12.8-18.6)	15.3 (13.1-18.4)	46.1 (38.6-53.5)	47.5 (40.4-62.4)
Female	15.1 (13.0-18.1)	14.1 (13.0-17.7)	43.9 (34.6-58.1)	44.6 (31.7-55.2)

[a] Data are mean (range)

Of the 370 adolescent responders, 60% (*n* = 222) reported having chronic pain and 72% (*n* = 265) were girls. Of the 323 responding parents, 13 had to be excluded because their child failed to complete the questionnaire. Ninety-four percent of the parent responders were female. Almost 60% (*n* = 178) of 301 parents (nine missing values) reported having chronic pain complaints themselves.

Pain

Headache (*n* = 70), back (*n* = 43), limb (*n* = 38) and abdominal (*n* = 37) pain were the most frequently mentioned pain complaints in 33, 20, 18 and 17% of the adolescents with chronic pain, respectively. Boys were significantly more inclined to report back pain (29.1%) and limb pain (27.3%), whereas girls were significantly more inclined to report headache (35.8%) and abdominal pain (21.0%).

The mean intensity of the pain in the previous month was 43 mm (SD = 22). The intensity of chronic pain was significantly higher for girls than for boys (46 mm versus 33 mm; $t(215) = 3.95$, $P < 0.001$).

Of the chronic pain sufferers, 51% reported having pain complaints at least twice a week. For pain duration, almost 20% of the adolescents with chronic pain reported that their pain complaints persisted for at least 1 - 2 days. There were no differences between boys and girls regarding frequency and duration of pain episode.

Adolescents with chronic pain experienced significantly poorer quality of life in all domains of the QLA-CP (Table 2). Based on Cohen's *δ*, most differences were moderate in size, functional status was, however, strongly impaired in the pain group (Cohen, 1988).

Table 2: Differences in quality of life between adolescents with and without chronic pain[a]

	Adolescents with chronic pain (n=222)		Adolescents without chronic pain (n=148)			
	Mean	(SD)	Mean	(SD)	P-value	Cohen's δ
Quality of life						
Psychological Functioning	2.0	(0.43)	2.3	(0.38)	<0.001	0.65
Social Functioning	1.8	(0.46)	1.9	(0.52)	0.025	0.21
Physical Functioning	2.2	(0.51)	2.5	(0.41)	<0.001	0.68
Functional Status	2.3	(0.44)	2.9	(0.29)	<0.001	1.11
Satisfaction with life in general	67.9	(24.15)	76.1	(21.62)	<0.001	0.36
Satisfaction with health	60.0	(26.13)	77.4	(23.83)	0.001	0.69

[a] Student's t-tests were used to compare adolescent with and without chronic pain (one-sided)

Differences between adolescents with and without chronic pain

Vulnerability

Compared with adolescents without chronic pain, adolescents with chronic pain reported significantly more feelings of insufficiency (*t* (354) = -6.51, *P* < 0.001), less social acceptance by others (*t* (352) = 2.49, *P* = 0.007), higher negative fear of failure (*t* (366) = -3.64, *P* < 0.001), and lower positive fear of failure which leads to better achievement (*t*(368) = 2.45, *P* = 0.008). The effect sizes were small, except for neuroticism which reached medium effect size (Table 3).

Table 3: Differences in psychosocial factors between adolescents with and without chronic pain[a]

	Adolescents with chronic pain (n=222)		Adolescents without chronic pain (n=148)		P-value	Cohen's δ
	Mean	(SD)	Mean	(SD)		
Vulnerability						
Neuroticism	18.9	(10.7)	12.4	(8.6)	<0.001	0.66
Social acceptance	22.2	(2.8)	22.9	(2.3)	0.007	0.25
Negative fear of failure	8.3	(3.6)	6.9	(3.4)	<0.001	0.39
Positive fear of failure	9.0	(4.2)	10.1	(4.1)	0.008	0.26
Reinforcement – adolescents' perspective						
In pain situations						
By parents	21.1	(7.4)	23.1	(7.3)	0.005	0.27
By peers	14.8	(3.3)	15.9	(3.1)	0.001	0.33
In pain-free situations						
By parents	11.2	(4.1)	11.4	(4.4)	ns	0.06
By peers	17.5	(2.8)	18.5	(2.9)	<0.001	0.35
Modeling						
Significant others with pain[b]	65%		53%		0.012	0.24
Number of significant others	1.05	(1.0)	0.72	(0.8)	<0.001	0.36
Coping						
Approach	33.8	(9.3)	34.8	(10.9)	ns	0.10
Problem-focused avoidance	41.1	(11.1)	42.8	(11.7)	ns	0.14
Emotion-focused avoidance	18.6	(6.0)	15.5	(4.7)	<0.001	0.55

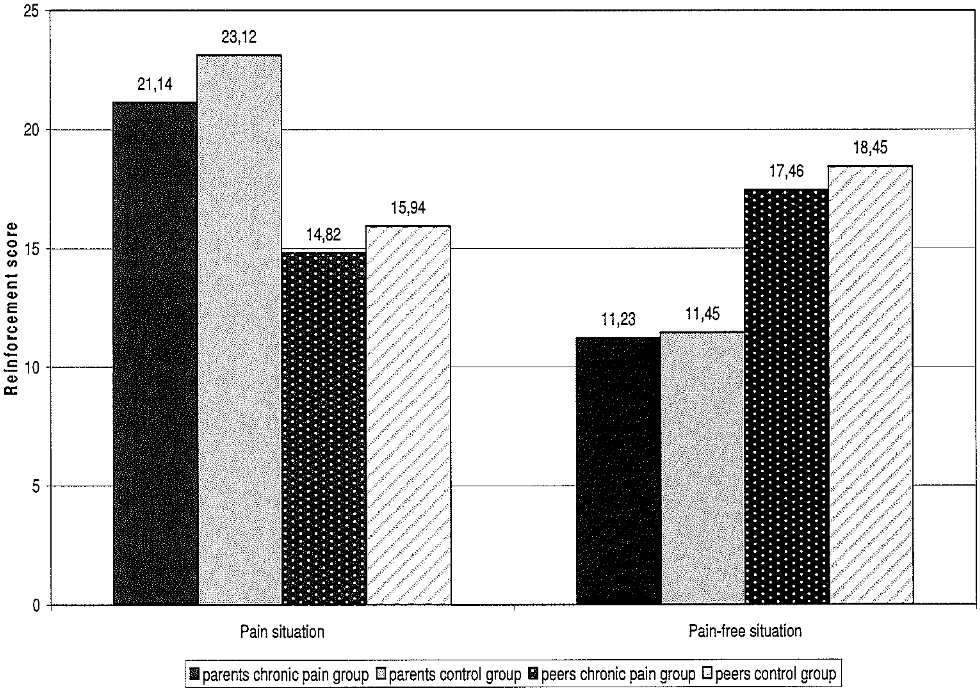
[a] Student's t-tests were used to compare adolescents with and without chronic pain (one-sided). ns, not significant.

[b] Differences were tested by χ^2 test.

Reinforcement

The reinforcement data were analyzed by ANOVAs with group (chronic pain versus control) as between-subject factor, and situation (pain versus pain-free situation) and reinforcer (parents versus peers) as within-subject factors. We found a main effect for situation (F(1,365) = 465.84, P < 0.001), with the highest experienced rewarding in pain situations. An interaction effect was found between situation and reinforcer ($F(1,365) = 1384.37$, $P < 0.001$). Posthoc paired t-tests showed that adolescents experience more rewarding from parents in a pain situation ($t(368) = 31.80$, $P < 0.001$), while they experience more rewarding from peers in a pain-free situation ($t(368) = -18.01$, $P < 0.001$) (Fig. 1). A significant interaction effect was found between group and situation, $F(1,365) = 6.16$, $P = 0.013$. Figure 1 shows that those with chronic pain perceive less rewarding than the control subjects in both pain-free situations ($t(367) = 2.136$, $P = 0.033$; Cohen's $\delta = 0.23$), and to a larger extent in pain situations ($t(366) = 3.366$, $P = 0.001$; Cohen's $\delta = 0.36$). A significant three-way interaction between group, situation and reinforcer ($F(1,365) = 5.14$, $P = 0.024$) underlines that this is, in particular, the case for the parents.

Figure 1



Modeling

Chronic pain sufferers were significantly more likely to report pain in significant others ($\chi^2 = 5.06$, d.f. $= 1$, $P = 0.013$) and reported a higher number of significant others with pain ($t(357) = -3.27$, $P < 0.001$) than their peers without chronic pain. These differences showed small effect size (Table 3).

Coping

With respect to coping with pain, adolescents with chronic pain were more likely to use emotion-focused avoidance coping strategies (t(355) = -5.41, $P < 0.001$); a moderate effect size. No differences between groups were found for problem-focused avoidance and the approach coping strategies (Table 3).

Differences between parents of adolescents with and without chronic pain

No significant differences were found between parents of adolescents with and without chronic pain on their experience of chronic pain, pain parameters, reported feelings of insufficiency, and coping strategies (Table 4).

Table 4: Differences between parents of adolescents with and without chronic pain[a]

	Parents of those with chronic pain (n=183)		Parents of those without chronic pain (n=127)			
	Mean	(SD)	Mean	(SD)	P	Cohen's δ
Vulnerability						
Neuroticism	10.9	(7.7)	9.6	(7.3)	ns	0.17
Reinforcement – parents' perspective						
In pain situations	22.5	(6.5)	24.9	(6.1)	<0.001	0.38
In pain-free situations	7.8	(2.9)	7.8	(3.6)	ns	0.15
Modeling						
Pain parents						
Chronic pain themselves[b]	60%		57%		ns	0.06
Intensity	64.9	(1.0)	66.2		ns	0.04
Frequency[c]	148.9	(34.9)	156.5	(35.3)	ns	0.10
Episode duration[c]	150.9		149.9		ns	0.06
Coping						
Active coping style	26.5	(20.5)	25.5	(4.6)	ns	0.11
Passive coping style	38.7	(20.8)	37.8	(9.5)	ns	0.06

[a] Student's t-tests were used to compare parents of adolescents with and without chronic pain on the other factors (one-sided). ns, not significant.

[b] Differences were tested by χ^2 test.

[c] Differences on mean rank were tested by Mann-Whitney test.

From the parents of chronic pain sufferers, 60% ($n = 108$) reported chronic pain themselves, compared to 57% ($n = 69$) of the parents of adolescents without chronic pain; this difference was not significant. In concordance with the adolescent's perception of parental pain rewarding, parents of adolescents with chronic pain reported less rewarding behavior for their children's pain than parents of adolescents without chronic pain ($t(264) = 3.24$, $P < 0.001$); a small effect size. No differences were found between the parents of both groups in expressing parental rewarding in pain-free situations (Table 4).

Regression Models

Adolescents with and without chronic pain

A logistic regression was fit using an enter elimination procedure to predict the presence of chronic pain (yes/no) for girls and boys.

Girls The odds of having chronic pain in girls increased as the number of pain models, neurotic symptoms and the use of emotion-focused avoidance coping strategies increased. The odds decreased as rewarding from peers in pain situations increased (Table 5).

Boys The odds of having chronic pain in boys increased as neurotic symptoms increased. The odds decreased as the use of problem and emotion-focused approach increased (Table 5).

Adolescents with chronic pain

Multiple regression models were fit using an enter elimination procedure to predict pain intensity, frequency and pain episode duration for girls and boys.

Girls The results revealed that the R^2 for the pain intensity regression model was significantly different from zero, $F(2,157) = 4.08$, $P = 0.019$ for girls. Only one vulnerability variable (neuroticism) contributed significantly to the prediction of pain intensity. The more neurotic the adolescent ($t = 2.44$, $P = 0.016$), the higher the intensity of pain. The model accounted for 4.9% (3.7% adjusted) of the variability in pain intensity.

The R^2 for the regression model for pain frequency was significantly different from zero, $F(3,155) = 13.89$, $P < 0.001$ for girls. One reinforcement variable and two pain locations contributed significantly to the prediction of pain frequency. The higher the rewarding from parents in regular situations without pain ($t = 2.14$, $P = 0.034$) and the presence of abdominal pain ($t = 4.02$, $P < 0.001$), the more frequent the pain. Headache ($t = -5.28$, $P < 0.001$) was related to less frequent pain. This model explained 21.2% (19.7% adjusted) of the variability in pain frequency.

Table 5: Predictors of chronic pain

Variable	*Girls*				*Boys*			
	B	*Sig*	*Exp(B)*	*CI*	B	*Sig*	*Exp(B)*	*CI*
Vulnerability								
Neuroticism	0.04	0.01	1.04	1.00-1.08	0.07	0.01	1.07	1.02-1.13
Social acceptance								
Negative fear of failure								
Positive fear of failure								
Reinforcement								
IBES1[a]								
IBES2[b]	-0.11	0.03	0.90	0.82-0.99				
IBES3[c]								
IBES4[d]								
Modeling								
Pain in family (y/n)								
Pain models	0.35	0.02	1.42	1.05-1.90				
Coping								
Approach					-0.06	0.01	0.95	0.91-0.99
Problem-focused avoidance								
Emotion-focused avoidance	0.09	0.003	1.10	1.03-1.17				
Pain								
Headache								
Abdomen								
Back pain								
Limb pain								
Existence of pain								
Age								

[a] Reinforcement of pain behavior by parents; b Reinforcement of pain behavior by peers;
[c] Reinforcement of behavior by parents in pain-free situations; [d] Reinforcement of behavior by peers in pain-free situations.

The results revealed that the R^2 for the pain episode duration regression model was significantly different from zero, $F(2,152) = 6.13$, $P = 0.003$. Modeling and a coping variable contributed significantly to prediction of the pain episode duration. The number of pain models ($t = 2.30$, $P = 0.023$), and the use of emotion-focused avoidance coping strategies ($t = 2.49$, $P = 0.014$) had a positive effect on pain episode duration. More pain models and more use of emotion-focused avoidance coping strategies are associated with longer pain episode duration. These variables explained 7.5% (6.2% adjusted) of the variability on the pain episode duration

Boys The results revealed that the R^2 for the pain intensity regression model was significantly different from zero, $F(2,44) = 5.67$, $P = 0.006$. Modeling and a coping variable contributed significantly to the prediction of pain intensity. Frequent use of emotion-focused avoidance ($t = 2.278$, $P = 0.028$), the higher the intensity of pain. More use of active coping strategies by the parent ($t = -2.09$, $P = 0.042$) is associated with a lower intensity of pain. The model accounted for 20.5% (16.9% adjusted) of the variability in pain intensity.

The R^2 for the regression model for pain frequency was significantly different from zero, $F(6,42) = 2.35$, $P = 0.048$ for boys. Separately, none of the variables showed a unique contribution to the prediction of pain frequency. The model explained 25.2% (14.5% adjusted) of the variability in pain frequency.

The results show that the R^2 for the pain episode duration regression model was significantly different from zero, $F(5,35) = 6.15$, $P < .001$. Two reinforcement variables, a vulnerability variable and pain location contributed significantly to the prediction of pain episode duration. The more rewarding by peers in a pain-free situation ($t = 2.33$, $P = 0.026$), the shorter the pain episode duration. More rewarding by parents in a pain-free situation ($t = -2.05$, $P = 0.048$), more negative fear of failure ($t = -2.085$, $P = 0.044$), and the presence of headache ($t = -3.58$, $P = 0.001$) resulted in a longer pain episode duration. This model explained 46.8% (39.2% adjusted) of the variability on the pain episode duration.

Discussion

This study compared adolescents with and without chronic pain on several psychosocial factors often reported to influence chronic pain experience. Further, an analysis of the contribution of these factors to chronic pain is provided.

In the current sample of 222 adolescents with chronic pain, the most frequently reported pain complaints (headache, back, limb and abdominal pain) were similar to those reported in a prevalence study by Perquin and colleagues.[1] Our results show that, compared to controls without chronic pain, adolescents with chronic pain experience significantly poorer quality of life in all domains. The effect size of these differences ranged from medium (psychological functioning, physical functioning and satisfaction with health) to large (functional status), except for the domain social functioning.[26] The latter result is in accordance with an earlier report that this latter scale is less reliable and valid than the scales representing the other domains of quality of life.[22]

In line with our hypothesis and earlier studies, we found that adolescents with chronic pain are more vulnerable in terms of neuroticism, fear of failure and less experienced social acceptance than adolescents without chronic pain.

In accordance with operant learning theories, we found a main effect for situation (pain versus pain-free), with the highest rewarding in pain situations. Interestingly, we found that adolescents experience the most rewarding by parents in pain situations, while they experience more rewarding by peers in a pain-free situation. An explanation for this finding might be that adolescents with pain withdraw from social situations and do not tell their peers. By staying home and thus avoiding school or other activities, rewarding of pain behavior is more likely from parents than from peers. While parents are more rewarding in pain situations, this was found to be significantly less in the chronic pain group than in the controls. This finding does not support our hypothesis, but is consistent with Osterhaus and colleagues who found a negative association between the parents' rewarding and the child's pain report.[11] Nevertheless, their explanation that the attention children receive when they are in pain should be seen as 'regular' parental care is not substantiated in our study. If so, we would also have found a similar difference for rewarding in pain-free situations, which was not the case.

Our results indicate that adolescents with chronic pain might be rewarded less often than one might expect on the basis of learning principles. Reasons for this may be that the family has grown accustomed to the pain and responds less, or that loss of credibility leads to adolescents with chronic pain receiving less attention for their pain behavior. In accordance with this explanation, Jamison and colleagues found that chronic pain patients perceived their families as non-supportive.[27] It is conceivable that a child's pain report will increase as a function of their parents' lack of attention to its complaints. Again, we emphasize that our sample was drawn from the general population. It is conceivable that operant learning theories are more applicable to adolescents from a clinical setting with more severe pain from a clinical setting. More research on the attitude and behaviors of parents towards the pain complaints of their children is needed. A possible limitation of our study on reinforcement concerns the use of self-reports of behavior. On the other hand, we do not rely only on the parents' report but also on the report of the child.

The finding that adolescents with chronic pain report more significant others with pain than adolescents without chronic pain supports our hypothesis and was consistent with other studies, but the difference is rather small.[12,13]

Overall, we found significant differences between adolescents with and without chronic pain. Neuroticism and pain coping strategies of the adolescents showed moderate effect sizes in the present study, and should be considered as differences with potential clinical importance. This deserves to be examined in further research.

As for the parents, we found no support for our hypothesis. No differences were found between parents of adolescents with and without chronic pain on their experience of chronic pain and the pain parameters. The same was true for the coping strategies parents used and the vulnerability of the parents. This latter finding is in contrast to Hotopf and colleagues who found that mothers of children with abdominal pain are more anxious (in terms of higher neuroticism scores).[28]

Analysis of the contribution of psychosocial factors to chronic pain showed that having chronic pain is associated with vulnerability (i.e. neuroticism) and pain coping strategies for both boys and girls. The number of pain models and the experienced rewarding by peers when in pain are also associated with chronic pain in girls. Although, more psychosocial factors are associated with chronic pain in girls, the psychosocial factors seem to play a larger role in predicting pain parameters for boys, considering the explained variance. The different associations found between the psychosocial factors and the pain parameters refer to the extensiveness of pain. However, due to the study's case-control design, the caused direction of the relation remains unclear. Despite this, psychological treatment aimed at pain management should consider multiple factors related to chronic pain rather than focusing on one factor only.
The gender differences in our study are in accordance with those reported in other studies.[9,29] Once again, these results underline the importance that pain management programs, take gender differences into account and embrace a variety of elements effective for both boys and girls.

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Chapter 5

Psychosocial factors associated with quality of life in adolescents with chronic pain

Submitted

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Abstract

This study examined the relationships between pain characteristics, psychosocial factors and quality of life among adolescents with chronic pain that existed for at least three months, either recurrently (i.e. pain with pain-free intervals) or continuously. We conducted a cross-sectional study in 194 adolescents aged 12 through 18 years who completed questionnaires on pain, psychosocial factors (i.e. vulnerability, reinforcement, modeling and coping) and quality of life, and also kept a diary about their pain complaints for 3 weeks. Multiple hierarchical regression analyses revealed that psychosocial variables accounted for a significant variance in the adolescent's quality of life, even when controlling for pain characteristics. Analysis of the independent variables showed that pain intensity and vulnerability contributed significantly and uniquely to the variance of most quality of life domains. In addition, we found that emotion-focused avoidance coping strategies strengthen the negative relation between pain intensity and psychological functioning. Beside pain, psychosocial factors (vulnerability, reinforcement, modeling and coping) are strongly associated with quality of life in adolescents with chronic pain. These results may contribute to interventions focused on psychological adaptation in young pain patients in order to improve their quality of life.

Introduction

The prevalence of chronic pain increases in adolescence.[1] Common complaints are headache, back pain and limb pain,[1-3] often resulting in considerable functional disability[4,5] and use of health services.[6] A number of psychosocial factors have been associated with the onset, exacerbation, and/or maintenance of chronic pain in adolescents. The most frequently studied factor is psychological vulnerability (i.e. feelings of inadequacy or insufficiency). Recurrent pain is related to psychological distress, anxiety and depression in adolescents.[7-11] Passchier and Orlebeke found that school-related stress, negative fear of failure, and headache complaints in children and adolescents are positively correlated.[12] Children complaining of headache showed a tendency towards high achievement motivation at school.[13]

Beside vulnerability, adolescents with chronic pain differ from healthy controls on psychosocial factors such as modeling, reinforcement and coping.[14] The vulnerability and coping strategies of the adolescent further affect the intensity of pain. Higher vulnerability and more use of emotion-focused avoidance coping strategies are associated with more intense pain. [14]

Chronic pain has substantial impact on the life of children and adolescents[15-18] as revealed by significantly worse psychological functioning, physical functioning, functional status, lower satisfaction with life, poorer health status [14,19] and social functioning[18] in adolescents with chronic pain. A high intensity and frequency of the pain is related to a lower self-reported quality of life in most quality of life domains. [16,20] Although chronic pain influences the quality of life, the explained variance of quality of life by pain characteristics was shown to be relatively low.[16,21] These results suggest that, although pain is an important factor, it may have only a limited influence on quality of life in children and adolescents with chronic pain. Pre-existing characteristics, such as age and gender, as well as psychosocial factors might also influence the quality of life in adolescents with chronic pain.

The influence of psychosocial factors on quality of life has mostly been studied in headache patients. The ability to cope with pain and adaptive family daily routine predicted better quality of life in children[22] while the vulnerability was related to lower quality of life in adolescents[23]. Results are conflicting with regard to the influence of family characteristics. Some studies found that the way parents deal with the pain of a child is associated with adjustment outcomes of the child, such as quality of life[22,24], while others concluded that family characteristics and social support are not significant predictors of quality of life.[25]

Psychosocial factors were also found to moderate (i.e. weaken or strengthen) the relation between pain and quality of life, for instance vulnerability and coping. In adolescents with high experienced (school) stress, more headache was associated with less quality of life but not in those with low stress.[23] The relation between irritable bowel symptoms and disability was stronger for adolescents with lower levels of perceived academic competence.[26] In addition, coping was found to strengthen the relation between the severity of juvenile rheumatoid arthritis and adjustment.[24] Stress was inversely related to adjustment, but at levels of high stress coping acted as a stress buffer. Although considerable research has focused on the relationship between pain, psychosocial factors (vulnerability, family factors and coping) and quality of life of adolescents with a specific chronic pain condition, few studies examined the influence of these factors side-by-side or studied the moderating effect of psychosocial factors on the relation between pain (irrespective of pain location) and quality of life.

Therefore, this study was designed to address the following questions: 1) Which psychosocial factors have a unique contribution (independent of relationships with pain) to quality of life of adolescents with chronic pain? 2) Which psychosocial factors moderate the relation between pain and quality of life? 3) Is there a difference in the influence of psychosocial factors in adolescents with high and low pain intensity on quality of life? Knowledge on these items may enable psychological interventions to better focus on specific psychological processes that influence the pain-related quality of life.

Methods

Study population

A total of 1607 adolescents aged 12 through 18 years (48% boys and 52% girls) from the general Dutch population completed a short structured pain questionnaire in the classroom. They were all students of six secondary schools in Rotterdam, selected by the Municipal Health Services in order to reach a representative group of adolescents. The questionnaire was administered to students of all years and educational levels.

The current study focused on adolescents with chronic pain with unknown organic etiology that existed for three months or longer, either recurrently (i.e. pain with pain-free intervals) or continuously. For this, we applied the definition of pain introduced by the International Association for the Study of Pain (IASP).[27] Adolescents were asked whether they had consulted a family physician or specialist for their pain. They were asked to report the diagnosis of the doctor. Adolescents with pain resulting from a specific chronic disease (e.g. rheumatic arthritis, malignancies) were excluded.

Of the 1607 adolescents, 631 adolescents (39%) reported having chronic pain with unknown organic etiology and 330 of these adolescents were willing to participate in the present study. This group was older (Student's *t*-tests, $t\ (629) = -2.69$, $P < 0.01$), reported more frequent pain (Mann-Whitney *U*-test, $Z = -2.98$, $P < 0.01$) and higher pain intensity in the previous month ($t\ (592) = 4.35$, $P < 0.001$) than those who were not willing to participate. No differences were found between participants and non-participants for gender, duration of pain episodes and pain location.

Procedure

A booklet of questionnaires and a pain diary were mailed to the adolescents. The adolescents were first invited to complete the questionnaires and to keep the diary to register pain intensity for three successive weeks. Adolescents with more than one chronic pain location were asked to refer only to the pain that generally troubled them most.

To increase the response rate, the adolescents were telephoned after one week to remind them to complete and return the booklet and the diary; when this was done they received a compact disc voucher (value 5 euros).

Questionnaires

Demographic data

Demographic data included the adolescent's date of birth, gender, nationality, educational level and current school year.

The Pain Questionnaire collected information about the pain: location, frequency (answering options ranged from less than once a month to every day), duration of the pain episodes (answering options ranged from less than 30 minutes to one week or longer), history (number of months the adolescent experienced pain) and pain intensity in the previous month (using a Visual Analogue Scale; VAS), were measured with a retrospective format to select adolescents with chronic pain.[1]

Vulnerability

Vulnerability was measured with the Inadequacy Scale of the Dutch Personality Questionnaire–Junior.[28] The scale contains 28 items with response categories ranging from 0 to 2. Examples of items are: 'I often think that I am not worthwhile' 'I often worry about the way I look'. Adolescents with a high score are more likely to describe themselves as tense and fearful and report more feelings of insufficiency; they are frequently characterized as having difficulty in making contact with others. The internal consistency of the inadequacy scale is $\alpha = 0.87$.[28] As to the validity, the scale has a high correlation with other questionnaires which measure the same construct.[29] In the present study the internal consistency for this scale was also $\alpha = 0.87$.

Reinforcement

We translated the Illness Behavior Encouragement Scale (IBES) to measure the extent to which pain behavior is rewarded.[30] This questionnaire measures the rewarding of pain behavior by parents (12 items) and peers (6 items) as perceived by the child. For both scales the response categories ranged from 0 to 4, with a higher score indicating more rewarding of (pain) behavior. The internal consistency for the child-report version regarding parents proved to be high and support was found for the validity of the IBES.[30] Test-retest reliability scores for the child-report versions yielded Pearson correlation coefficients of $r = 0.77$ and $r = 0.72$ for maternal and paternal rewarding of illness behavior, respectively.[30] In the present study the internal consistency for the two IBES scales (rewarding by parents and peers in a pain situation) is 0.78 and 0.55, respectively.

Modeling

To assess modeling, adolescents were asked whether they had significant others with pain complaints. When the answer was positive, additional information was requested about their relationship (i.e. a father or mother) and the location of the pain complaints of these significant others.

Coping

We assessed pain coping strategies of the participants with a translated version by Bandell-Hoekstra and colleagues[31] of the Pain Coping Questionnaire (PCQ).[32] This questionnaire assesses three higher-order strategies: Problem- and emotion-focused approach (14 items about information seeking and problem solving), Problem-focused avoidance (15 items about behavioral and cognitive distraction, and positive self-statements) and Emotion-focused avoidance (10 items about externalizing, catastrophizing behavior). For all strategies, the response categories ranged from 0 to 5, with a higher score indicating the likelihood to use the coping strategies in pain situations. The subscales and higher-order scales are internally consistent.[32] Factor analysis on the translated version confirmed the subscales that were previously reported by Reid and colleagues[32], and a test-retest showed moderate to high reproducibility of the PCQ.[31] In the present study, the internal consistency for the higher-order scales was also high (Cronbach's alpha = 0.89 for Approach; α = 0.90 for Problem-focused avoidance; α = 0.81 for Emotion-focused avoidance).

Quality of life

To measure the quality of life we used the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP).[33] The QLA-CP is subdivided into six domains, each measuring a particular aspect of quality of life: (1) Psychological Functioning, (2) Functional Status, (3) Physical Functioning, (4) Social Functioning, (5) Satisfaction with Life in General, and (6) Satisfaction with Health. The first four domain scores were obtained by averaging the subscale scores pertaining to a domain. This resulted in domain scores ranging from 0 to 3. The last two domains (5 and 6) were measured with a VAS; a 100-mm long horizontal line with the anchors 'completely dissatisfied' and 'completely satisfied'. A higher score on each domain of the QLA-CP represents a better quality of life. The QLA-CP has shown suitable internal consistency and construct validity against the Dartmouth Primary Care Cooperative Information Project (COOP) Functional Health Assessment Charts/WONCA (COOP/WONCA charts).[33] In addition, it discriminates between adolescents with chronic pain on both intensity and frequency of the pain.[33] In the present study the first four QLA-CP domains showed internal consistencies of α = 0.88, 0.80, 0.64 and 0.64, respectively.

Diary

To assess the pain intensity (prospectively measured), adolescents were asked to register their pain for three successive weeks. Pain intensity was recorded three times daily, at breakfast, dinnertime and bedtime with a VAS with the anchors 'no pain' and 'the worst pain you can imagine'. The adolescents were asked to mark a position on the VAS that best matched their pain at that moment. The VAS is a valid measure for the assessment of self-reported pain intensity in chronic pain patients.[34] Participants with more than 25% missing VAS values in the diary were excluded from analyses.

Data analysis

Prospective pain intensity scores consisted of the means of all pain intensity recordings. These scores were calculated by adding up all pain intensity scores (from 0 to 100) and dividing them by the number of recording times (3 weeks * 3 times daily = 63). We were also interested in the fluctuations in pain, since these give an indication of the nature and characteristics of the pain attacks. Pain attacks that show high fluctuations in time might have a greater impact on the quality of life of the adolescents due to their unpredictable occurrence. Pain fluctuations were calculated by measuring the average difference between pain intensities for each patient at three recording times: breakfast, dinnertime and at bedtime. Based on the median difference of all participants for each recording time as cut off, the individual average score was classified as reflecting a high or low fluctuation of pain intensity for breakfast, dinner and bedtime. When an adolescent had a high fluctuation at all recording times he or she was considered as having pain with high fluctuations.

To explore the inter-relationships between demographic factors, pain characteristics and psychosocial factors, Pearson correlations were computed for continuous variables. For ordinal variables, Spearman rank-order correlations were computed. To reduce the risk of type I errors, given that there were 120 correlations in the analyses, the alpha level was set at 0.001 for all correlations reported here.

Univariate analyses were performed to examine the extent to which the individual variables were associated with the quality of life domains. If a given variable had an association with a significance level of $P < 0.15$, that variable was included in the multiple regression analysis.

Multiple hierarchical regression models were fit using a stepwise elimination procedure. Demographic variables (age and gender) were entered as the first block in the regression analysis to control for any influence they might have on quality of life. The pain characteristics were entered as the second block in the regression analysis to control for any influence pain might have on the quality of life domains. Psychosocial factors were entered in the third block, and the fourth block contained interactions between prospective pain intensity and the psychosocial factors. Interaction variables were used to examine the presence of variables which could moderate the association between pain and quality of life in adolescents. For this, the psychosocial factors with interval format were dichotomized using the median scores on cut off.

Tabachnick and Fidell[35] suggest that multicollinearity may be a problem if the Pearson correlation between two variables in a multivariate analysis is above 0.70. Inspection of Pearson correlation coefficients indicated that there was a problem of multicollinearity between the interaction variables. Correlations between predictor variables in the same multivariate analysis were less than 0.01 for the first block of the variables and 0.51 for the second and third block of the variables. Correlations higher than 0.70 were found in the fourth block. The interaction terms with the lowest correlations (with the dependent variable) were excluded from this block in the regression analysis (see Table 2).

For subgroup analysis, we focused on adolescents in the lowest 25 percentile of prospective pain intensity (the low pain group) and adolescents in the highest 25 percentile of prospective pain intensity (the high pain group). Differences between subgroups were analyzed by cross-tabulation, Chi-square tests and Mann-Whitney *U* (M-W) tests for categorical variables. Student's *t*-tests were used to compare the psychosocial factors between the subgroups. Cohen's δ was used to assess effect sizes of these results. Cohen δ: $0.20 \leq \delta < 0.50$ is considered a small effect size; $0.50 \leq \delta < 0.80$ a medium effect size and $\delta \geq 0.80$ a large effect size (Cohen, 1988). Further, we performed the multiple hierarchical regression analyses for both subgroups.

Results

Respondents

Of the 631 eligible adolescents with chronic pain, 330 adolescents were willing to participate and received the questionnaire and diary. Of these, 224 adolescents responded, giving a response rate of 35%. Two adolescents who reported chronic pain as the result of a diagnosed chronic disease were excluded. Of the respondents, 3 adolescents omitted greater than 25% of the reporting periods and 25 (11%) returned only the questionnaire but not the diary and were excluded from analysis, leaving a study sample of 194 adolescents. Compared to the original screening population of adolescents with chronic pain, adolescents in our final sample reported more frequent (M-W, $Z = -3.18$, $P = 0.01$) and more intense pain in the previous month (t (258.56) = - 4.66, $P < 0.001$). No differences were found between participants and non-participants for age, gender, duration of a pain episode and pain location, retrospectively measured.

The sample consisted of 145 (75%) girls and 49 (25%) boys. The average age was 14.7 years (Standard Deviation (SD) = 1.4 years).

Headache (n = 59), back (n = 38), limb (n = 36) and abdominal (n = 34) pain were the most frequently mentioned pain locations in 30%, 19%, 18% and 17% of the adolescents with chronic pain, respectively. Table 1 shows the characteristics of the adolescents for all pain locations. Overall, the mean intensity of pain in the previous month was 41.7 mm (SD = 22.6). Most adolescents reported recurrent pain complaints (90%) and the mean pain intensity in three weeks (over 63 recording moments) was 16.1 mm (SD = 14.7). For subgroup analysis, 48 adolescents in the lowest 25 percentile of prospective pain intensity (the low pain group) were compared with 48 adolescents in the highest 25 percentile of prospective pain intensity (the high pain group).

Table 1: Characteristics of the study population (n=194)

	Scale range	**Total sample mean (SD)**
Demographic data		
Age (*years*)	12 - 18	14.7 (1.4)
Gender (*N, %*)		
Boys		49 (25.3)
Girls		145 (74.7)
Pain characteristics		
Intensity – prospective (VAS)[1]	0 – 100	16.1 (14.7)
Intensity – retrospective (VAS)[1]	0 – 100	41.7 (22.6)
Course of the pain *(N,%)*		
Continuous		19 (9.8)
Recurrent		175 (90.2)
Psychosocial Factors[2]		
Vulnerability		
Neuroticism	0 – 56	18.3 (10.3)
Reinforcement		
Rewarding by parents	0 – 48	21.2 (7.4)
Rewarding by peers	0 – 24	14.9 (3.4)
Modeling (N,%)		
Father in pain		45 (23.2)
Mother in pain		69 (35.6)
Coping		
Approach	0 – 70	33.9 (9.4)
Problem-focused avoidance	0 – 75	41.4 (11.2)
Emotion-focused avoidance	0 – 50	18.4 (5.8)
Quality of life[3]		
Psychological Functioning	0 – 3	2.0 (0.4)
Physical Functioning	0 – 3	2.2 (0.5)
Functional Status	0 – 3	2.4 (0.4)
Social Functioning	0 – 3	1.8 (0.5)
Satisfaction with life in general (*VAS*)	0 – 100	67.9 (24.3)
Satisfaction with health (*VAS*)	0 - 100	60.6 (26.4)

1) The higher the scores the more intense the pain

2) The higher the scores the higher the psychosocial factors (more vulnerable, more rewarding, more pain models and more use of coping strategies)

3) The higher the scores the better the self-reported quality of life of the adolescents

Preliminary analyses

Table 2 gives the inter-relationships between the investigated variables and the quality of life domains.

None of the demographic variables were significantly correlated with any of the quality of life domains. Pain intensity (prospectively measured) correlated negatively with all quality of life domains, except Social Functioning. Other pain characteristics correlating with quality of life were fluctuation, intensity (retrospectively measured) and frequency. The greater the fluctuation in pain, the higher the pain intensity and the more frequent the pain, the lower the quality of life. Of the psychosocial factors, especially vulnerability and coping correlated with most quality of life domains. The higher the scores on vulnerability, the greater the use of approach coping and the greater the use of emotion-focused avoidance coping, the lower the quality of life. Interaction effects were related to Physical Functioning and Functional Status. For adolescents with high scores on psychosocial variables, higher levels of pain intensity (prospectively measured) were associated with lower quality of life.

Overview of regression analyses

Multiple hierarchical regression models were fit using an enter elimination procedure to examine the relation between demographic variables, pain characteristics, psychosocial factors, interaction variables and quality of life. The results are given in table 3. All variables are presented, but only those included in the multiple regression analysis are presented with their standardized regression coefficient.

Pain characteristics

At the second stage of the analyses, the pain characteristics were entered as a block into the regression equation. The results indicated that pain characteristics explained an additional amount of the variance of the quality of life domains Psychological Functioning (15%), Physical Functioning (17%), Functional Status (29%), Satisfaction with Life in General (14%)and Satisfaction with Health (13%). When these variables were examined individually, only pain intensity (prospectively measured) was independently and significantly associated with all quality of life domains. Higher pain intensity was associated with lower quality of life. History of the pain was significantly associated with the quality of life domains Psychological Functioning and Satisfaction with Life in General. The longer the pain existed, the lower the quality of life. Finally, pain frequency was significantly associated with Functional Status. The more frequent the pain, the lower the quality of life.

Psychosocial factors

After controlling for the effects of demographic variables and pain characteristics, the block of psychosocial factors explained an additional amount of the variance of the quality of life domains Psychological Functioning (29%), Physical Functioning (15%), Functional Status (5%), Social Functioning (23%), Satisfaction with Life in General (30%) and Satisfaction with Health (7%).

Table 2: Pearson correlations between pain, psychosocial factors and quality of life scores

	Psychological Functioning[1]	**Physical Functioning**[1]	**Functional Status**[1]	**Social Functioning**[1]	**Satisfaction with life**[1]	**Satisfaction with health**[1]
Demographics						
Age	- 0.09	- 0.06	0.00	0.05	- 0.09	- 0.05
Pain[2]						
Pain-intensity prospective	- 0.35*	- 0.38*	- 0.47*	- 0.03	- 0.28*	- 0.32*
^ Fluctuation	- 0.25*	- 0.31*	- 0.38*	- 0.04	- 0.18	- 0.25*
^ Course of the pain	- 0.17	- 0.13	- 0.15	- 0.09	- 0.10	- 0.21
Pain-intensity retrospective	- 0.40*	- 0.32*	- 0.30*	- 0.00	- 0.24*	- 0.15
^ Frequency	- 0.21	- 0.28*	- 0.45*	- 0.03	- 0.13	- 0.25*
^ Duration	0.05	- 0.08	- 0.15	0.03	- 0.05	- 0.04
History	- 0.17	- 0.09	- 0.10	0.00	- 0.18	- 0.07
Psychosocial factors[3]						
Vulnerability	- 0.56*	- 0.45*	- 0.23*	- 0.41*	- 0.54*	- 0.17
Reinforcement						
Rewarding by parents	0.07	0.04	- 0.03	0.15	0.13	0.02
Rewarding by peers	0.17	- 0.01	- 0.06	0.27*	0.08	0.06
Modeling						
^Mother with pain	- 0.02	- 0.08	- 0.17	0.05	- 0.01	0.00
^Father with pain	- 0.08	- 0.05	- 0.03	0.05	- 0.02	- 0.13
Coping						
Approach coping	- 0.17	- 0.23*	- 0.25*	0.05	- 0.25*	- 0.15
Problem-focused avoidance	0.08	- 0.07	0.01	0.09	0.04	0.09
Emotion-focused avoidance	- 0.50*	- 0.40*	- 0.25*	- 0.23*	- 0.39*	- 0.32*
Interactions						
Pain & neuroticism	- 0.44*	- 0.37*	- 0.28*	- 0.23*	- 0.36*	- 0.20
Pain & rewarding parents	- 0.11	- 0.08	- 0.20	0.09	- 0.09	- 0.12
Pain & rewarding peers	- 0.06	- 0.21	- 0.25*	0.13	- 0.10	- 0.21
Pain & mother with pain	- 0.24	- 0.33*	- 0.43*	0.04	- 0.27	- 0.11
Pain & father with pain	- 0.26	- 0.34*	- 0.33*	0.01	- 0.17	- 0.29*
Pain & approach coping	- 0.31*	- 0.33*	- 0.39*	0.01	- 0.29*	- 0.30*
Pain & problem-focused coping	- 0.06	- 0.17*	- 0.10	0.02	- 0.07	- 0.09
Pain & emotion-focused coping	- 0.47*	- 0.42*	- 0.39*	- 0.09	- 0.34*	- 0.27*

* $P \leq 0.001$ ^ Spearman rank-order correlation, a nonparametric version of the Pearson correlation coefficient was used to analyse ordinal data

1) The higher the scores the better the self-reported quality of life of the adolescents

2) The higher the scores the less favorable the level of pain

3) The higher the scores the higher the psychosocial factors (more vulnerable, more rewarding, more pain models and more use of the specific coping strategies)

Table 3: Predictors of quality of life (Multiple hierarchical regression analyses)

	Psychological Functioning				**Physical Functioning**				**Functional Status**				**Social Functioning**				**Satisfaction with life**				**Satisfaction with health**			
	R^2	ΔR^2	ΔF	Beta ^{b}	R^2	ΔR^2	ΔF	Beta ^{b}	R^2	ΔR^2	ΔF	Beta ^{b}	R^2	ΔR^2	ΔF	Beta ^{b}	R^2	ΔR^2	ΔF	Beta ^{b}	R^2	ΔR^2	ΔF	Beta ^{b}
1. Demographics					0.02	0.02	3.08^{T}																	
Age																								
Gender								- 0.13^{T}																
2. Pain characteristics	0.15	0.15	4.65***		0.18	0.17	8.97***		0.29	0.29	13.93***		0.01	0.01	2.33		0.14	0.14	5.87***		0.13	0.13	5.11***	
Pain intensity – prospect.				- 0.17^{T}				- 0.19*				- 0.19*								- 0.24*				- 0.21*
Fluctuation				- 0.08				- 0.12				- 0.09								- 0.05				- 0.12
Course of the pain				0.04																				0.08
Intensity – retrospect.				- 0.07				- 0.12				- 0.12								- 0.09				0.04
Frequency				- 0.11				- 0.12				- 0.29***												- 0.08
Duration												- 0.09				0.11								
History				- 0.18*																- 0.17*				
Pain location																								
													0.24	0.23	13.75***									
3. Psychosocial Factors	0.44	0.29	20.30***		0.33	0.15	7.43***		0.33	0.05	2.90*					- 0.35***	0.43	0.30	15.01***		0.20	0.07	3.53**	
Neuroticism				- 0.40***				- 0.33***				- 0.12^{T}				0.11				- 0.46***				- 0.03
Rewarding by parents																0.19**				0.16*				
Rewarding by peers				0.15*																				
Mother with pain								- 0.01				- 0.07								0.05				
Father with pain								- 0.12^{T}																- 0.15*
Approach				- 0.05				- 0.08				- 0.17*								- 0.09				- 0.07
Problem-focused avoidance																- 0.07								
Emotion-focused avoidance				- 0.21**				- 0.09				0.08								- 0.10				- 0.19*
													0.25	0.01	0.86									
4. Interactions	0.48	0.04	2.58*		0.35	0.02	0.73		0.36	0.02	0.96					- 0.03	0.44	0.01	0.70		0.21	0.01	0.59	
Pain & neuroticism				- 0.05				0.03				0.18								0.07				0.09
Pain & rewarding parents				0.11								0.09				0.11								0.11
Pain & rewarding peers								- 0.01				0.02												- 0.06
Pain & mother with pain												0.08												
Pain & father with pain				- 0.11				- 0.13				- 0.04								0.00				- 0.10
Pain & approach coping																								- 0.11
Pain & problem-focused								- 0.04				0.04												
Pain & emotion-focused				- 0.26*				- 0.17												- 0.14				

a $^{*}P \leq 0.05$; $^{**}P \leq 0.01$; $^{***}P \leq 0.001$; $^{T}P \leq 0.10$

b Standardized regression coefficient

c ΔR^2 = R Square Change; ΔF = F change

An examination of the beta weights associated with the psychosocial factors indicated that in particular vulnerability was independently and significantly associated with almost each domain, i.e. Psychological Functioning, Physical Functioning, Social Functioning and Satisfaction with Life in General. The higher the vulnerability scores, the lower the quality of life of adolescents. Rewarding by others was significantly but positively associated with Psychological Functioning, Social Functioning and Satisfaction with Life in General. The presence of a father with pain was significantly related to lower Satisfaction with Health. Finally, the coping strategy problem- and emotion-focused approach was significantly associated with Functional Status, while emotion-focused avoidance coping was significantly associated with Psychological Functioning and Satisfaction with Health. The more those coping strategies were used, the lower the quality of life.

Interactions

Interaction effects accounted for an additional 4% in the variance of Psychological Functioning beyond that explained by the pain characteristics and psychosocial factors. However, the beta weights associated with the interactions indicated that only the interaction between pain intensity and emotion-focused avoidance coping was independently and significantly associated with Psychological Functioning (see Figure 1). For adolescents who reported higher levels of emotion-focused avoidance coping, higher levels of pain intensity were associated with lower levels of Psychological Functioning. In contrast, for adolescents reporting lower levels of emotion-focused coping, pain intensity level was not associated with the levels of Psychological Functioning.

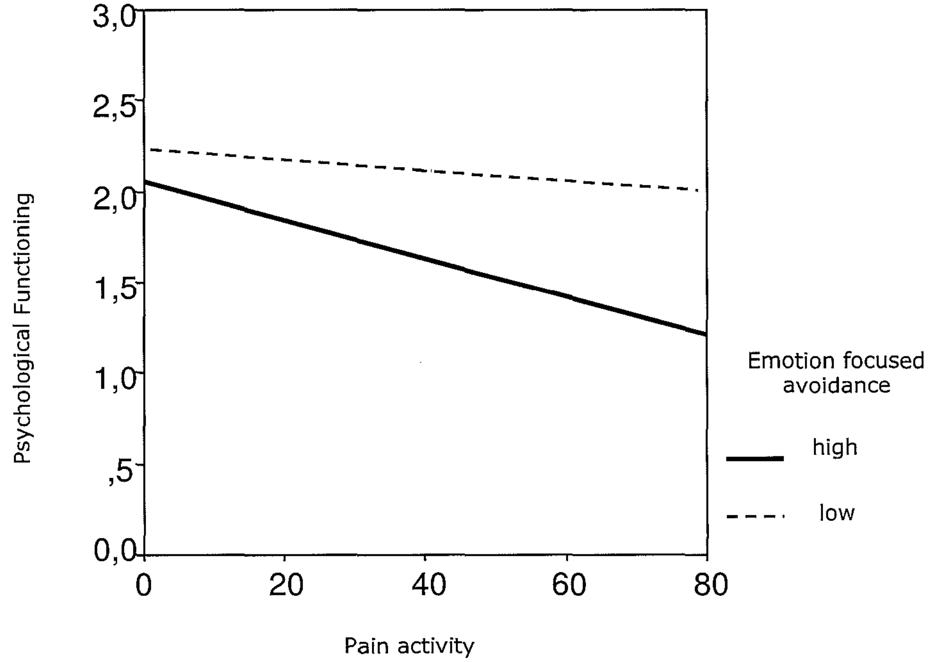
High and low pain

Besides the obvious differences in pain characteristics, we found that the adolescents from the high pain group (i.e. adolescents in the highest quartile) were more vulnerable ($t(94) = -3.29$, $P = 0.001$), were more likely to use emotion-focused avoidance coping strategies ($t(94) = -4.46$, $P < 0.001$) and more often had a mother with chronic pain ($\chi^2 = 5.10$, d.f. = 1, $P = 0.02$) than those from the low pain group. Further, adolescents with intense pain suffer a significant reduction in all quality of life domains, except for Social Functioning. The meaningfulness of the differences in quality of life ranged from moderate effect sizes for Satisfaction with health ($t(92) = 4.11$, $P < 0.001$) (Cohen's $\delta = 0.67$) to large effect sizes for Psychological Functioning ($t(83.2) = 4.37$, $P < 0.001$), Functional Status ($t(94) = 7.81$, $P < 0.001$) and Physical Functioning ($t(94) = 5.41$, $P < 0.001$) (Cohen's $\delta > 0.80$).

The multiple hierarchical regression analysis showed that our model explained a larger amount of variance of most quality of life domains for those with high pain compared to those with low pain (Psychological Functioning: 68% versus

48%; Physical Functioning: 73% versus 34%; Functional Status: 38% versus 35%; Social Functioning: 29% versus 40%; Satisfaction with life: 64% versus 41%; and Satisfaction with health: 34% versus 30%, respectively). Further, we found that the psychosocial factors explained considerably larger amounts of variance of Psychological Functioning (53% versus 4%) and Physical Functioning (56% versus 10%) for adolescents in the high pain group than for those in the low pain group. For the other quality of life domains, the explained variance by the psychosocial factors was about the same for both groups.

Figure 1: Significant effects of interactions between Emotion Focused Avoidance Coping (High, Low) and pain activity on the quality of life domain Psychological Functioning.



Discussion

The present study examined the association between demographic factors, pain characteristics, psychosocial factors and quality of life for adolescents with chronic pain with unknown organic etiology. Overall, we found that demographics did not account for a significant proportion of the variance in quality of life. However, pain characteristics explained a significant and considerable amount in almost each quality of life domain, ranging from 13%

(Satisfaction with Health) to 29% (functional Status). After controlling for the effects of age, gender, and pain characteristics, the block of the psychosocial factors explained an additional proportion of the variance in all quality of life domains (ranging from 5% for Functional Status to 30% for Satisfaction with Life). Finally, a small but significant additional proportion of the variance (4%) was explained by interactions between pain intensity and coping for the domain Psychological Functioning.

Considering our first research question 'which psychosocial factors have a unique contribution to quality of life of adolescents with chronic pain?', inspection of regression coefficients indicated that vulnerability is the strongest significant and independent psychosocial factor associated with quality of life (Psychological Functioning, Physical Functioning, Social Functioning and Satisfaction with Life). The importance of the vulnerability of the adolescent is in accordance with other findings[23,24,36]. Beside vulnerability we also found that more rewarding for pain behavior by others is related to a higher quality of life (Psychological Functioning, Social Functioning and Satisfaction with Life). This finding supports studies that emphasized the importance of studying the attitudes and perceptions parents have with regard to themselves, their child and the stressor.[22,25] In addition, we found that coping was associated with quality of life in adolescents with chronic pain. The fact that maladaptive coping is associated with lower Psychological Functioning and Satisfaction with Health supports earlier studies on chronic pain.[37-39]

Regarding the second research question 'Which psychosocial factors moderate the relation between pain and quality of life?', we found that the relationship between pain and quality of life (psychological functioning) is strengthened by the level of emotion-focused avoidance coping. Consistent with Degotardi and colleagues[24], our results indicate that for adolescents who report more emotion-focused avoidance coping, a higher level of pain is associated with lower level of psychological functioning.

As for our third research question 'Is there a difference in the influence of psychosocial factors in adolescents with high and low pain intensity?', the subgroup analysis provided additional indication that the presented model was particularly suitable for adolescents with high pain. Associations between psychosocial factors and Psychological Functioning and Physiological Functioning were considerably stronger for adolescents with high pain compared to those with low pain intensity.

The present study has a number of methodological strengths. In particular, the regression analyses controlled for the potentially confounding effects of demographics and pain characteristics as well as for psychosocial factors in adolescents with chronic pain (irrespective of pain location). Another strength is the measurement of pain intensity which was carried out with a questionnaire and with a diary. Previous studies have shown that the use of a pain diary enhances both validity and reliability of pain parameters.[16,19,20,40,41] In accordance with other findings, we found lower prospective pain scores, compared with retrospectively measured pain scores.[16,42,43] An explanation for this finding might be memory recall bias; i.e. peaks of intense pain are more likely to be remembered than the moments of less intense pain.

It should also be noted that, in line with Schipper et al.[44] and Langeveld et al.[19], the quality of life measure used is based on the idea that quality of life is a multidimensional concept, incorporating social, psychological and physical health. At the same time, this multidimensional concept contributed to a methodological limitation of the study. Use of six independent regression analyses makes it impossible to control the possible correlations between the domains of quality of life. Further, all measures were self-report and the study design was cross-sectional. Therefore, cause-effect relations between pain, psychosocial factors and quality of life cannot be determined. There is a need for longitudinal studies with repeated measurements of pain, psychosocial factors and quality of life so that longitudinal relationships among these variables can be studied. Another limitation concerns the response rate in our study. Of the 631 eligible adolescents in the general population 194 adolescents participated in our study. Retrospectively measured, the pain frequency and intensity of the participating 194 adolescents was significantly higher than those of all adolescents with chronic pain from the general population (n=631), which may reflect a bias. However, we believe the bias to be small, considering the rather small differences in pain intensity and reported pain location between the prevalence sample of Perquin and colleagues[1] and our sample.

Our results show that factors other than demographics and pain characteristics alone are reflected in quality of life. Beside the main effects of vulnerability, reinforcement and modeling, coping strategies strengthen the relationship between pain and quality of life of adolescents with chronic pain with unknown organic etiology. Considering this, we suggest that psychological intervention programs directed at reducing the impact of pain on quality of life in adolescents with chronic pain should primarily focus on techniques aiming at stress-coping skills of the adolescents, in order to enhance their efficacy.

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Chapter 6

A cognitive-behavioral program for adolescents with chronic pain - a pilot study

Patient Education and Counseling, in press

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Abstract

The purpose of this pilot study is to evaluate the feasibility of a cognitive-behavioral training program for adolescents with chronic pain irrespective of pain localisation. A secondary aim was to give an impression of the effect of the program on pain and quality of life. Eight adolescents (14-18 years) with chronic pain and their parents recruited from the general population participated in this pilot study. The intervention included five group meetings alternated with four telephone contacts (during the self-management weeks) over a period of nine weeks. The training aimed to change pain behavior through pain education, relaxation strategies, problem-solving techniques, assertiveness training, cognitive restructuring and by stimulating the adolescent's physical activity level. The training further addresses the social context of pain by inviting parents to attend two meetings for the parents only, and by asking the adolescents to bring a peer to one of the meetings. Adolescents and their parents were positive about the program. Adolescents felt they were more in control of their pain and parents valued the support they experienced in helping their children to master the pain. The training was considered to be feasible in daily life. Further, the preliminary data showed an effect on pain and quality of life in the expected direction. The results underline the need for a definitive study with a larger sample size and a random clinical controlled design.

Introduction

Chronic pain is often associated with complex social and psychological problems leading to considerable medical consumption,[1] school absenteeism and nuisance in the adolescents' life[2]. As a consequence of the pain adolescents with chronic pain evaluate their quality of life as being less satisfactory than their healthy peers.[3] There is a growing interest in improving the quality of life of pain patients, but pain relief remains the main goal of treatment.[4] A recent systematic review showed that psychological treatments are effective in reducing the severity and frequency of chronic headache in children and adolescents.[5] However, few psychological treatments for adolescents with chronic pain other than headache have been evaluated in a randomized controlled study design. Significant reductions in pain and improvements in functioning were achieved in children and adolescents with recurrent abdominal pain[6] and complex regional pain syndromes[7]. Although psychological treatments based on the principle of cognitive-behavioral therapy are effective for adolescents with chronic pain, until now they have been limited to a single pain complaint. A program suitable for a wider spectrum of chronic pain sufferers might enhance its utility.

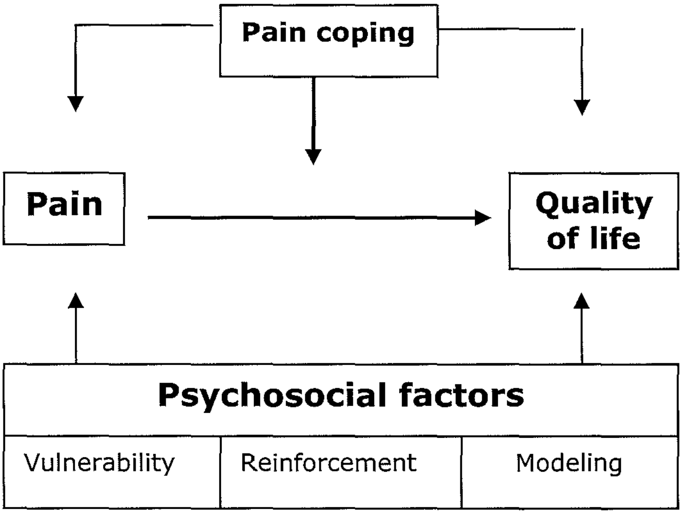
To facilitate the applicability of clinically-based treatments, several psychological interventions have a self-administered format.[8] In the field of chronic pain in children and adolescents, self-management programs have been shown to be as effective as psychological treatment guided by a therapist, but less expensive.[9-11] On the other hand, a therapy group has been reported to be more appealing to adolescents.[12] Groups give adolescents the opportunity for modeling, problem solving, helping others and relating to peers who share similar circumstances, all of which are more difficult to arrange through individual therapy or self-administered programs.[13,14] A combination of both forms, i.e. self-management alternated with group sessions, enables to benefit from the positive aspects of both.

Most psychological interventions involve the chronic pain sufferer only, whereas the importance of the social context of chronic pain is increasingly emphasized.[15] Although only a few treatments involve the parents of children and adolescents with chronic pain, the results are promising. For example, parents rated treatments in which they were involved as more satisfying and effective.[6,16] These experiences were supported by clinical improvements in pain severity and frequency.[6,17] Maternal caregiving strategies have been shown to be significant and independent predictors of clinical improvement in pain behavior.[17] Parent-mediated guidelines for pain behavior management may therefore be considered as an important addition to management programs for chronic pain in children and adolescents. With the growing influence of peers in adolescence, the social network is no longer limited to the adolescent's family; this may implicate the need for the involvement of peers in psychological interventions.

We have developed a cognitive-behavioral program for adolescents with chronic pain at different localisations based on a model of the quality of life of adolescents with non-specific chronic pain. The model is shown in Figure 1. In this model factors playing a role in chronic pain and in the quality of life of adolescents with chronic pain were tested with regression analysis.[3,18] Psychosocial factors and pain coping strategies are related to chronic pain and its parameters (chapter 4).[3] Besides the pain psychosocial factors and pain coping strategies have a significant impact on the quality of life of adolescents with chronic pain (chapter 5).[18] Besides, pain coping strategies moderate the relationship between pain and quality of life (chapter 5). [18]

The present pilot study evaluates the feasibility of this program. A secondary aim was to give an impression on its potential beneficial effect on pain and quality of life.

Figure 1: Model on pain and quality of life in adolescents with chronic pain



Methods

Subjects

Adolescents with recurrent or continuous chronic pain (with unknown organic etiology) that persisted for at least three months once a week with an intensity of 30 mm or more (retrospectively measured with a VAS) and resulted in pain-associated disability, were selected from a previously identified population of adolescents with chronic pain.[3]

Procedure

The Medical Ethics Committee of the Erasmus Academic Medical Center Rotterdam approved the study. Adolescents and their parents were telephoned and invited to participate in a pilot study of our cognitive-behavioral program. We explained that the program was developed based on the results of our earlier study in which they had participated,[3] and that the main goals of the pilot study were to evaluate the feasibility of the program and to explore its effects.

The outcome variables pain and quality of life were assessed two weeks prior to the initiation of the training at pre-treatment (T0), directly after the 9 weeks of intervention at post-treatment (T1), and one year after the intervention at follow-up (T2). Adolescents and their parents anonymously completed the evaluation form at post-treatment (T1).

Cognitive-behavioral program

The intervention consisted of a combination of five group meetings and four telephone contacts, during a period of nine weeks. The meetings were held at the Erasmus MC from 5.00 p.m. until 6.30 p.m. The group meetings were alternated with self-management weeks (telephone contacts) and supported with a training book. Parents were invited to attend two meetings for parents only; one at the beginning and one at the end of the intervention period. In session five the adolescents were asked to bring a peer to the meeting.

Rationale of the program

To develop active coping skills we emphasized the important role that individuals play in changing their own behavior. The goal of self-regulation is to help the adolescent develop skills to control their thoughts, feelings, behavior, and physiological responses. The program emphasizes changes in pain behavior through education and training in relaxation strategies, problem-solving techniques, assertiveness training and cognitive restructuring. If the physical activity level is decreased because of the pain, adolescents choose an individual goal (gradually rebuilding a physical activity) based on pain-specific disability.

Program content: adolescents

Each week of the program addresses a specific theme. Theoretical aspects of these themes are introduced in the meetings and further developed during the self-management weeks. The written material provides additional information on the themes. Thereafter, each theme is followed by new exercises and homework assignments related to the theory already discussed. Table 1 presents an outline of the training program.

Program content: parents

In the first meeting the rationale for pain management procedures and pain coping are explained and the content of the program is presented. Parents are asked to apply the pain model of Loeser[19] to the pain of their child. This is used as a starting point to discuss the impact of having a child with chronic pain in the family. General guidelines (based on Allen and Shriver [17]) to deal with pain (behavior) are offered to the parents. For instance, the parents are advised to take the pain of their child seriously and reward healthy behavior. In the second parents' meeting (in the ninth week) perceived changes in the child's pain behavior during the training are discussed. Attention is given to the way parents deal with the pain of their child. The influence of parental behavior on pain is elucidated and discussed. The parents' own pain complaints and the way they cope with them are also discussed and compared with the coping strategies of their children. The second parents' meeting also contained an evaluation of the program.

Table 1: Content of the cognitive-behavioral program, described for every week.

	Adolescents	*Parents*
Week 1	**Meeting** Introduction and rationale for pain management procedures and pain coping Explanation on how pain works Relaxation abdominal respiration Physiology of mind-body connections	
Week 2	**Self-management –telephone** Relaxation; adominal respiration Pain and stress RET: cognitive restructuring	**Meeting** Introduction and rationale for pain management procedures The content of the program The parents´ role
Week 3	**Meeting** Physical activity level Watch over your own border Progressive relaxation Recognizing and replacing negative pain thoughts with positive and rational ones	
Week 4	**Self-management –telephone** Living with pain: making plans Attention and distraction Progressive relaxation and physical exercise	
Week 5	**Meeting (with peers)** How does pain work? Sharing your pain Pain is a milestone round your neck	
Week 6	**Self-management-telephone** Positive thinking Progressive relaxation Error of reasoning and rational thoughts	
Week 7	**Meeting** Assertiveness training Relaxation through guided fantasy	
Week 8	**Self-management-telephone** The environment; others in pain? Fear of failure	
Week 9	**Meeting** Relapse prevention training Relaxation through guided fantasy Evaluation	**Meeting** How to proceed in the future Evaluation

Questionnaires

Feasibility

To evaluate the feasibility of the program an evaluation form was developed for the adolescents and for their parents. These forms were administered at post-treatment (T1) and consist of open-ended questions on how the adolescents and their parents experienced the training program: e.g. which aspects they valued most, what were the shortcomings, and how could the program be improved. The items on practical issues (e.g. concerning the timing and location of the meetings) give an indication about the applicability of the program in daily life.

Pain

The Pain Questionnaire[1] collected data at pre-treatment (T0), post-treatment (T1) and follow-up (T2) on the adolescent's date of birth, gender, nationality, educational level and school year, as well as data on the localisation, frequency, duration and intensity of the pain.

Pain diary

Adolescents were asked to register their pain intensity during two successive weeks at pre-treatment (two weeks prior to the intervention), post-treatment (directly after ending the intervention) and follow-up (six months after ending the intervention). Pain intensity was recorded three times daily, at breakfast, dinnertime and bedtime using a VAS with the anchors 'no pain' and 'the worst pain you can imagine'. The adolescents were asked to mark a position on the VAS that best matched their pain at that moment. The VAS is a valid measure for the assessment of self-reported pain intensity in chronic pain patients.[20]

Quality of life

Because we were mainly interested in whether the training reduced the impact of pain on daily life and leisure activities, we measured the quality of life in terms of Functional Status as addressed on the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP).[21] This is a shortened version of the Quality of Life Headache–Youth (QLH-Y) from Langeveld and colleagues.[22] The QLA-CP is reported to be reliable, valid and has suitable internal consistency and construct validity against COOP/WONCA charts.[21] In the present study we administered the Functional Status at pre-treatment (T0), post-treatment (T1) and follow-up (T2) with a higher score presenting a better quality of life.

Data analysis

The feasibility of the program was tested by categorizing relevant items of the evaluation of both adolescents and their parents. Pain intensity scores were calculated by summing all pain intensity scores (from 0 to 100) in the pain diary and dividing them by the number of recording times (2 weeks * 3 times daily =

42). Scores of adolescents who completed only one week of the pain diary were multiplied by two; this was the case for one of the adolescents. A decrease in pain of more than 50% was considered to be a large reduction, and a decrease from 20 to 50% a moderate reduction. Mean scores were calculated for the outcome variable quality of life in terms of Functional Status. All adolescents who completed the questionnaires on at least two of the three data collection points were included in the analysis.

Results

Subjects

Of the 86 eligible adolescents, we randomly selected 8 adolescents. If a selected adolescent refused participation we randomly selected another adolescent. Finally, 21 adolescents with chronic pain had been randomly selected to obtain a pilot sample of eight participants.

Table 2: Sample characteristics

Case	Age	Education level†	Pain	Frequency	Pain history§	Intensity*
1	14	Middle secondary school	Limb pain	At least 2x a week	30	38
2	14	Lower vocational training	Limb pain	At least 2x a week	8	32
3	16	Middle secondary school	Headache	Every day	40	33
4	16	Higher secondary school	Abdominal pain	Every day	96	66
5	16	Middle secondary school	Headache	At least 2x a week	84	55
6	16	Lower vocational training	Abdominal pain	At least 2x a week	3	66
7	17	Middle secondary school	Headache	At least 2x a week	-	-
8	18	Higher secondary school	Limb pain	Once a week	36	77

† Education levels were categorised into four groups: 1. lower vocational training; 2. lower secondary school, which is a four-year program; 3. middle secondary school, which is a five-year program and allows students to attend professional training; 4. higher secondary school, a six-year program and the prerequisite for admittance to university. §Pain history presented in months *Retrospectively measured with a visual analogue scale; scores ranged from 00 (no pain) to 100 (worst pain you can imagine).

Of these 21 adolescents (17 girls and four boys), 12 adolescents decided not to participate because their pain was no longer severe enough or because the pain had already disappeared. One of the adolescents requested to participate in a future group due to lack of time during the current study period. Besides the differences in gender (all selected boys decided not to participate), no differences were found in pain parameters between the 12 non-participants and the eight participants.

Table 2 gives the characteristics of the eight adolescents participating in the program. During the first meeting it became clear that three girls knew each other from the same school.

Feasibility

Participation

Of the eight adolescent girls, one missed a meeting due to illness and another due to school examinations; the remaining six girls attended all meetings. The participants were cooperative, showed respect for each other, and over time became a cohesive and trustworthy group, allowing frankness during the discussions about their pain and daily lives. The variation in age and education level caused some delays due to the need to clarify some theoretical aspects of the program. During the telephone contacts in the self-management weeks, we noticed that some adolescents had not read the information and/or completed the assignments, and some were difficult to reach at the arranged contact times. Four adolescents eported a reduction in their physical activity level because of their pain. These adolescents did not participate in any type of sport anymore. They were assisted in rebuilding their physical activit level. All four adolescents were active in sports again at the end of the program. Two of them started swimming, while the other two joint a fitnes centre.

Two adolescents did not bring a peer to the meeting in week 5; one because the peer could not meet the time schedule and another because she wanted to avoid telling her participation in the training.

Response rate of the evaluation study

Of the 8 participants, 7 girls completed the questionnaires and 6 the pain diary at pre-treatment (T0); 5 girls completed the questionnaires and 4 the pain diary at post-treatment (T1); and 5 girls completed both the questionnaires and the pain diary at follow-up (T2). Several contacts (by mail and telephone) were needed to gather the data. Of the 8 participants, 7 girls and the parents of five girls completed the evaluation form. These 6 parents (both parents of one girl were present) attended both of the parents meetings. The parents of one girl were present in the first, but not in the last meeting. One mother was unable to come because of young children at home, and another mother did not want to be involved in the intervention. Her daughter did not complete the questionnaires, diary or evaluation form, and both during and after the intervention the girl gave several reasons for her lack of compliance.

Evaluation by the adolescents

Most reactions about the program were positive: the adolescents valued being in a group with others. They reported that the program taught them how to influence their own pain and most experienced a change in their attitude towards pain and their life in general. Overall, they reported to practice the exercises about four days a week for 10 to 15 minutes a day. They found the assignments useful, even though they sometimes forgot to do them or lacked time to do them thoroughly. The theoretical aspects of pain and self-management coping were reported to be easy to understand. All adolescents said they would recommend the training to others with pain.

The adolescents valued the participation of their parents and a peer. One girl was originally reluctant to bring a peer to the program (fearing their reaction), but afterwards reported that they both had enjoyed it. Suggestions to improve the training included more practice during the meetings (e.g. relaxation) and preferably groups with boys and girls.

Evaluation by the parents

As expected (because of their age), the girls did not tell their parents much about the program. Consequently, all parents found that the information given during the parents' meetings was useful and helped them to support their child in the training. All parents said that their child valued the training; one parent thought that the training was sometimes difficult because of the additional assignments and registrations. The girls did not ask their parents for help at any time during the training, and most parents were unaware of when their child was practicing at home. Parents valued the contact with other parents and found their exchange on how to deal with pain in the family as instructive and supportive. Additionally, the guidelines were considered useful in practice. Most parents changed their attitude towards their child when in pain and felt that they were more consistent in their behavior.

Suggestions to improve the training were mainly practical, e.g. to start the adolescents' meetings at a later time, and to increase the number of participating parents to increase the exchange of experiences. One parent recommended more individual guidance of parents and/or children if required.

The trainer's perspective

The pilot study was conducted by a child psychologist and a graduate student in clinical psychology to allow a more individual approach when necessary (e.g. when additional explanation of the theory was needed). The allocation of tasks required both observation and guidance of the adolescents. The trainers exchanged

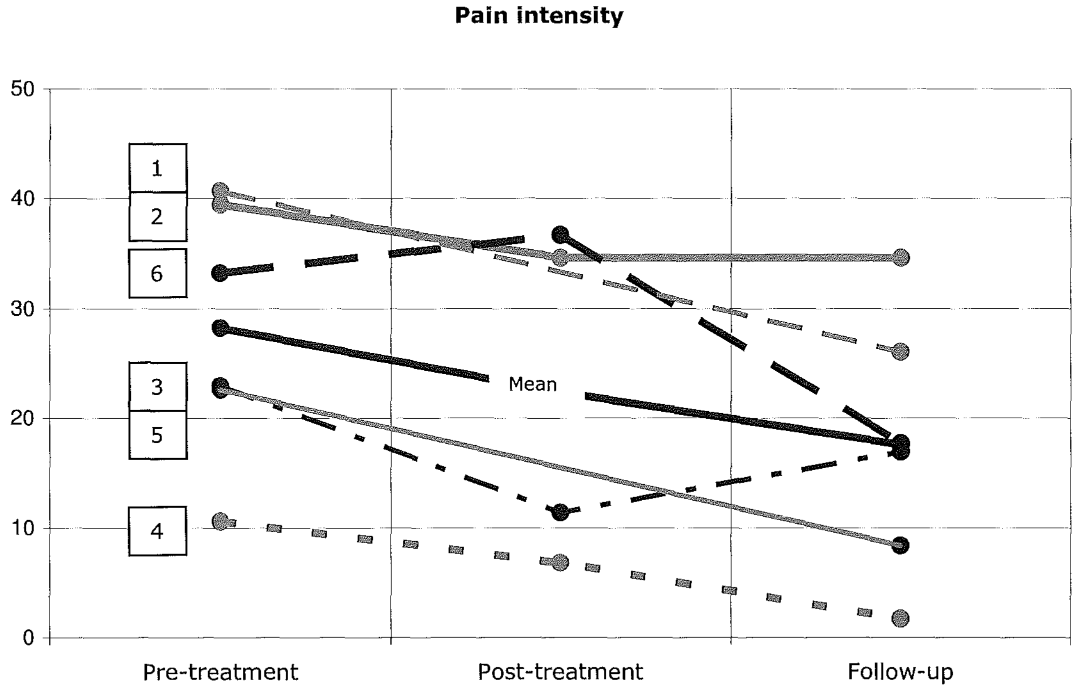
experiences after the meetings and possible changes in approach were discussed. Because the trainers complemented each other they considered their cooperation to be valuable and essential in getting a more complete picture of each participant.
The trainers reported that the first meeting had insufficient detailed information because most adolescents already knew how pain works (physical aspects). Not all adolescents completed their home assignments, and too much theory after school could be demotivating. In contrast, sufficient time to discuss their pain and its influence on their daily life seemed very important for the girls.
Preparations for the training, especially the preparatory talks in which individual tasks were allocated and materials were prepared for each meeting, were time consuming (about two hours for each meeting), whereas the preparations for the telephone contacts took only about 10 minutes for each adolescent and the telephone discussion itself lasted about 15 minutes per child. Other preparations included arranging a suitable room for the sessions (including a gymnasium or suitable field for the relay race in week 5), and some refreshments during the training.

Impressions on the effect of the program: pain and quality of life

Based on our model, pain and quality of life were chosen as variables to assess therapy outcome. Figure 2 shows the changes in pain intensity at post-treatment and follow-up compared to pre-treatment. At post-treatment (T1) two of the six girls showed no changes compared to pre-treatment whereas four girls achieved a moderate to large decrease in (prospective) pain intensity which was maintained during the follow-up period (T2). Three adolescents showed a clinically significant decrease (> 50%) in pain at T2 (figure 2).

Figure 3 shows the changes in quality of life. Overall, we found either no changes, small deteriorations or improvements in the Functional Status of the group. The improvements ranged from moderate at post-treatment (T1) to large at follow-up (T2) (figure 3).

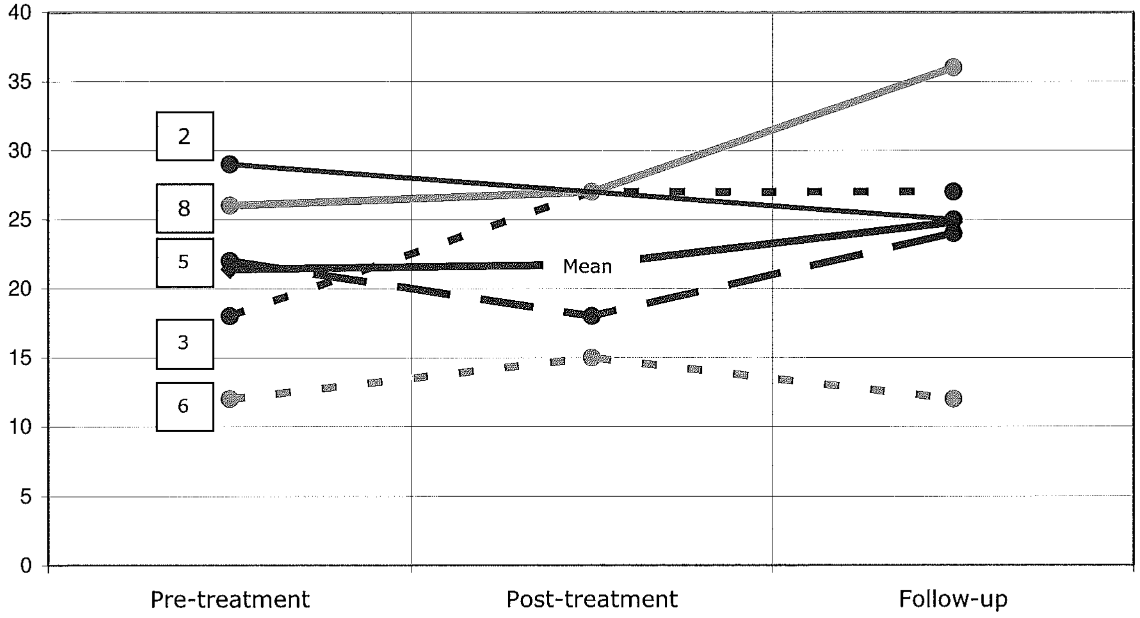
Figure 2: Prospective absolute pain intensity scores of the adolescents on pre-treatment, post-treatment and follow-up. Change in percentage (pre-post and pre – follow-up) is shown at right from the figure.



Case	T1	T2
1	*20%*	36%
2	12%	*12%*
3	50%	26%
4	36%	84%
5	20%	63%
6	10%	51%

Note: Lines represent the pain intensity scores of the participants (case number) on pre-treatment, post-treatment and follow-up. The percentage change in pain intensity (pre – post; T1 and pre – follow- up; T2) is shown for each participant (case number) at the right.

Figure 3: Quality of life (functional status) scores of the 5 adolescents at pre-treatment, post-treatment and follow-up. Change in percentage (pre-post and pre – follow-up) is shown at right from the figure.



Case	T1	T2
2	10%	-14%
3	50%	56%
5	-18%	9%
6	25%	0%
8	4%	39%

Note: Lines represent the pain intensity scores of the participants (case number) on pre-treatment, post-treatment and follow-up. The percentage change in pain intensity (pre – post; T1 and pre – follow- up; T2) is shown for each participant (case number) at the right.

Discussion

The present study provide evidence for the feasibility of a cognitive-behavioral program for adolescents with chronic pain. Both adolescents and their parents were positive about the content of the program. The adolescents reported that the training helped them gaining more control over their pain, and to feel less like a victim of pain. Parents evaluated the training and their involvement as supportive and informative because this enabled them to support their child in mastering their pain.

The compliance level in attending the meetings indicates that it is feasible to start immediately after school and finish before oter activities (e.g. sport) begin. Although the compliance with and participation in the meetings was high, during the self-management weeks the assignments were not always completed. The adolescents might be better moivated if, in future, the assignments are started during the self-management weeks and then evaluated at the star of the following group meeting. In addition, emphasizing self-responsibility may increase the return of questionnaires and pain diary after the training. A non-optimal response rate could be due to a perceived reduction of the usefulness and self-interest of completing questionnaire after the training. It should be stressed, therefore, that the follow-ups are part of the training and enable to determine the course of pain.

In this pilot study the participants varied in age and educational level. Although they remained respectful towards each other, a more homogenous group is preferable and might enhance the sharing of experiences. The fact that three of the eight girls were from the same school had a positive effect on this group. However, trainers should be aware that this could also have some neagtive effects (e.g. gossip) and take appropriate action when necessary.

Regarding the second aim of this pilot study, the training showed an effect on pain and quality of life in the expected direction. Adolescents reported a lower level of pain intensity at post-treatment, which continued during follow-up. At the final data collection point three adolescents (50%) had achieved a large clinically significant reduction in pain, and the remainder showed a moderate change. Assuming all those lost to follow-up would still report pain as severe as on pre-treatment level, the adjusted proportion of adolescents that reached a reduction of at least 50% would be 38%. The reached reductions in pain after our program is in line with other studies on effectiveness of cognitive-behavioral treatment for children and adolescents with chronic pain.[6,11,16,17,23-25] However, the design of our pilot study does not allow us to conclude that the reductions in pain are only a result of our program. For example, Perquin and colleagues found that 51% of the children and adolescents with chronic pain at pre-treatment assessment show spontaneous remissions at one year follow-up.[26]

In line with other studies that showed that pain is negatively related to quality of life in adolescents with chronic pain[21,27], we found an improvement in quality of life (i.e. in functional status) as a consequence of the decrease in pain intensity. In contrast with Bandell-Hoekstra[25] we found that the impact of pain on daily and leisure activities (Functional Status) was reduced after training. Despite these successful results, no definitive conclusions can be drawn. The preliminary data from this pilot study emphasize the need for a definitive study with a larger sample size and a random clinical controlled design.

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Chapter 7

A cognitive-behavioral program for adolescents with chronic pain - a randomized controlled trial

Submitted

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Abstract

Chronic pain has a significant negative influence on daily functioning of adolescents. A cognitive-behavioral intervention was developed to improve the quality of life of adolescents with chronic pain.

This study presents the efficacy of the intervention in a randomized controlled trial design. Thirty-one adolescents with chronic pain were randomly assigned to the intervention group (n=18) and the control group (n=13). The adolescents completed questionnaires (on quality of life and psychosocial factors) and a pain diary (for 3 weeks) at pre-treatment, post-treatment and at follow-up (6 months after the program ended).

There were no significant differences between the intervention group and the control group in the improvement of quality of life and reduction of pain at both post-treatment and follow-up. A main effect for medication use indicated that adolescents in the intervention group display lower levels of medication use than adolescents in the control group. Overall, there were no clear significant intervention effects of our cognitive-behavioral program for the psychosocial factors. These findings suggest that the intervention is not effective in improving adolescents' quality of life. Explanations for this finding and future directions are discussed.

Introduction

Chronic pain in adolescence is a common experience[1,2] and because symptoms mostly disappear over time[3,4,5] pain might be considered as part of normal life. However, longitudinal studies showed that, despite the spontaneous remission of chronic pain in most children and adolescents, some suffer from persistent chronic pain. Of the children and adolescents who reported chronic pain at baseline in some prospective studies, about 50% still experience chronic pain at one-year follow-up[3,4] and a third at two-year follow-up[3]. In this group of children, pain characteristics and impact of pain remains stable over a period of three years.[6] This stability of symptoms and negative influence on quality of life emphasize the need for effective treatments. Enhancing coping strategies might prevent the chronicity of pain or improve the quality of life.

Several pain management programs are available for children and adolescents with persistent chronic pain. A systematic review on all randomized controlled trials for children and adolescents with chronic pain showed that the most studied psychological interventions were developed for headache. These appeared to be effective in reducing the pain of headache.[7] Recently, a systematic review performed by Weydert and colleagues[8] found evidence for the efficacy of behavioral interventions (e.g. cognitive-behavioral therapy, biofeedback) in children and adolescents with recurrent abdominal pain.

Although these programs are effective in reducing pain, they all focus on children with a single specific pain location. Beside the lack of programs suitable for a wider spectrum of children with chronic pain, existing programs focus on pain relief as the primary outcome for evaluating treatment efficacy. No studies are available that have a primary focus on enhancing children's functioning.[9,10] Turk and colleagues[11] suggested that clinical trials evaluating the efficacy of pain treatments should present a more complete reporting of outcomes representing multiple core domains. Beside pain relief, outcomes concerned with a patients' functioning (e.g. physical and emotional functioning) and their disposition (reasons for withdrawal) and satisfaction with treatment should also be assessed.[11]

The purpose of the present study was to evaluate the efficacy of the cognitive-behavioral intervention program which we developed for adolescents with non-specific chronic pain irrespective of pain location (see chapter 6). This program addresses elements of a model on quality of life of adolescents with chronic pain that was based on previous studies of our research group. These studies showed that psychosocial factors (like vulnerability, reinforcement, modeling and coping) have significant influence on pain characteristics (chapter 4) and quality of life of adolescents with chronic pain (chapter 5).

A pilot study showed that the intervention program is feasible to incorporate in daily life of adolescents and that its effect on pain and quality of life occurred in the expected direction (chapter 6). The aim of the present trial was to explore the effectiveness of the intervention in a randomized controlled design. In this study, quality of life was considered as the primary outcome and pain relief, medication use and school absence were secondary outcome measures. Additionally, we were interested whether changes in the outcome measures are associated with changes in psychosocial factors (such as vulnerability, reinforcement and coping), because the intervention is directed at the psychosocial factors of our model on quality of life in adolescents (see chapter 1).

We studied the following research questions:

1. Is there a difference in change of quality of life (in terms of satisfaction with life, satisfaction with health, psychological functioning, physical functioning, functional status and social functioning) at post-treatment or at follow-up between the intervention group and a control group?
2. Is there a difference in change of pain, pain related problems, medication use and school absence post-treatment or at follow-up between the intervention group and a control group?
3. Is there a difference in change of psychosocial variables (vulnerability, reinforcement and coping) at post-treatment or at follow-up between the intervention group and a control group?

Methods

Participants

Adolescents aged 12 through 18 years with chronic pain were recruited (March 1, 2002 - May 31, 2003) from the general population and clinical population. The population sample was recruited via articles in (local) newspapers, ads in monthly magazines and distribution of folders with information in 3 secondary schools in Rotterdam. A clinical sample was obtained (March 31, 2003 – May 31, 2003) by informing the pediatricians of 8 hospitals in Rotterdam area, 5 family physicians and 3 pharmacies' of our study. They were requested to recruit patients who met the inclusion criteria with information by means of a folder. Inclusion criteria were: age between 12-18 years, recurrent (i.e. pain with pain-free intervals) or continuous chronic pain (with unknown organic etiology) that existed for at least three months, mean pain intensity in the previous month of at least 30 mm on a Visual Analogue Scale (from 0-100mm), impaired quality of life due to pain and sufficient knowledge of the Dutch language.

Of the 80 adolescents who reacted and requested additional information on the program (including a baseline measure), 44 (55%) were from secondary schools, 35 (44%) responded on the articles and ads and 1 (1%) enrollment was obtained from a hospital in Rotterdam-area. Thirty-nine adolescents eventually met the inclusion criteria, returned an informed consent and completed the screening measure (baseline assessment). Before randomization, one of them withdrew from the study due to hospitalization because of pain, leaving a sample of 38 adolescents that were included in the randomization procedure (table 1). After randomization, 2 adolescents from the intervention group and 1 adolescent from the control group withdrew. Reasons for withdrawal were: lack of time (n=1), difficulties in traveling – Erasmus University Medical Center Rotterdam was to far from home (n=1) and spontaneous improvement (n=1). Four adolescents from the control group were excluded from analysis because of lack of pre-treatment data (n=2) and lost to track down (n=2). Thus, the initial sample consisted of 31 adolescents of 12 to 18 years old. During the project, 2 adolescents withdrew from the study at post-test, and another two at follow-up. Both withdrawals at post-test were from the control group, at the follow-up one was from the intervention group and the other from the control group. Reasons for withdrawal were: unwillingness to be confronted with the pain (n=2), spontaneous improvement (n=1) and admittance in an in-patient psychiatric hospital (n=1). In order to lose as little information as possible, the drop-outs from the follow-up were not excluded from analyses if data from the post-test was available.

Procedure

The protocol of this study was approved by the Medical Ethics Committee of the Erasmus University MC Rotterdam. The study was designed as a randomized controlled clinical trial, involving an experimental group and a control group with a pre-treatment, post-treatment and 6 months follow-up.

After receiving a written informed consent, adolescents were assigned by a project team member, unaware of the identity of the participants, to the experimental or control group based on a randomly generated list. The experimental group followed the cognitive-behavioral intervention, while the control group did not receive any intervention, although they were free to consult a physician or undergo treatments.

Multidimensional assessments were made at baseline, pre-treatment, post-treatment and follow-up. The baseline assessment included the screening assessment before randomization (demographics, pain characteristics, pain medication use, school absence due to pain, satisfaction with life and satisfaction with health). Before the commencement of the intervention phase for the intervention group, adolescents from both groups completed three weeks of pain diaries and the questionnaires (pre-treatment). Following the last intervention session, again all adolescents from the intervention and control group self-monitored their pain for three weeks and were asked to complete the questionnaires (post-treatment). Six months after the conclusion of the intervention program, the follow-up assessment took place, including the same measurements as at the pre-treatment and post-treatment assessments.

Intervention

The intervention in this study was a cognitive-behavioral program for adolescents with chronic pain, irrespective of pain localization (for a complete description see chapter 6). The program consisted of five group meetings of 1½ hours over 9 weeks. The meetings were alternated with four self-management weeks supported by telephone contacts. Parents were invited to attend two additional meetings dedicated to the parents only. In session five the adolescents were asked to bring a peer to the meeting.

The program was outlined in a treatment protocol and emphasized changes in pain behavior through education and training in relaxation techniques (breathing exercises, progressive relaxation, mental imagery), problems-solving techniques, assertiveness training and cognitive restructuring (monitoring and generating positive self-statements during pain to replace negative self-statements about pain). As the program proceeded, adolescents received written information about all aspects of the program which built into a manual. They further received an exercise book for their homework assignments.

Measures

Self-report measures were included to evaluate the efficacy of the cognitive-behavioral intervention. Quality of life, pain characteristics, medication use and school absence were chosen as outcome measures. Additionally, we were interested whether changes in the outcome measures are associated with changes in psychosocial factors (such as vulnerability, reinforcement and coping).

Questionnaires at baseline:

Pain-retrospectively The pain questionnaire collected demographic data on the adolescent's date of birth, gender, nationality, educational level and school year.[2] Information about the pain was requested concerning location, frequency, duration, intensity, pain medication use and school absence. From a list of possible locations (head, abdomen, back, limb, neck, ear, throat, chest and elsewhere) subjects were asked to indicate all locations where they had experienced chronic pain for at least three months. In case of more than one location, respondents were asked to answer questions about the pain location they suffered the most. Pre-coded categories were used to assess the frequency of occurrence and the duration of a pain episode. The intensity of pain was assessed with a Visual Analogue Scale (VAS; a 100-mm long horizontal line with the anchors 'no pain' and 'the worst pain you can imagine') by asking "How bad is the pain usually?"

Quality of life To measure the satisfaction with life and satisfaction with health, we used two domains of the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP; see chapter 2). These domains, 'Satisfaction with Life in General' and 'Satisfaction with Health', were measured with a VAS (a 100 mm long horizontal line with the anchors 'completely dissatisfied' and 'completely satisfied'). A higher score represents more satisfaction.

Questionnaires at pre-treatment, post-treatment and follow-up

Quality of life To measure the quality of life, we used the Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP). This is a shortened version of the Quality of Life Headache – Youth (QLH-Y) from Langeveld and colleagues[12]. The QLA-CP is subdivided into six domains, each measuring a particular aspect of quality of life: (1) Psychological Functioning, (2) Functional Status, (3) Physical Functioning, (4) Social Functioning, (5) Satisfaction with Life in General, and (6) Satisfaction with Health. The assessed items were assigned to 1 of 4 response categories, ranging from 0 to 3 (0 = rarely or never; 1 = sometimes; 2 = often; and 3 = very often; or 0 = not at all; 1 = quite a bit; and 3 = very much). In addition, the QLA-CP included two VASs to measure Satisfaction with Life and Satisfaction with Health. The anchor points for both items ranged from 0 (completely dissatisfied) to 100 (completely satisfied). The reference period for answering the QLA-CP was the previous week. Mean items scores were calculated per domain. The higher the scores, the better the self-reported quality of life. The QLA-CP has shown suitable internal consistency and construct validity against COOP/WONCA charts (chapter 2).

Vulnerability Vulnerability was measured with the Inadequacy scale of the Dutch Personality Questionnaire - Junior.[13] The items on this scale address vague physical complaints, depressed mood, vague fears and feelings of insufficiency. Adolescents with a high score are more likely to describe themselves as tense and fearful and report more feelings of insufficiency; they are frequently characterized as having difficulty in making contact with others. The internal consistency of the Inadequacy scale is $\alpha = 0.87$.[13] As to validity, the scale has a high correlation with other questionnaires which measure the same construct.[14]

Reinforcement We literally translated the Illness Behavior Encouragement Scale (IBES) to measure the extent to which pain behavior is rewarded.[15] There is a version for children and one for their parents. The children's version measures the rewarding of pain behavior by the parents as perceived by the child. Because of the importance of peers for adolescents we generated another version for use with peers. This latter version measures the child's perception of the rewarding behavior from peers when he/she is in pain. The rewarding behaviors covered by the items are, e.g., giving small gifts, spending more time than usual with the child, giving special privileges, pampering or spoiling, excusing the child from regular chores. A higher score indicates more rewarding of (pain) behavior. The internal consistency for the child-report versions proved to be high ($\alpha = 0.81$ for maternal rewarding and 0.83 for paternal rewarding). Test-retest reliability scores for the child-report versions yielded Pearson correlation coefficients of $r = 0.77$ and $r = 0.72$ for maternal and paternal rewarding of illness behavior, respectively.[15]

Coping Pain coping strategies of adolescents were assessed with the Pain Coping Questionnaire.[16] This questionnaire assesses three higher-order strategies: Approach (with the subscales: information seeking, problem solving, seeking social support); Problem-focused avoidance (subscales: positive self-statements, behavioral distraction, cognitive distraction); and Emotion-focused avoidance (subscales: externalizing, and internalizing / catastrophizing). Approach-coping strategies are directed toward the stressor while avoidance strategies are directed away from the stressor.

The adolescents were asked to indicate how often they used each of the 39 coping behaviors. A higher score indicates the likelihood to use the coping strategies in pain situations. The subscales and higher-order scales are internally consistent.[16]

Pain-related problems To measure the pain-related problems adolescents experience, we used the Pain-related Problem List (PPL; see chapter 3). The PPL is subdivided into four domains: problems related to (1) concentration, (2) mobility, (3) adaptability, and (4) mood. The assessed items were assigned to 1 of 4 response categories, ranging from 0 to 3 (0 = rarely or never; 1 = sometimes; 2 = often; and 3 = very often; or 0 = not at all; 1 = quite a bit; and 3 = very much). The reference period for answering the PPL was the previous week. Pain problems were calculated by summing all domain scores. The higher the scores, the more self-reported pain problems. The PPL has good internal consistency and adequate validity against PedMIDAS (chapter 3).

Pain diary (pre-treatment, post-treatment and follow-up) To assess the pain intensity and pain frequency adolescents were asked to register their pain for three successive weeks. Pain intensity was recorded three times daily, at breakfast, dinnertime and bedtime with a VAS with the anchors 'no pain' and 'the worst pain you can imagine'. The adolescents were asked to mark a position on the VAS that best matched their pain at that moment. The VAS is a valid measure for the assessment of self-reported pain intensity in chronic pain patients.[17] We further asked the adolescents to register daily in their pain diary if they used medication for their pain and if they missed school due to their pain.

Statistical analysis

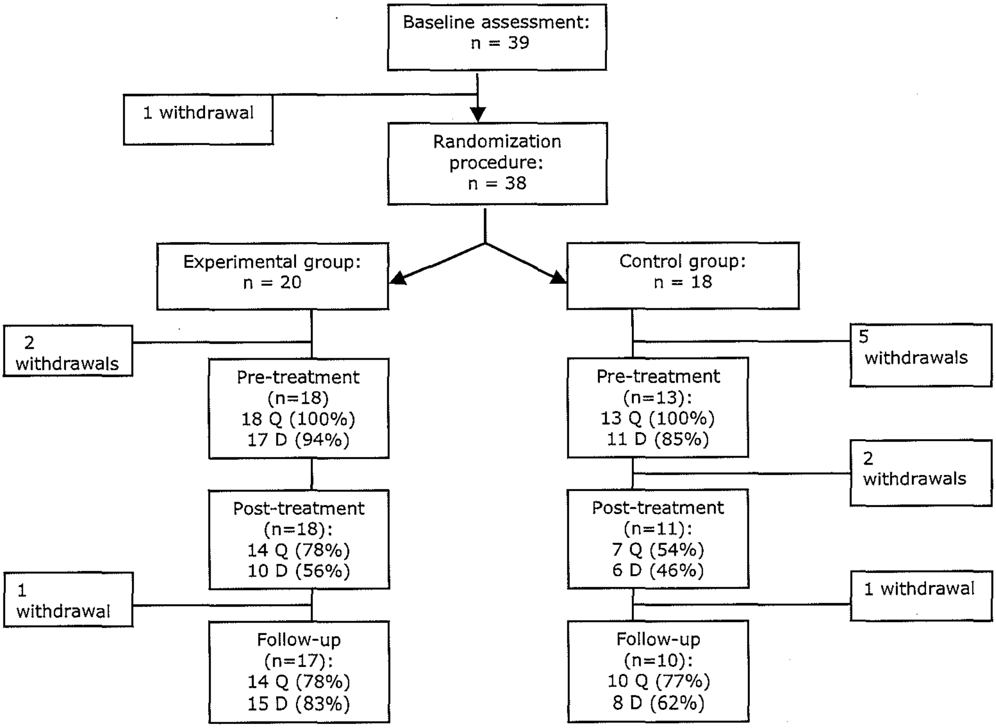
Prospective pain intensity scores consisted of the means of all pain intensity recordings. These scores were calculated by adding up all pain intensity scores (from 0 to 100) and dividing them by the number of recording times (3 weeks * 3 times daily = 63). Pain frequency was defined as the number of VAS recordings indicating pain in the 3-week pain diary (range: 0 - 63). Medication use and school absence were defined as the number of days medication was used for pain or school was missed due to pain in the 3-week pain diary. Scores ranged from 0 to 21 and 0 to 17 for medication use and school absence respectively.

T-tests were performed to analyse differences between the intervention group and the control group on outcome measures at baseline assessment. Chi-Square tests and Mann-Whitney *U* (M-W) tests were used to test differences for categorical variables. Subsequently, 2 series of ANCOVA's were executed. In the first series, intervention group and control group were compared on their scores at post-treatment assessment. In the second series, intervention group and control group were compared on their scores at the follow-up test. Scores at pre-treatment were included as a covariate. In every ANCOVA, the covariate was identical to the selected outcome measure. This was done to account for between-condition differences at pre-treatment and to reduce the error-variance.

Results

Of the 31 adolescents that entered the present trial, 29 (93%) were girls. The mean age was 15.2 years (sd = 2.0 yrs). Eighteen adolescents were randomly assigned to the intervention group and 13 adolescents to the control group. Sample size in both groups at each assessment moment is presented in table 1. Complete measurements at post-test and follow-up were obtained from 42% of the adolescents. Questionnaires were completed by 61% of the adolescents at both post-test and follow-up, while only 45% completed all diary assessments.

Table 1: Sample size in the intervention and control group at pre-treatment, post-treatment and 6 months follow-up with percentages compared to pre-treatment between brackets



NB. Q = questionnaires completed ; D = diaries completed

Baseline assessment

Both groups were comparable with regard to gender, age and baseline measures on overall quality of life, pain characteristics, medication use or school absence (see table 2). Adolescents from the intervention group reported more continuous pain complaints compared to the control group.

Table 2: Randomization check: baseline characteristics of study participants.

		Intervention group n=18	Control group n=13
Age *Years (range)*		14.9 (12-17)	15.6 (12-18)
Gender *N (%)*			
Boys		2 (11%)	0
Girls		16 (89%)	13 (100%)
Quality of life (VAS) *mm (sd)*			
Satisfaction with Life		57.7 (23.1)	57.7 (20.8)
Satisfaction with Health		48.6 (24.4)	40.5 (28.4)
Pain characteristics (retrospective)			
Intensity (VAS) *mm (sd)*		65.7 (17.0)	58.2 (15.9)
Frequency *N (%)*	weekly	16 (88.9%)	11 (84.7%)
	less than weekly	2 (11.1%)	2 (15.3%)
Duration* *N (%)*	≥ a whole day	10 (58.9%)	8 (61.9%)
	less than a day	7 (41.1%)	5 (38.1%)
Course *N (%)*	continuous	13 (72.2%)	5 (38.5%)
	recurrent	5 (27.8%)	8 (61.5%)
History *months (sd)*		32.8 (20.3)	28.9 (24.4)
Localisation *N (%)*	head	8 (44.4%)	5 (38.5%)
	abdomen	4 (22.2%)	2 (15.4%)
	back	1 (5.6%)	2 (15.4%)
	limb	2 (11.1%)	4 (30.8%)
	other	3 (16.7%)	0 (0%)
Days absent from school due to pain in the previous month *days (sd)*		4.8 (6.0)	4.1 (5.6)
Pain medication used in the previous month *N (%)*		15 (83%)	12 (92%)

* 1 case had incomplete data on the duration of a pain episode and was not included.

Post-treatment ANCOVAs with pre-treatment scores as covariate

Outcome variables: No significant differences in change in quality of life, pain characteristics or school absence were found between the intervention group and control group, but there were differences for medication use at post-test (see table 3 and 4). Adolescents from the intervention group used significantly less medication than adolescents from the control group. Further, the intervention group reported significantly more pain-related problems compared to the control group.

Psychosocial factors: Differences in change were found in coping strategies and pain-related problems between the intervention group and control group (see table 5). The intervention group tended to use more emotion-focused avoidance coping strategies at post-treatment than the control group. No significant differences in change were found for the other psychosocial variables.

Follow-up test ANCOVAs with pre-treatment scores as covariate

Outcome variables: The intervention group tended to report higher levels of Social Functioning at follow-up compared to the control group. There was a significant main effect for the subscale 'Functioning at home'. Adolescents in the intervention group displayed higher levels of functioning at home than adolescents in the control group. Differences were also found in the level of medication use between both groups at follow-up. At follow-up, adolescents in the intervention group used less medication than adolescents in the control group at follow-up. No training effects were found for the other variables (see table 3 and 4).

Psychosocial factors: There were no significant differences in change for the psychosocial factors between the intervention and control group (see table 5).

Table 3: Mean scores at pre-treatment, mean change scores on post-treatment and follow-up on quality of life, 95% confidence interval of the difference score between both groups (adjusted for pre-treatment values).

		Pre-treatment			Post-treatment minus pre-treatment			Follow-up (6 months) minus pre-treatment		
		mean (sd)	n	[95% CI]	mean Δ (sd)	n	mean difference* [95% CI]	mean Δ (sd)	n	mean difference* [95% CI]
Satisfaction Life[a]										
	E	57.7 (24)	18	[45.7 – 69.7]	9.2 (17.8)	9	8.0 [-11.8 - 27.8]	7.7 (25.1)	13	2.5 [-14.9 - 19.8]
	C	52.7 (23)	12	[38.1 – 67.3]	10.4 (27.3)	8		12.5 (21.4)	10	
Satisfaction Health[a]										
	E	43.6 (22)	18	[32.7 – 54.4]	14.6 (28.9)	13	- 1.1 [-20.5 - 18.3]	12.4 (33.1)	14	-14.1 [-34.1 - 5.9]
	C	42.2 (26)	13	[26.4 – 58.1]	19.0 (13.5)	8		- 3.9 (33.3)	10	
Psychological funct[b]										
	E	1.8 (0.4)	18	[1.6 – 2.0]	0.1 (0.4)	13	0.0 [-0.4 - 0.4]	0.1 (0.4)	14	0.0 [-0.3 - 0.3]
	C	1.8 (0.5)	13	[1.5 – 2.1]	0.0 (0.3)	8		-0.0 (0.4)	10	
Functional Status[b]										
	E	2.2 (0.5)	18	[1.9 – 2.4]	0.0 (0.9)	13	0.0 [-0.5 - 0.5]	0.2 (0.5)	14	- 0.1 [-0.5 - 0.3]
	C	1.9 (0.4)	13	[1.6 – 2.1]	0.3 (0.3)	8		0.2 (0.6)	10	
Physical funct[b]										
	E	1.7 (0.5)	18	[1.4 – 2.0]	0.1 (0.6)	13	0.3 [-0.1 - 0.8]	0.2 (0.5)	13	0.1 [-0.5 - 0.6]
	C	1.8 (0.4)	13	[1.6 – 2.1]	0.2 (0.6)	7		0.2 (0.8)	10	
Social functioning[b]										
	E	2.0 (0.5)	18	[1.7 – 2.2]	0.0 (0.5)	13	0.0 [-0.4 - 0.3]	0.1 (0.5)	14	- 0.4 [-0.8 - 0.0]
	C	1.7 (0.5)	13	[1.4 – 2.1]	0.0 (0.3)	8		-0.0 (0.5)	10	

* Mean difference in the change scores (Δ) between intervention and control group (adjusted for differences in pre-treatment values)

[a] The higher the score (range 0 – 100), the better the quality of life. [b] The higher the score (range 0 – 3), the better the quality of life.

Table 4: Mean scores at pre-treatment, mean change scores on post-treatment and follow-up on pain intensity, pain frequency, medication use and school absence (based on pain diary data), 95% confidence interval of the difference score between both groups (adjusted for pre-treatment values).

		Pre-treatment			Post-treatment minus pre-treatment			Follow-up (6 months) minus pre-treatment		
		mean (sd)	n	[95% CI]	mean Δ (sd)	n	mean difference* [95% CI]	mean Δ (sd)	n	mean difference* [95% CI]
Pain intensity[a]										
	E	38.7 (21.7)	17	[27.5 – 49.9]	- 5.2 (15.6)	9	2.1 [-13.6 - 17.9]	- 10.1 (11.5)	14	- 3.6 [-22.5 - 15.3]
	C	39.6 (20.3)	11	[25.9 – 53.2]	- 5.6 (10.4)	6		- 15.5 (29.1)	6	
Pain frequency[b]										
	E	51.1 (16.6)	17	[42.6 – 59.6]	- 7.2 (12.7)	9	4.9 [-9.5 - 19.3]	- 9.9 (11.0)	14	- 5.7 [-23.3 - 11.9]
	C	52.5 (13.5)	11	[43.4 – 61.5]	- 2.2 (10.1)	6		- 15.3 (26.1)	6	
Medication use[c]										
	E	4.2 (5.0)	17	[1.6 – 6.7]	- 3.0 (2.9)	9	3.4 [1.8 - 5.0]†	- 3.4 (5.2)	14	6.7 [1.5 - 12.0]†
	C	4.9 (5.6)	11	[1.2 – 8.7]	0.0 (2.5)	6		4.8 (10.1)	6	
School absence[d]										
	E	2.8 (4.6)	17	[0.5 – 5.2]	1.7 (6.4)	9	- 0.3 [-6.5 - 5.9]	- 1.0 (3.5)	14	1.5 [-1.4 - 4.4]
	C	2.2 (5.1)	11	[-1.2 – 5.6]	0.8 (2.4)	6		- 0.2 (2.3)	6	

* Mean difference in the change scores (Δ) between intervention and control group (adjusted for differences in pre-treatment values).

† the 95% CI does not include zero, which means that there is a statistically significant difference between both groups.

[a] Intensity of the pain was assessed using the Visual Analogue Scale, a 100mm long line with the verbal anchors (0) 'no pain' versus (100) 'the worst pain you can imagine, at both sides. [b] Frequency scores ranged from 0 (no pain episodes) to 63 (maximum number of pain episodes). [c] The scores on medication use ranges from 0 days of medication use to 21 days during three weeks of pain registration in the pain diary. [d] The school absence varied from 0 days to maximum 17 school days missed because of pain during the three weeks of registration.

Table 5: Mean scores at pre-treatment, mean change scores on post-treatment and follow-up on psychosocial factors (vulnerability, reinforcement, coping) and pain-related problems, 95% confidence interval of the difference score between both groups (adjusted for pre-treatment values).

		Pre-treatment			Post-treatment minus pre-treatment			Follow-up (6 months) minus pre-treatment		
		mean (sd)	n	[95% CI]	mean Δ (sd)	n	mean difference* [95% CI]	mean Δ (sd)	n	mean difference* [95% CI]
Vulnerability [a]	E	18.6 (6.7)	18	[15.3 – 21.9]	1.9 (7.0)	13	- 4.7 [-10.4 - 1.0]	- 2.2 (6.8)	13	- 0.6 [-6.6 - 5.3]
	C	20.2 (13.1)	13	[12.2 – 28.1]	- 2.5 (3.3)	8		- 3.1 (6.9)	10	
Reinforcement [b] parents	E	22.2 (7.1)	18	[18.7 – 25.7]	- 1.4 (5.2)	13	0.4 [-4.9 - 5.7]	- 1.8 (6.0)	14	- 0.4 [-6.9 - 6.0]
	C	18.2 (6.7)	13	[14.2 – 22.3]	- 0.5 (5.8)	8		0.1 (8.7)	10	
Reinforcement [c] peers	E	13.7 (3.3)	18	[12.1 – 15.4]	0.8 (3.5)	13	0.6 [-2.7 - 3.9]	0.1 (3.4)	14	0.3 [-2.4 - 2.9]
	C	13.2 (4.0)	13	[10.8 – 15.6]	0.3 (3.4)	6		- 0.2 (4.6)	9	
Prob emot appr [d]	E	2.4 (0.7)	18	[2.1 – 2.8]	- 0.2 (0.5)	13	0.2 [-2.3 – 0.6]	- 0.3 (0.7)	14	0.2 [-0.3 - 0.7]
	C	2.6 (0.6)	13	[2.2 – 2.9]	- 0.1 (0.3)	8		- 0.2 (0.3)	10	
Probl foc avoid [d]	E	2.6 (0.8)	18	[2.2 – 3.0]	0.3 (0.4)	13	- 0.3 [-0.8 – 0.2]	0.0 (0.8)	14	0.1 [-0.5 - 0.7]
	C	2.7 (0.8)	13	[2.3 – 3.2]	- 0.1 (0.6)	8		0.0 (0.4)	10	
Emot foc avoid [d]	E	2.0 (0.7)	18	[1.7 – 2.4]	0.0 (0.5)	13	- 0.4 [-0.7 – 0.0]†	- 0.2 (0.6)	14	- 0.2 [-0.7 - 0.3]
	C	2.4 (0.6)	13	[2.0 – 2.8]	- 0.5 (0.4)	8		- 0.5 (0.4)	10	
Pain-rel. probl [e]	E	12.5 (7.3)	18	[8.9 – 16.1]	1.3 (5.1)	12	- 6.9 [-13.4 - -0.5]†	- 2.0 (6.6)	14	- 1.9 [-8.9 - 5.1]
	C	19.0 (6.6)	13	[15.0 – 23.0]	- 6.5 (7.0)	8		- 5.4 (9.3)	10	

* Mean difference in the change scores (Δ) between intervention and control group (adjusted for differences in pre-treatment values)
† The 95% CI does not include zero, which means that there is a statistically significant difference between both groups
Scale range: [a] 0 – 56; [b] 0 – 48; [c] 0 – 24; [d] 0 – 3; [e] 0 – 54. The higher the scores, the higher the presence of psychosocial factors

Discussion

The present study did not show that the cognitive-behavioral intervention program results in higher quality of life improvement compared with a control group. Additionally, no significant differences in changes in pain intensity, pain frequency and school absence were found at post-treatment and follow-up between both groups. However, adolescents in the intervention group used significantly less medication than adolescents in the control group at both post-treatment and follow-up.

Before interpreting these results a few remarks concerning our sample have to be made. In the present study, 31 adolescents with chronic pain participated. They were recruited from the general population (school newspaper, magazines) and from clinical settings (family doctor's practice, pediatrics in hospitals and via announcements in pharmacies). Only 80 adolescents showed interest in participating in our study. Compared with the prevalence of severe chronic pain in Dutch adolescents this is an extremely low enrollment. Up to 33% of the Dutch adolescents report persistent or recurrent pain.[2] A smaller proportion, 16% of the adolescent girls and 6% of the boys, report severe disabling chronic pain indicating their eligibility for our intervention. Although the number of enrollments was disappointing, these findings support recruitment for participants in the general population. As a consequence of our small sample size the results should be interpreted with caution.

Considering our first research question, we found no significant differences in change in quality of life between the intervention group and the control group. The absence of a significant difference is in accordance with the study of Bandell-Hoekstra and colleagues.[18] Their self-management program for children and adolescents with headache did not show efficacy for improving quality of life when the data of the experimental group was compared those of a placebo group. Our results concerning quality of life might be explained by the fact that there were no clear treatment effects on pain characteristics and vulnerability. In a previous study we found that pain intensity and especially vulnerability have a strong influence on the quality of life of adolescents with chronic pain (chapter 5). However, we did find a significant difference in change in functioning at home between the intervention group and the control group. This effect might be a consequence of the participation of parents in our intervention. Adolescents in the intervention group reported that they got on better with their parents and reported that they could talk more to their parents about personal things or problems than adolescents in the control group. Adolescents and their parents might have learned other ways to communicate with each other which resulted in better functioning at home. Positive effects of the involvement of parents in interventions for children with chronic pain were also found in other studies.[9,19-21]

Regarding our second research question, we did not find differences in changes between the intervention group and the control group for pain characteristics at post-test and follow-up. These results are in contrast with the conclusions of (Cochrane) systematic reviews on the effectiveness of psychological treatments for chronic headache[7] and recurrent abdominal pain[8]. Most psychological treatments evaluated in controlled trials were effective in reducing pain characteristics. [19,21-26] Though, in accordance with our results the efficacy in pain reduction of a cognitive-behavioral intervention could not be established in recent studies on adolescents with chronic headache in the general population[18] and tertiary referred adolescents with chronic pain[9].

In the present study, the absence of differences in changes in pain reduction might indicate that adolescents in the intervention group learned other ways to cope with their pain instead of using pain medication.

The intervention group, surprisingly, displayed more pain-related problems compared to the control group after the intervention period. This difference might be caused by the mechanism that adolescents are confronted with their disabilities during intervention.

With regard to our third research question, significant differences in change were found in the psychosocial variable coping at post-treatment. Higher levels of change in emotion-focused avoidance coping strategies were found in the intervention group at post-treatment. The unexpected difference in both pain related disabilities as well as in coping might be caused by the mechanism that adolescents are, increased aware of their pain related disabilities and pain coping styles during the intervention. A study conducted by Howard and colleagues[27] showed that treatment subjects experience changes in their perceptions of their pretreatment levels of functioning (i.e. coping or pain-related problems), whereas non-treatment control subjects do not. Howard and Dailey[28] additionally report that this response-shift bias might cause the absence of treatment results established with questionnaire measures.

The absence of clear effects of our training program may be related to the lack of power for demonstrating small effects. Our sample size was rather small and the compliance to the post-treatment and follow-up measurement was not optimal. Future research may include electronic diaries with compliance-enhancing features because they are a more effective way of collecting diary information[29] and show, even in children, significantly greater compliance and accuracy compared to traditional paper diaries[30].

The absence of effects may also be explained by the fact that our program was aimed at a broad range of adolescents. Adolescents with pain intensity higher than 30 mm on a VAS were included in our study. We argued that these adolescents are at risk of adjustment problems and may therefore profit from a cognitive-behavioral pain program. As a consequence of the absence of specific clinical based criteria, the pain intensity at pre-treatment was relatively low in comparison with adolescents that were included in other clinical-based[9] or general population[18] studies. It will be more difficult to demonstrate an intervention effect, because the pain reduction is expected to be larger in adolescents with higher pre-treatment pain intensity scores.

Finally, there is a lack of information on the responsiveness of the questionnaires used in this study. We do not know whether the questionnaires are able to detect changes in quality of life or psychosocial factors due to treatment, and do not have any information on the size of detectable changes.

The relatively small number of enrolled cases raises some questions for the direction of future research. The first, most obvious question would be: Are adolescents already able to cope with their pain in an effective manner, which makes participation in a cognitive-behavioral program unnecessary? Hunfeld and colleagues[31] concluded - based on interviews with adolescent pain sufferers from the general population - that pain had become part of the daily life of adolescents. Adolescents structure their activities and sleeping hours to prevent aggravation of pain. This could mean that our program is best offered to adolescents who are referred to secondary care because they are in need for help in managing their pain. Smith and colleagues[32] show that adolescent who seek behavioral treatment for recurrent headache report headaches of longer duration, missed more school days due to headache, and report higher initial sustained discomfort scores. Future research on establishing effectiveness of our program should be aimed at obtaining a clinical sample. To enlarge the number of participants a multicentre randomized controlled would be required.

The second question concerns the design of our intervention. Is the design of our program interesting enough for adolescents nowadays? Considering the frequent use of computers by adolescents, an interactive website offering cognitive-behavioral treatment might be more appealing for them. Previous studies have shown that computer assisted cognitive behavioral therapy is effective for individuals with increased depressive symptoms at the community level[33] and in general practice[34]. Internet sites offer feasible and powerful public health interventions. This should be further addressed in future research.

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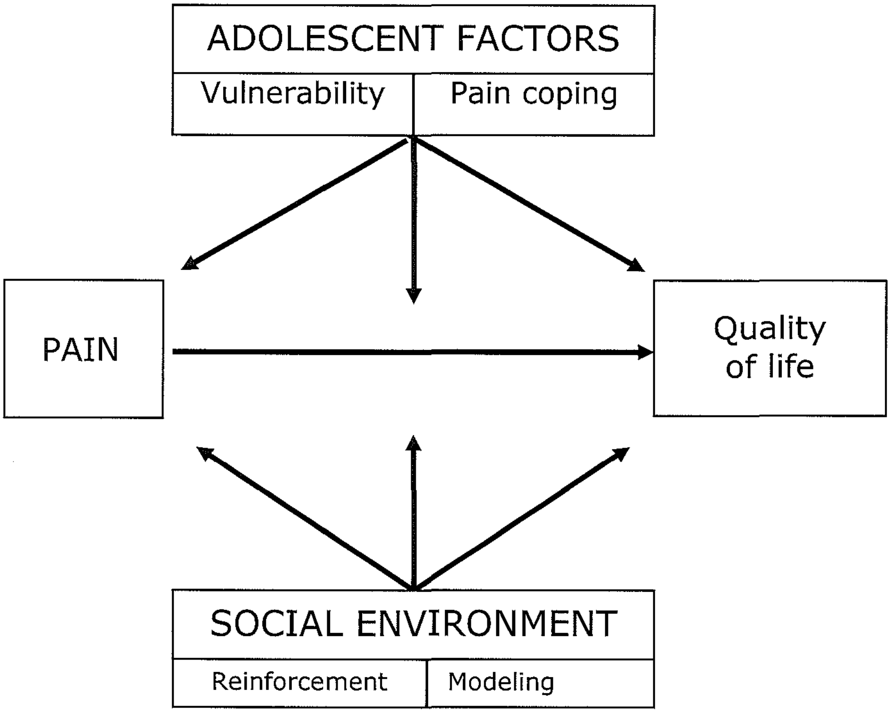
Chapter 8

General discussion

Overview of the results

The main objective of this thesis was to evaluate a model on pain and quality of life in adolescents with chronic pain (figure 1). This model was evaluated by means of two designs: a cross-sectional and a randomized controlled design.

Figure 1: Model on pain and quality of life in adolescents with chronic pain



To study the impact of chronic pain on the daily life of adolescents, we shortened a quality of life questionnaire for adolescents with chronic pain and developed a pain-related problem list. Based on the data, the Quality of Life Questionnaire for Adolescents with Chronic Pain (QLA-CP) and the Pain-related Problem List (PPL) were both considered reliable and valid instruments to measure quality of life and pain-specific problems, respectively, in adolescents with chronic pain irrespective of their pain location *(chapter 2 and 3)*.

In the cross-sectional studies chronic pain, quality of life and psychosocial factors were studied in adolescents with chronic pain and controls. Compared to controls, adolescents with chronic pain are more vulnerable, report more significant others with chronic pain and use more emotion-focused avoidance coping. Contrary to our expectations, the chronic pain group experienced less reinforcement for their pain behavior by parents and peers than adolescents without chronic pain *(chapter 4)*. Additionally, the model was evaluated by investigating the contribution of variables from our model to chronic pain and quality of life. Regression analyses on the contribution of psychosocial factors to chronic pain and its parameters sustained the positive relation between pain on the one hand and vulnerability, the number of significant others with pain (pain models), (less) pain reinforcement, and emotion-focused avoidance coping with pain on the other *(chapter 4)*. The results of our population based cross-sectional study further show that psychosocial factors account for a significant variance in the adolescent's quality of life, even when controlling for pain characteristics. In addition, we found that coping moderates the relation between pain intensity and quality of life *(chapter 5)*.

Based on these results we developed a cognitive-behavioral training program for adolescents with chronic pain irrespective of their pain location *(chapter 6)*. A pilot study showed that our cognitive-behavioral intervention is feasible and fits well in daily life of adolescents. The preliminary data showed an effect on quality of life and pain in the expected direction *(chapter 6)*. A randomized controlled trial design was used to explore the effect of the intervention on quality of life and chronic pain *(chapter 7)*. In this study, the model was evaluated by studying changes in pain and quality of life due to (expected) changes in the psychosocial factors as a consequence of the intervention program. No clear intervention effects could be demonstrated; a similar improvement in quality of life and decrease in pain intensity and frequency was found between the intervention and control group. However, there was a significant main effect for the subscale functioning at home at follow-up, indicating that adolescents in the intervention group display better levels of functioning at home than adolescents in the control group. Differences are also found in the level of medication use between both groups at post-treatment and follow-up. Adolescents from the intervention group display a significantly lower level of medication use than adolescents in the control group. Additionally, no clear significant differences in change in the psychosocial factors were found between the intervention and control group. At post-treatment differences were found in coping strategies and pain-related problems. Surprisingly, the adolescents from the intervention group display significantly more pain-related problems and more use of emotion-focused avoidance coping than those from the control group *(chapter 7)*.

In this chapter we discuss these major findings. The chapter will finish with concluding remarks on our model on pain and quality of life in adolescents with chronic pain and recommendations for future clinical and research directions.

The development of instruments for adolescents with chronic pain

Quality of Life questionnaire for Adolescents with Chronic Pain (QLA-CP)

Validating the QLA-CP we, unexpectedly, found a positive correlation between pain intensity and the domain social functioning (chapter 2), indicating the higher the pain the better the social functioning of the adolescents. Other studies have stressed the negative influence of chronic pain on social functioning of children and adolescents with chronic pain.[1,2] Our finding might indicate that the domain social functioning of the QLA-CP refers to the experienced social support rather than the actual functioning in social situations. Adolescents with intense pain refer more to the social support of significant others. This assumption is supported by the moderate correlation between social functioning of the QLA-CP and social support of the COOP/WONCA charts. An alternative explanation might be that the domain social functioning was found to be less reliable and valid than the scales representing the other domains of quality of life.[3]

Pain-related Problem List (PPL)

One of the strengths of the development of the PPL is the use of interviews with adolescents with chronic pain to construct the items. These items reflect the experiences of the adolescents themselves, rather than the opinion of professionals or theoretical assumptions. The questionnaire can be used in adolescents with chronic pain irrespective of their pain location. Other questionnaires mainly focus on the impact of a single pain location on daily functioning. A limitation of our study could be that we used the Chronic Pain Disability Inventory (CPDI) and the Pediatric Migraine Disability Assessment (PedMIDAS) to validate the PPL with. However, these questionnaires appeared to be the best instruments available, given their disease-specific focus and the fact that they are best attuned to our focus group.

We found some indication for the validity in a clinical sample, but this sample was too small to draw any conclusions. So far, the results can only be generalised to adolescents with chronic pain from the general population.

Further study is advised on the test-retest reliability, cross-cultural validation and the responsiveness of both questionnaires.

Evaluating the model:

the importance of psychosocial factors for pain and quality of life

Adolescent factors

The cross-sectional studies showed that of the psychosocial factors, vulnerability (in terms of neuroticism) and coping have the strongest association with pain *(chapter 4)* and quality of life *(chapter 5)*. We found significant differences on these psychosocial factors between adolescents with and those without chronic pain. Differences between both groups in vulnerability and emotion-focused avoidance coping strategies showed moderate effect sizes and should be considered as differences with potential clinical importance. The importance of vulnerability and coping was sustained by the analysis of the contribution of psychosocial factors to chronic pain *(chapter 4)* and quality of life *(chapter 5)*. The fact that neuroticism and emotion-focused avoidance coping are associated with lower quality of life supports earlier studies on chronic pain.[4-8] The results further show that the relationship between pain and quality of life (psychological functioning) is strengthened by the level of emotion-focused avoidance coping. Consistent with Degotardi and colleagues[6] our results indicate that for adolescents who report more emotion-focused avoidance coping, a higher level of pain is associated with a lower level of psychological functioning.

To explain the strong correlations between neuroticism (i.e. vulnerability) and chronic pain Watson and Pennebaker[9] formulated the symptom-perception model. According to this model, persons scoring high on neuroticism (i.e. vulnerability) are more likely to notice and attend to internal physical sensations and minor aches because their attentional scanning of both the external and internal environment is fraught with anxiety and uncertainty. Other studies concluded that neuroticism should be considered as a vulnerability factor that could lower the threshold at which pain is perceived as threatening.[10,11] Additionally, recent studies in children with chronic pain showed that pain catastrophizing (emotion-focused avoidance coping) mediated the relationship between negative affectivity (i.e. vulnerability) and somatic complaining.[12]

Social environment factors: parents and peers

Reinforcement In line with operant learning theories, we found the highest rewarding for the adolescents' behavior occurred in pain situations. Interestingly, we found this to be true only for parents and not for peers. Peers are more rewarding in a pain-free situation. An explanation for this finding might be that adolescents with pain show withdrawal in and from social situations, become more silent and do not tell their peers about their pain. By staying home and thus avoiding school or other activities, rewarding of pain behavior is more likely coming from parents than from peers.

However, while parents are more rewarding in pain situations, surprisingly this was found to be significantly less in the chronic pain group compared to the control group *(chapter 4)*. Rewarding of the adolescents' pain behavior by parents and peers was associated with a better quality of life (in terms of psychological functioning, social functioning and satisfaction with life in general) *(chapter 5)*. These findings are consistent with Osterhaus and colleagues[13]; they found a negative association between the parents' rewarding and the child's pain report.

Our results indicate that adolescents with chronic pain might be rewarded less often in pain situations than one might expect on the basis of the learning principles of (pain) behavior. Fordyce[14] argued that positive and negative reinforcement of pain behavior lead to the development of chronic pain. May be the family has grown accustomed to the pain and responds less, or loss of credibility leads to adolescents with chronic pain receiving less attention for their pain behavior. In accordance with this explanation, Jamison and colleagues[15] found that chronic pain patients perceived their families as non-supportive. It is conceivable that a child's pain report will increase as a function of their parents' lack of attention for its complaints. Considering the fact that our sample was drawn from the general population, it is conceivable that operant learning theories are more applicable to adolescents from a clinical setting with more severe pain as their pain might be more frequent and more worrying for their parents.

Our results support studies that emphasized the importance of studying the attitudes and perceptions parents have with regard to their child and the stressor.[16,17] More research on the attitude and behaviors of parents towards the pain complaints of their children is needed.

To obtain the story behind these intriguing results semi-structured interviews were held (not systematically reported in this thesis) with 25 adolescents with chronic pain and 5 without chronic pain and their mothers. The interviews consisted of open-ended questions covering pain, vulnerability, reinforcement, modeling and pain coping. The interviews demonstrated support for the identified differences between pain sufferers and controls in the cross-sectional studies. Parents of adolescents with chronic pain reported that most of the time they do not take the adolescents' pain very seriously. They feel that the pain is useful for their children in order to avoid less pleasant activities. On the other hand, they also reported feelings of insecurity, guilt and helplessness. Dealing with the chronic pain of their children is also for parents a difficult task. They reported difficulties in differentiating actual pain from acted pain. As a consequence parents may show an intermittent pattern of reinforcement, indicating that they sometimes provide emotional support and at other moments they indicate that they do not have time to assist their children when they are in pain. This inconsistent behavior pattern may also play a role in the maintenance of chronic pain in adolescents. The interviews show that the process of reinforcement is complex and difficult to entangle.

Modeling The finding that adolescents with chronic pain report more significant others with pain than adolescent without chronic pain is consistent with other studies,[18,19] but the differences are rather small. Additional analyses (not reported in this thesis) show that adolescents with severe pain report more significant others in their social environment with pain at a similar location as the adolescent. Comparing the parents of adolescents with and without chronic pain, we found no differences regarding their own chronic pain, pain parameters, pain coping strategies, and vulnerability. The latter is in contrast with a study of Hotopf et al.[20] who found that mothers of children with abdominal pain are more anxious (in terms of neuroticism).

Evaluating the model:

changing the psychosocial factors by means of an intervention program

Quality of life

Although the pilot study showed significant improvements in quality of life following the intervention, the randomized controlled trial did not show overall significant differences in change between the intervention group and the control group. This absence is in accordance with the study of Bandell-Hoekstra and colleagues.[21] Their self-management program for children and adolescents with headache did not show improved quality of life when compared with a placebo intervention.

Our results concerning quality of life might be explained by the fact that there were no clear treatment effects on pain characteristics and vulnerability. In our cross-sectional study we found that pain intensity and especially vulnerability have a strong influence on the quality of life of adolescents with chronic pain (chapter 5). The absence of significant improvements in quality of life might therefore be caused by a lack of substantial change in the pain and vulnerability of the adolescents.

We did find a significant difference in change in quality of life (in terms of functioning at home) between the intervention group and the control group at follow-up. This effect might be a consequence of the participation of parents in our intervention. Adolescents in the intervention group reported that they got on better with their parents and reported that they could talk more to their parents about personal things or problems than adolescents in the control group. Adolescents and their parents might have learned other ways to communicate with each other, which resulted in better functioning at home. Positive effects of the involvement of parents in interventions for children with chronic pain were also found in other studies.[22-25]

Pain

The pilot study showed that pain reduces after the intervention *(chapter 6)*. The randomized controlled trial showed that these changes did not differ significantly from the changes in a control group at post-treatment and follow-up *(chapter 7)*. This latter result is in contrast with the conclusions of (Cochrane) systematic reviews on the effectiveness of psychological treatments for chronic headache[26] and recurrent abdominal pain[27] in children and adolescents. Most psychological treatments evaluated in controlled trials were effective in reducing pain characteristics in both the general population and clinical samples.[23,25,28-32] However, in accordance with our results the efficacy in pain reduction of a cognitive-behavioral intervention could not be established in some studies on adolescents with chronic headache in the general population[21,33] and tertiary referred adolescents with chronic pain.[22]

Considering the increased medication intake among adolescents in the general population,[34,35] the decrease in medication use following intervention is promising. This treatment effect might indicate that adolescents in the intervention group learned other ways to cope with their pain instead of using pain medication.

Pain-related problems and psychosocial factors

In the randomized controlled trial, significant differences in change were found in psychosocial variable 'coping' and outcome variable 'pain-related problems' (PPL) at post-treatment. The intervention group, surprisingly, displayed more pain-related problems compared to the control group after the intervention period. This difference might be caused by the mechanism that adolescents are increased aware of their disabilities and coping styles due to chronic pain during the intervention. A study conducted by Howard and colleagues[36] showed that treatment subjects experience changes in their perceptions of their pre-treatment levels of functioning (i.e. coping or pain-related problems), whereas non-treatment control subjects do not. Howard and Dailey[37] additionally report that this response-shift bias might cause the absence of treatment results established with questionnaire measures.

Concluding remarks on our psychological model

The anticipated relationships within our model are partially confirmed by the results of our studies. The cross-sectional studies showed evidence for the supposition that the psychosocial factors are associated with chronic pain and quality of life. Individual examination of the factors showed that vulnerability and emotion-focused avoidance coping had the strongest significant associations with chronic pain and quality of life *(chapter 4 and 5)*. The randomized controlled study was conducted to evaluate the influence of expected changes in the psychosocial factors (by means of the intervention program) on chronic pain and quality of life. However, the psychosocial factors were not successfully manipulated with our intervention. As a consequence, there were no changes in quality of life and pain characteristics after the intervention *(chapter 7)*. Therefore, we are not able to make any conclusions on our model concerning the influence that changes in the psychosocial factors have on chronic pain and quality of life.

Methodological considerations

Definition of chronic pain

In our studies chronic pain was defined as 'continuous or recurrent (i.e. pain with pain-free intervals) pain with unknown organic etiology that exists for at least three months'. Adolescent reporting their pain as part of a diagnosed chronic disease (rheumatic arthritis, malignancies) were excluded. The rareness of these conditions would require a different design and the treatment of the disease and of the pain would be difficult to separate. In our cross-sectional sample we had to exclude only 2 adolescents from further analysis because of reported chronic pain as a result of a diagnosed chronic disease. This is comparable with the prevalence study of Perquin and colleagues.[38] In this study only one percent of the representative sample of Dutch children and adolescents was excluded because of known pathology.

Sample selection

In the cross-sectional studies, we did not impose restrictions regarding frequency or severity of chronic pain in order to obtain a community-based adolescent population. In theory, it would have been possible that adolescents who experience a mild pain that occurred once a month in the previous 3 months were considered as chronic pain sufferers while adolescents who experience severe daily pain in the previous two months were considered as controls. In practice however, only 9 (6%) of the 148 adolescents in the control group in our sample reported pain complaints that existed less than 3 months.

Our definition of chronic pain provides a broad picture on long-term pain in the general population and enables comparisons with the Dutch prevalence study of Perquin and colleagues.[38]

The selection criteria were slightly adapted in the intervention studies. In order to obtain a sample of adolescents for whom it would be possible to establish any treatment effects, the inclusion criteria for the intervention program were more stringent. Adolescents who experienced a mean pain intensity less than 30 mm on a Visual Analogue Scale (from 0 – 100 mm) in the previous month were excluded for the intervention studies. Of the enrollments for the randomized controlled trial, none of the adolescents reported pain less than 30 mm.

Response rate

The sampling procedure and the response rate in our studies deserve attention. The cross-sectional study sample was a convenience sample, instead of a representative random sample of the total population. Only a third of the eligible adolescents with chronic pain from the general population participated in our cross-sectional studies. However, we believe that our results are generalizable for adolescents with chronic pain in general because the ratio of boys and girls, the age of the adolescents, the pain intensity and reported pain location in our sample are similar to those in the representative prevalence study of Perquin and colleagues.[38]

In the randomized controlled study, the response rate was also disappointing. Only 31 adolescents with chronic pain participated. Adolescents were recruited from the general population (school newspaper, magazines) and from clinical settings (family doctor's practice, pediatrics in hospitals and via announcements in pharmacies). During the recruitment period (15 months for the general population and 2 months for the clinical population) 80 adolescents were initially interested in our study and requested additional information on the program. Of these, 44 (55%) were from secondary schools, 35 (44%) responded on the articles and ads and 1 (1%) enrollment was obtained from a hospital in Rotterdam-area. Considering the prevalence of (severe) chronic pain in Dutch adolescents this is an extremely low enrollment. Up to 33% of the Dutch adolescents report persistent or recurrent pain.38 A smaller proportion, 16% of the adolescent girls and 6% of the adolescent boys, report severe disabling chronic pain indicating their eligibility for our intervention. Our difficulties in obtaining an adolescent sample might be comparable to the sample selection of Bandell-Hoekstra and colleagues.[21] They recruited more children attending elementary school than to those in the age of high school for their pain-coping program. Of the 940 children and 1418 adolescents with headache from the general population who participated in their previous sample, 433 (18%) were eligible for the intervention. Eventually, they included 107 (11%) children and only 51 (3.5%) adolescents in the intervention study. It remains unclear what the reasons were for this small percentage of participating adolescents, but it does indicate that obtaining an adolescent sample was more difficult to realise than a sample of younger children. These difficulties could be explained from developmental perspective. Socially, adolescence is marked by an increasing independence from parents. Younger children are more susceptible and might be enrolled by their parents because they value the intervention. Claar and Walker39 found that many mothers acknowledge the contribution of psychosocial factors to their children's abdominal pain. They may be more receptive to behavioral interventions for their children addressing this issue.

During adolescence a more stable and consolidated sense of identity is developing. This occurs due to the fact that adolescents model their identity and behavior on others. Adolescents are increasingly able to think in an abstract and hypothetical manner. Thus, they may well reflect on how they are perceived by hypothetical others (peers), and may feel ashamed for their pain towards peers. The additional stress of pain during adolescence, a period of rapid physical and psychological change may be particularly difficult for the adolescent to cope with, but does not guarantee enrollment for intervention programs. Enrollment for an intervention because of chronic pain emphasizes differences with other adolescents and might therefore be avoided by the adolescents.

Unfortunately, we do not have data available on the reasons why the 41 adolescents who initially were interested in participating withdrew before the study started. Beside developmental aspects, another reason may be the amount of questionnaires and the pain diary they had to complete at three assessment periods and the additional uncertainty of not knowing beforehand whether they could expect personal benefit from the intervention at short notice (allocated to the intervention group) or not (allocated to the control group). In addition, the pilot study (chapter 6) showed that the most common reason for eligible adolescents not to participate in our program was because their pain was not (or no longer) severe enough for an intensive program or because the pain had already disappeared.

Pain measurement

Considering the subjective nature of pain, self-report measures on pain are presumably the most valid and reliable method of assessment. Research has demonstrated that prospective diaries, as compared to retrospective questionnaires increase the validity of children's pain report. Brink and colleagues40 found that headache intensity and headache duration were, compared to the diary measurement, overestimated in the questionnaire. Unlike retrospective methods, prospective diaries do not require children to summarize or average their behavior and can elicit more accurate descriptions.41 A limitation in our first cross-sectional study (chapter 4) is that we did not include a prospective pain measure (diary). Considering the results of Brink and colleagues40 we should be aware of the fact that recall errors might have occurred in our study on the relationship between psychosocial factors and pain (parameters) (chapter 4). The overestimation of pain intensity when retrospectively measured was supported by the results in chapter 5 in which we found lower prospective pain scores compared with retrospectively measured pain scores, and by the results of others studies.[42,43]

The absence of treatment effects on quality of life and pain

The absence of clear effects of our training program may be related to the lack of power for demonstrating small effects. Our sample size was rather small and the compliance at post-treatment and follow-up measures was not optimal. Besides the small sample size, several other considerations concerning the absence of effects have to be made. The first concerns the intervention program. Considering the effectiveness of pain programs in previous studies,[26,27] the use of cognitive-behavioral techniques in our pain management program is supported by evidence. However, the transfer of knowledge may not have been adequately succeeded and the change in behavior may not have been acquired by the adolescents. The lack of substantial change in vulnerability of the adolescents might be explained by the notion that neuroticism (i.e. vulnerability) is a stable personality characteristic and could only be influenced indirectly by a short intervention (e.g. by learning adolescents to cope in daily stressful situations). Although a cognitive-behavioral method is an appropriate method used in a wide variety of groups, no empirical evidence is available regarding which elements of the training are most beneficial for adolescents with chronic pain.

Secondly, our program was aimed at a broad range of adolescents. Adolescents with retrospective pain intensity of at least 30 mm on a VAS were included in our study. We argued that these adolescents are at risk of pain adjustment problems and may therefore profit from a cognitive-behavioral pain program. As a consequence of the absence of specific clinical based criteria, the pain intensity (prospectively measured) at pre-treatment was relatively low in comparison with adolescents that were included in other clinical-based[22] or general population[21] studies. It will be more difficult to demonstrate an intervention effect, because the pain reduction is expected to be larger in adolescents with higher pre-treatment pain intensity scores.[44] Third, the questionnaires used in our study may not be sensitive enough for treatment evaluation. The fourth consideration is related to our actions to enhance the compliance for the assessments. We sent the adolescents questionnaires and the pain diary with a small present and we further stayed in contact with them (sending Christmas-cards, holiday-cards, telephone contacts to motivate them to complete and return the questionnaires and pain diary). These actions might be interpreted as attention for the adolescents and might bring about a non-specific therapeutic effect.

Clinical implications

With the shortened QLA-CP health care providers are able to obtain a reliable and valid indication of the quality of life in adolescents can be assessed. This multidimensional questionnaire is suitable for adolescents with chronic pain irrespective of their pain location. In combination with the PPL a complete picture on the impact of chronic pain on daily functioning of adolescents with chronic pain can be obtained.

The results of the cross-sectional studies indicate that pain management programs should consider multiple psychosocial factors related to chronic pain rather than focusing on a single factor only. Based on the results in *chapter 5,* interventions aiming at reducing the impact of pain on quality of life in adolescents with chronic pain should primarily focus on techniques aiming at stress-coping skills of the adolescents in order to enhance their efficacy in dealing with stressful situations (e.g. pain). Besides a focus on these techniques, the role of the social environment (i.e. parents and peers) deserves attention in interventions.

Adolescents who participated in the intervention reported improvements in quality of life and reductions in pain characteristics at post-treatment and follow-up *(chapter 6 and 7)*. However, because there was no difference in change in quality of life and pain characteristics between the intervention group and the control group no positive conclusions on the effectiveness of the intervention can be drawn yet. If used in daily practice or clinical studies, adolescents and their parents should be told that the effect of treatment is not yet confirmed. The significant effect concerning the functioning of adolescents at home indicates the potentially important role of parents in treatment. Health care providers should address the role of social-environment factors in chronic pain and the difficulties parents experience in parenting chronic pain children.

Recommendations for further directions

1. Given the cross-sectional design in this thesis, caution in interpreting the causality of our results is warranted. Prospective longitudinal designs and treatment intervention studies are needed to disentangle the issue of causality. The Generation-R study in Rotterdam is such a prospective study. In this study, among others, the development of chronic pain is examined in a sample of approximately 10.000 mothers and infants from the prenatal period to adolescence. This study focuses on maternal vulnerability as main

determinant of chronic pain in the child and investigates the moderating effect of parenting attitudes and child vulnerability.

2. A replication of our randomized trial with a larger sample is advisable. Researchers need to consider the following changes:

   a. Another format. Considering the frequent use of computers by adolescents, an interactive website offering cognitive-behavioral treatment might be more appealing to them. Previous studies have shown that computer assisted cognitive behavioral therapy is effective for individuals with increased depressive symptoms at the community level[45] and in general practice.[46] Internet sites offer feasible and powerful public health interventions. The effectiveness of such formats for adolescents with chronic pain should be addressed in future research.

   b. Other ways of data collection. Future research should preferably include electronic diaries with compliance-enhancing features because they are a more effective way of collecting diary information[46] and show – even in children - significantly greater compliance and accuracy compared to traditional paper diaries.[48]

   c. Another sample. A replication could be conducted with adolescents who are referred to secondary care. Smith and colleagues[49] show that adolescents who seek behavioral treatment for recurrent headache report headaches of longer duration, missed more school days due to headache, and report higher initial sustained discomfort scores. To enlarge the number of participants a multicentre randomized controlled would be required.

3. Considering the fact that important aims of interventions are improving one's quality of life, daily functioning and coping strategies, it is essential that the responsiveness of the QLA-CP and the PPL are further established as well as their test-retest reliability and cross-cultural validation.

4. As the intervention consisted of a combination of various elements (e.g. relaxation, cognitive techniques) we cannot comment on the effect of the separate therapeutic strategies, but this would make an interesting focus for future research.

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Summary

Chapter 1 presents an overview of psychosocial factors (vulnerability, reinforcement, modeling and coping) that are often associated with the development and maintenance of chronic pain in adolescents. Previous studies on these psychosocial factors are discussed. Additionally, the chapter describes psychological theories on chronic pain and available intervention programs for adolescents with chronic pain. The psychosocial factors are incorporated in a model on pain and quality of life in adolescents with chronic pain. The main aim of this thesis is to evaluate this model. The accompanying research questions are presented.

Chapter 2 describes the shortening of the Quality of Life questionnaire for adolescents with Headache-Youth (QLH-Y). We were interested in obtaining a quality of life questionnaire suitable for adolescents with chronic pain irrespective of their pain location. We reduced the original 71 items of the QLH-Y to 44 items in our new version of the questionnaire, the Quality of Life Questionnaire for Adolescents with Chronic Pain (QLA-CP). The original domains of quality of life (psychological functioning, functional status, physical functioning, social functioning, satisfaction with life in general and satisfaction with health) were maintained. The questionnaire was validated for adolescents with chronic pain irrespective of their location and showed suitable internal consistency and construct validity against COOP/WONCA charts. The QLA-CP discriminates between adolescents with pain on both intensity and frequency. We conclude that the QLA-CP is a reliable and valid instrument to measure the quality of life of adolescents with chronic pain irrespective of their pain location.

In **chapter 3** the development of the Pain-related Problem List (PPL) is described. This short questionnaire (18 items) is based on the personal experiences of adolescents with chronic pain. The items were concerned with problems related to concentration, mobility, adaptability and mood. The internal consistency of the domains and the total scale proved to be adequate. Evidence was found for the validity of the total scale and four domains in both a general population and a clinical sample. The PPL discriminates between adolescents with high pain intensity and low pain intensity. Overall, we conclude that the PPL is a reliable and valid instrument to measure pain-specific problems in adolescents with chronic pain irrespective of their pain location, especially in the general population. Combining a generic quality of life questionnaire like the QLA-CP with a pain-specific instrument is advisable for use in studies evaluating interventions. This enables the assessment of pain related quality of life of adolescents with different types of chronic pain.

Chapter 4 and 5 report on the evaluation of our model with a cross-sectional study design.

Chapter 4 describes a comparison between 222 adolescents with chronic pain and 148 controls (12 through 18 years). Psychosocial factors of the model (vulnerability, reinforcement, modeling and coping), pain and the quality of life of both groups of adolescents were compared. Parents of the adolescents of both groups were compared on pain and psychosocial factors. Compared to healthy controls, adolescents with chronic pain are more vulnerable in terms of neuroticism, negative fear of failure, and they experience less social acceptance. Contrary to our expectations, adolescents from the chronic pain group experience less reinforcement for their pain behaviour by parents and peers than those from the control group. While the number of significant others suffering from pain was higher in the chronic pain group, no differences were found between their parents and those of the adolescents without chronic pain in pain experience, pain parameters, and pain coping. Regression analyses on the contribution of psychosocial factors to chronic pain and its parameters sustained the positive relation between pain (parameters) and the psychosocial factors vulnerability, the number of pain models, (less) pain reinforcement, and coping with pain.

In **chapter 5** the examined relationships between demographics, pain characteristics, psychosocial factors and quality of life among 194 adolescents with chronic pain are presented. The results of this population based cross-sectional study showed that psychosocial factors account for a significant variance in the adolescent's quality of life, even when controlling for pain characteristics. Overall, vulnerability (in terms of neuroticism) contributed significantly and uniquely to the variance of most quality of life domains. In addition, we found that pain coping styles moderate the relation between pain intensity and quality of life. The negative relation between pain intensity and psychological functioning was stronger at higher levels of emotion-focused avoidance coping.

Chapter 6 and 7 report on the evaluation of our model with a randomized controlled study design. Our model was tested by studying changes in pain and quality of life due to changes in the psychosocial factors as a consequence of the intervention program.

Chapter 6 presents our cognitive-behavioral training program for adolescents with chronic pain irrespective of their pain location. This program is based on the results of the cross-sectional studies presented in chapter 4 and 5. The purpose of the pilot study was to evaluate the feasibility of the program. The results in the pilot study indicate that our intervention program is feasible and fits well in daily life of adolescents. Both adolescents and their parents were positive about the program. The adolescents reported that the training helped them in gaining more control of their pain, and feeling less like a victim of pain. Parents evaluated the training and their involvement as supportive and informative because this enabled them to support their children in mastering their pain. The preliminary data showed an effect on quality of life and pain in the expected direction.

Chapter 7 describes a randomized controlled trial that was used to explore the effect of the intervention on quality of life and pain. Subjects (n=31) were randomly assigned to the experimental group or the control group. No clear intervention effects could be demonstrated in this randomized controlled trial. We found a similar improvement in quality of life and decrease in pain intensity and frequency between the intervention and control group. However, there was a significant main effect for the quality of life subscale functioning at home, indicating that adolescents in the intervention group display better functioning at home than adolescents in the control group. Differences are also found in the level of medication use between both groups at post-treatment and follow-up. Adolescents from the intervention group display a significantly lower level of medication use than adolescents in the control group. Additionally, no clear significant differences in change in the psychosocial factors were found between the intervention and control group. At post-treatment differences were found in coping strategies and pain-related problems. The intervention group displays significantly more pain-related problems and more use of emotion-focused avoidance coping than the control group.

In **chapter 8** the main results of these studies are discussed, and conclusions and suggestions for future research are given.

Samenvatting

Hoofdstuk 1 is de inleiding van dit proefschrift. In dit hoofdstuk worden verschillende psychosociale factoren beschreven die vaak gerelateerd worden aan de ontwikkeling en/of het blijven bestaan van chronische pijnklachten bij adolescenten. De factoren die toegelicht worden zijn: 1) de psychologische kwetsbaarheid van de adolescent; 2) de mate van beloning van pijngedrag door ouders en leeftijdsgenoten; 3) de aanwezigheid van pijnklachten bij anderen in de omgeving van de adolescent (bijv. ouders met chronische pijn); en 4) de manier waarop adolescenten omgaan met hun pijnklachten. Dit hoofdstuk presenteert een literatuuroverzicht van eerder onderzoek gericht op de relatie tussen de genoemde psychosociale factoren en chronische pijnklachten bij kinderen en adolescenten. Aansluitend worden gangbare psychologische theorieën over chronische pijnklachten en beschikbare interventieprogramma´s voor kinderen en adolescenten met chronische pijnklachten beschreven.
Dit proefschrift is gericht op het verwerven van inzicht in de relatie tussen psychosociale factoren, chronische pijnklachten en de kwaliteit van leven van adolescenten. Deze factoren zijn samengebracht in een model welke bestudeerd zal worden in verschillende deelstudies. De bijbehorende onderzoeksvragen worden gepresenteerd aan het einde van dit hoofdstuk.

Hoofdstuk 2 beschrijft de inkorting van een kwaliteit van leven vragenlijst voor adolescenten met hoofdpijn (QLH-Y). Naast het inkorten van de vragenlijst was het doel van deze deelstudie het verkrijgen van een kwaliteit van leven vragenlijst dat geschikt is voor adolescenten met chronische pijnklachten ongeacht de pijnlocatie. De oorspronkelijke 71 items van de QLH-Y konden gereduceerd worden tot 44 items in de nieuwe versie van de vragenlijst, de kwaliteit van leven vragenlijst voor adolescenten met chronische pijn (QLA-CP). Alle oorspronkelijke domeinen van kwaliteit van leven (psychologisch functioneren, functionele status, fysiek functioneren sociaal functioneren, tevredenheid met leven in het algemeen en tevredenheid met gezondheid) bleven gehandhaafd. De vragenlijst is gevalideerd voor adolescenten met chronische pijn ongeacht hun pijnlocatie en heeft een acceptabele interne consistentie en construct validiteit ten opzicht van COOP/WONCA kaarten. De QLA-CP maakt onderscheid tussen adolescenten met hoge pijn intensiteit / frequentie en adolescenten met lage pijn intensiteit / frequentie. Geconcludeerd wordt dat de QLA-CP vragenlijst een betrouwbaar en valide instrument is om de kwaliteit van leven van adolescenten met chronische pijnklachten te meten.

In **hoofdstuk 3** wordt de ontwikkeling van de pijn-gerelateerde problemen lijst (PPL) beschreven. Deze korte vragenlijst (18 items) is gebaseerd op de persoonlijke ervaringen van adolescenten met chronische pijn. De items zijn gericht op problemen gerelateerd aan concentratie, fysiek functioneren, aanpassingen en de stemming van adolescenten. De interne consistentie van de domeinen en de totale schaal bleek adequaat. De validiteit van de totale schaal en de vier domeinen is aangetoond voor zowel een algemene als klinische populatie. De PPL maakt onderscheid tussen adolescenten met hoge pijn intensiteit en lage pijn intensiteit. Geconcludeerd kan worden dat de PPL een betrouwbaar en valide instrument is om, met name in de algemene populatie, pijn-specifieke problemen te meten bij adolescenten met chronische pijn ongeacht de pijnlocatie. Het hoofdstuk wordt afgesloten met de aanbeveling dat een generieke kwaliteit van leven vragenlijst zoals de QLA-CP gecombineerd kan worden met een pijn-specifiek instrument zoals de PPL voor het evalueren van interventies. Een dergelijke combinatie maakt een zorgvuldige waardering van de pijn-gerelateerde kwaliteit van leven van adolescenten met chronische pijnklachten mogelijk.

Hoofdstuk 4 en 5 beschrijven de evaluatie van het model met een cross-sectioneel design.
Hoofdstuk 4 presenteert een vergelijking tussen 222 adolescenten met chronische pijn en 148 adolescenten zonder chronische pijn (12 t/m 18 jaar). Beide groepen werden vergeleken op de psychosociale factoren van het model (psychologische kwetsbaarheid, beloning voor pijngedrag, aanwezigheid van pijnklachten bij mensen in de omgeving van de adolescent en de manier waarop adolescenten omgaan met pijnklachten), pijn en de kwaliteit van leven. Ouders van adolescenten uit beide groepen zijn daarnaast vergeleken op pijnklachten en psychosociale factoren. Vergeleken met adolescenten zonder chronische pijn, zijn adolescenten met chronische pijn psychologisch kwetsbaarder (zij rapporteren een hoge mate van neuroticisme, aanwezigheid van negatieve faalangst, en een lage sociale acceptatie door leeftijdsgenoten). In tegenstelling tot onze verwachting ervaren adolescenten met chronische pijn minder beloning voor pijngedrag dan adolescenten zonder chronische pijnklachten. Terwijl het aantal mensen met pijnklachten in de omgeving hoger is in de chronische pijn groep, blijken er geen verschillen te zijn tussen de ouders van beide groepen in de aanwezigheid van pijnklachten, pijn intensiteit en frequentie en het omgaan met pijnklachten. Regressie analyses naar de bijdrage van psychosociale factoren op chronische pijn en pijn karakteristieken (intensiteit en frequentie van pijn) ondersteunen de positieve relatie tussen pijn (karakteristieken) en de

psychosociale factoren (psychologische kwetsbaarheid, aantal pijnmodellen, lage beloning voor pijngedrag en manieren van omgaan met pijn).

In **hoofdstuk 5** wordt de relatie tussen demografische gegevens, pijn karakteristieken, psychosociale factoren en kwaliteit van leven bij 194 adolescenten met chronische pijn beschreven. De resultaten van deze cross-sectionele studie laten zien dat de kwaliteit van leven van adolescenten met chronisch pijn redelijk goed voorspeld kon worden op basis van psychosociale factoren, ook nadat er gecorrigeerd is voor kenmerken van pijn. Binnen de groep psychosociale factoren bleek dat de variabele psychologische kwetsbaarheid (neuroticisme) sterk geassocieerd was met het merendeel van de kwaliteit van leven domeinen. Daarnaast laten de resultaten zien dat de manier waarop omgegaan wordt met pijn van invloed is op de relatie tussen pijn en kwaliteit van leven. Deelnemers met veel pijn die gebruik maken van emotiegerichte vermijding in het omgaan met pijn bleken een slechtere kwaliteit van leven (psychologisch functioneren) te rapporteren dan deelnemers die evenveel pijn ervaren maar minder gebruik maken van emotiegerichte vermijding.

Hoofdstuk 6 en 7 zijn gericht op het beschrijven van de inhoud en het effect van een cognitieve gedragsmatige interventie.
Hoofdstuk 6 beschrijft de cognitief-gedragsmatige training voor adolescenten met chronische pijn. Dit programma is gebaseerd op de resultaten van de cross-sectionele studies (hoofdstuk 4 en 5). Met de pilot studie werd de toepasbaarheid van de interventie geëvalueerd. De resultaten geven te kennen dat de interventie goed inpasbaar is in het leven van adolescenten. De interventie werd door adolescenten én hun ouders positief gewaardeerd. Adolescenten rapporteerden dat de training hen heeft geholpen in het hervinden van controle over pijn, waardoor ze zich minder een slachtoffer van de pijn voelden. Ouders evalueerden de training en hun betrokkenheid als ondersteunend en informatief. Ze voelden zich beter in staat om hun kind te ondersteunen in het de baas worden over pijn. De eerste resultaten laten een effect zien op kwaliteit van leven en pijn in de verwachte richting.

Hoofdstuk 7 beschrijft een gerandomiseerd gecontroleerde studie dat gebruikt is om het effect van de interventie op kwaliteit van leven en pijn te bestuderen. Deelnemers (n=31) werden willekeurig over twee groepen verdeeld, de interventie groep en een controle groep. De gerandomiseerde studie liet geen duidelijke interventie effecten zien. Er werd een vergelijkbare verbetering in kwaliteit van leven en vermindering van pijn gevonden tussen de interventie groep en de controle groep. Een significant hoofdeffect werd gevonden voor de kwaliteit van leven subschaal ´functioneren thuis´; adolescenten in de interventie groep blijken na de interventie thuis beter te functioneren dan adolescenten in de controle groep. Verschillen zijn eveneens gevonden in de mate van medicatiegebruik tussen beide groepen vlak na het doorlopen van de training en 6 maanden later. Adolescenten in de interventie groep zijn significant minder medicatie gaan gebruiken na de interventie in vergelijking met adolescenten in de controle groep. Geen duidelijke significante verschillen zijn gevonden in de verandering in psychosociale factoren tussen de interventie en de controle groep. Na afronding van de training zijn verschillen gevonden in de manieren waarop omgegaan wordt met pijn en pijn-gerelateerde problemen. Adolescenten in de interventie groep rapporteren meer pijn-gerelateerde problemen en gebruiken meer emotiegerichte vermijding in het omgaan met pijnklachten in vergelijking met adolescenten van de controle groep.

In **hoofdstuk 8** worden de belangrijkste resultaten van deze studies bediscussieerd. Conclusies en suggesties voor verder onderzoek worden gepresenteerd.

Dankwoord

Veel mensen hebben meegewerkt, geholpen en ondersteuning geboden gedurende de periode waarin ik aan dit proefschrift heb gewerkt. Graag wil ik van de gelegenheid gebruik maken om deze personen bedanken.

Allereerst dank ik alle deelnemers aan het onderzoek. Zonder jullie was er helemaal geen onderzoek geweest. De scholen die zo gastvrij waren ons te ontvangen, de adolescenten en hun ouders die de vragenlijsten hebben ingevuld en vooral de deelnemers aan de interventiestudie. Jullie hebben allen veel energie gestoken in het registreren van pijnklachten, het invullen van de vragenlijsten en de gesprekken die gevoerd zijn, zonder dat het tevoren vast stond of jullie zelf baat zouden hebben bij het onderzoek. Bedankt dat jullie je ervaringen met mij wilden delen.
Mijn promotoren, prof. Dr. Jan Passchier en prof. Dr. Bart Koes. Jan, je hebt me de ruimte gegeven en alle kansen geboden om te groeien in de wetenschap. Dit heeft mij gestimuleerd in mezelf te blijven geloven, ook als het soms wat minder ging. De ervaring die ik dankzij jou heb kunnen opdoen op het gebied van onderwijs geven ervaar ik als zeer waardevol. Bart, je oprechte betrokkenheid bij mij en mijn onderzoek zijn een stimulans geweest om dit proefschrift tot een goed einde te brengen. Door je heldere en kritische commentaar op alle tekst van dit proefschrift heb ik heel wat bijgeleerd over het schrijfproces. Gezien de omvang van het dankwoord blijkt echter wel dat ik de kunst van het beknopt weergeven nog niet helemaal onder de knie heb....
Copromotor en begeleider, Dr. Joke Hunfeld. Mijn dank is groot voor je betrokkenheid en begeleiding gedurende het project. Ik ben ervan overtuigd dat de consequente manier waarop je de voortgang van het project en het (re-) submitten van onze artikelen kritisch hebt gevolgd (en daarvoor heb je mij regelmatig achter de broek aan moeten zitten), er voor gezorgd heeft dat het onderzoeksproject uiteindelijk met dit proefschrift kan worden afgerond.
Prof dr. Siep Thomas, hoogleraar Huisartsgeneeskunde en instituutbeheerder, bedankt voor het interim-promotorschap.
De leden van de begeleidingscommissie ben ik dankbaar voor hun inzet en betrokkenheid. Dr. Hans van der Wouden, methodologisch en tekstueel een kei, maar belangrijker nog: erg betrokken en altijd bereid mee te denken en/of te adviseren. Dr. Alice Hazebroek-Kampschreur, beschikkend over waardevolle

ervaring op het gebied van werving in open populaties, tactvol en open in feedback. Dr. Lisette van Suijlekom-Smit, altijd oog voor de relevantie van onderzoeksresultaten voor de klinische praktijk. Bedankt dat je mij - in mijn eerste aio-jaar - enkele van jouw patiënten toevertrouwde waardoor ik naast een theoretisch kader ook de gelegenheid kreeg grip te krijgen op de belevingswereld van adolescenten met chronische pijnklachten.
Prof. Dr. S. Thomas, Prof. Dr. H.A. Büller en Prof. Dr. J.M. Koot. Ik wil u bedanken voor het zorgvuldig doornemen van het manuscript en uw bereidheid om deel uit te maken van de promotiecommissie.
Het Pijnkenniscentrum (PKC) te Rotterdam en de Stichting Algesiologie wil ik bedanken voor de financiële steun die zij gegeven hebben waardoor verschillende deelstudies van dit proefschrift uitgevoerd konden worden.

Gezien de omvang van het project heb ik op verschillende momenten het gevoel gehad ´door de bomen het bos niet meer te zien´ en ten onder te gaan in een overload aan administratieve taken. Gelukkig waren er herhaaldelijk mensen die mij op zo´n moment uit de brand wilden helpen. Jacqueline Mourik en Petra Jellema, bedankt voor jullie hulp bij het nummeren en verzenden van de eerste vragenlijsten en pijndagboekjes toen mijn Vivian-planning (weer eens) onhaalbaar bleek. Niels en Ewout, bedankt voor jullie hulp bij het uitvoeren van de telefonische reminders om de respons in de ´case-control´ studie zo hoog mogelijk te krijgen. Jacoba en Ankey, bedankt voor jullie hulp bij het verwerken van alle vragenlijsten en pijndagboekjes. Aansluitend wil ik ook graag de enthousiaste studenten noemen die in het kader van hun studie (doctoraal onderzoek, stageperiode of keuzeonderzoek) onder mijn supervisie hebben meegewerkt aan het onderzoek. Anneke, Marije, Sara, Conny, Laurien en Melanie. Bedankt voor jullie gezelligheid, maar vooral ook jullie behulpzaamheid bij het uitvoeren van de geprotocolliseerde behandeling en het consciëntieus verzamelen van data.

Larraine Visser-Isles en Michelle Peters, jullie engelse correcties hebben er voor gezorgd dat de publicaties uit dit proefschrift ook buiten Nederland gelezen kunnen worden.

Dank ook aan mijn collega´s van de afdeling Medische Psychologie en Psychotherapie. Hoewel iedereen heeft bijgedragen aan de goede tijd die ik op de afdeling heb gehad, wil ik Benno Bonke, Josien de Boer en Ingrid-Emilie Wouterlood- van Cleeff nog bij name noemen. Ik heb veel van jullie geleerd over onderwijs (geven) tijdens onze samenwerking (bij het opzetten van het attitudeonderwijs voor co-assistenten Kindergeneeskunde en/of tijdens het uitwisselen van cohag-ervaringen). Dankzij jullie ben ik als docent gegroeid.

De mensen van het secretariaat, Margreet, Ingrid, Loes†, Alice, Marja en Ankey, hebben mij regelmatig geholpen met allerlei praktische zaken. Het was altijd prettig om even bij jullie aan te waaien en gezellig te kletsen.
Veel lol en leed heb ik gedeeld met de ´jonge garde´van de afdeling. De regelmatig geplande vrijdagmiddagborrels, etentjes, verjaardagen, sinterklaasavonden, e.d. waren altijd gezellig en vormde een goede afwisseling van het wetenschappelijke. Anita, Litanja, Cecile en Marleen; ik vind het bijzonder dat we nog steeds van die gezellige etentjes hebben. Volgende keer weer als Chantal in het land is?
Mijn kamergenoten Jacqueline en Paula. Gezamenlijk zijn we van cf 308 naar onze eigen kamer beneden verhuisd!! Aan werken kwamen we soms bijna niet toe omdat onze opstelling wel heel gezellig was. Jacqueline, dankzij jou kon ik regelmatig overnachten in het huis van je moeder aan de Coolsingel. Dat was echt een uitkomst. Bedankt! Ook bij Annemarie was er altijd wel een plekje beschikbaar om te overnachten. Als ´buren´ hebben we later nog regelmatig hard gelopen, gezwommen in het Oostervant of gewoon lekker Bon Bon Bloc´s gegeten.
Saskia, je hebt veel PIPO-kwaliteiten! Je bent een ras-optimist en bleef aanhoudend het belang van onze werkgroep uitdragen. Ik vind het jammer dat onze PIPO-bijeenkomsten nooit echt helemaal van de grond zijn gekomen, maar aan jou heeft dat in ieder geval niet gelegen.
Adriaan, Cora, Iris, Jolie, Leonieke, en Silvia. Bedankt voor alle gezelligheid in de afgelopen jaren. Ik weet zeker dat ook nieuwe aio´s het bij mpp leuk zullen gaan hebben omdat jullie daar nog rondlopen.

De medewerkers van de afdeling Huisartsgeneeskunde dank ik voor hun interesse en collegialiteit. Ik heb veel goede herinneringen aan de tijd dat ik een kamer deelde met Christel. Regelmatig werd werk afgewisseld met leuke groepsactiviteiten (squashen, badminton, uiteten, tijdens de lunch met een goep trappen lopen naar de 21^{e} verdieping, houtenspeelgoed party's etc.). Annette, Arianne, Celinde, Christel, Claudia, Lya, Marco, Mariet, Petra, Sander, Sita en Wouter, bedankt voor die goede herinneringen.
Christel, jij was toch wel mijn grote voorbeeld. Mijn onderzoek was tenslotte een vervolg op jouw promotie onderzoek en jij liet me zelfs als paranimf van dichtbij meemaken hoe de verdediging van een proefschrift dient te verlopen. Uit de tijd dat wij een kamer deelden zal ik je cappuccino- en muntendrop verslaving niet snel vergeten. Bedankt voor alle gezelligheid, ook na werktijd (zoals squashen, weekje vakantie in Finland en logeerpartijtjes bij jou, Gerrit, Floor en Lotte)!
Petra, ik heb zelden iemand ontmoet die zo duidelijk oog heeft voor de life-events van een ander en daarbij perfect aanvoelt hoe je diegene een hart onder de riem moet steken. Het is een gemis voor de psychologie dat je gestopt bent met de opleiding. Het contact dat we hebben opgebouwd tijdens het werk in

Rotterdam (dat ooit begonnen is met gezamenlijk pizza's eten in de keuken van hag en daarbij discussieren over de - psychologische - ontwikkelingen in het Big Brother huis) zetten we gelukkig nog steeds voort tijdens gezellige etentjes (waarbij pizza is vervangen door saté met friet!!) en mail wisselingen.

Naast het werk is er ook nog een heel andere wereld! Deze wereld heeft mij steeds weer laten zien dat er meer is dan werken alleen. Allereerst mijn vrienden, trouwe supporters aan de zij-lijn.
Carolien, we hebben in de afgelopen 6 jaar (en in de 14 daarvoor) veel mooie maar ook moeilijke dingen gedeeld. Steeds hebben we elkaar onvoorwaardelijk kunnen steunen en dat maakt onze vriendschap zo bijzonder voor mij.
Susan en Annelies, als een soort 'drie musketiers' weten we elkaar steeds weer te vinden. Vaak hebben we weer een hoop bij te praten en is er een betere manier dan dat met lekker eten en een wijntje te doen? WW-A, met veel goede herinneringen denk ik terug aan onze ´zon-zee-rosé´ fietsvakantie in Frankrijk. Ik geloof niet dat ik ooit in twee weken zoveel gelachen heb!!

Axel, Mischa, Carin en Jeannette, goede studievrienden sinds de intro-week 1992. Door het hele traject heen hebben jullie al mijn verhalen over de wetenschap aangehoord. Meerdere malen hebben jullie mij voor gek verklaard om te willen promoveren (gezien jullie psychologische achtergrond moet ik die diagnose misschien toch eens serieus nemen...). Bedankt voor jullie relativerende steun, alle gezellige dingen die we gedaan hebben maar vooral voor jullie vriendschap.

Ubel en Janneke, bedankt voor jullie oprechte belangstelling. Onze culinaire etentjes met Marcel, Irina, Pieter en Ingeborg blijf ik een geweldig initiatief en een leuke uitdaging vinden. Wij hebben inmiddels lekkere ideeën voor het nagerecht...

Tennissen is de laatste jaren een bron van ontspanning geworden. Ik wil Hilda, Chantal, Susan, Lida, Mike, Albert, Mirjam en Dieter, dan ook graag bedanken voor de ontspanning en gezelligheid tijdens het tennissen en het nazitten in het clubhuis.

Graag wil ik ook een woord van dank uiten naar Henk en Hennie. Door alle vakanties in Chabottes en weekjes weg met Hapimag heb ik de wetenschap regelmatig kunnen laten voor wat het is. Bedankt voor al die weken rust en ontspanning! Met alle plezier ga ik weer met jullie, Hans, Rian en Erik de dag na de promotie mee skiën.

Mijn paranimfen, Anita en Yvonne. Lieve Anita, ooit was je alleen ´een collega´, maar inmiddels zijn we goede vrienden geworden. Onze fietsweekenden, gezellige etentjes, bioscoop-bezoeken (nadat we eerst de boeken gelezen hadden), worden nu ook afgewisseld met ´partner-uitjes´. Wie weet worden het ooit hele gezins-uitjes? Ik vind het een mooie symbolische uiting van onze vriendschap dat jij, ter steun, 22 december naast mij zal staan.
Lieve Yvonne, al staat de wetenschappelijke wereld ver van jou af, je bent altijd geïnteresseerd geweest in hoe ik het er in die andere wereld vanaf bracht. Het geeft me een gerust gevoel dat ik straks mijn ´grote zus´ naast me heb staan tijdens de verdediging van mijn proefschrift. Ik heb bewondering voor de manier waarop jij jouw leven vorm geeft. Ik hoop dat Nienke, Wessel en Jelmer, de kinderen van jou en Theun, in veel opzichten op je zullen gaan lijken. Ik ben er trots op dat jij mijn zus bent en hoop dat we nog regelmatig zullen genieten van onze zussen-dagen.

Lieve papa en mama, jullie zijn er altijd voor mij! Ik geloof niet dat woorden echt goed kunnen beschrijven hoe bijzonder ik dat vind. Jullie onvoorwaardelijke liefde en steun hebben mij het vertrouwen gegeven om mijn eigen weg te gaan in dit leven. Als ik ook maar iets van jullie warme belangstelling, oprechte aandacht en zorg voor anderen heb meegekregen dan hoop ik dat dat tot uiting mag komen in mijn werk als psycholoog.

Lieve Hans, gelukkig bleef jij altijd jezelf als ik weer eens gestrest rondliep. Je hebt me regelmatig uit mijn eigen wereldje weten te halen met de opmerking: "Maak er geen scriptie van!" Jouw nuchtere en rationele kijk op de wereld is vaak verhelderend (net zo als je excel-sheets) en heeft er voor gezorgd dat ik deze fase van mijn leven met een goed gevoel kan afsluiten.

Amersfoort, 2004

Curriculum Vitae

Vivian Merlijn werd geboren op 22 februari 1973 te Hoorn. Na het behalen van het HAVO diploma aan het Marcus College te Grootebroek (1991), is zij begonnen aan de Hogere Beroepsopleiding tot Verpleegkunde (HBO-V) aan de Hogeschool van Alkmaar. Na het behalen van het propedeuse diploma startte zij in 1992 de studie Psychologie aan de Vrije Universiteit te Amsterdam, met als afstudeerrichting Klinische Kinder- en Jeugdpsychologie. Voor haar doctoraalscriptie heeft zij een retrospectief onderzoek uitgevoerd naar kinderobesitas en levensgebeurtenissen. Haar klinische afstudeerstage heeft zij doorgebracht bij de afdeling Medische Psychologie - sectie kinderen, van het VU medisch centrum te Amsterdam. Na het behalen van haar doctoraalexamen werd zij bij deze afdeling aangesteld als medisch psycholoog (september 1996 tot maart 1997). Van maart 1997 tot januari 1998 heeft zij als onderzoeksassistent meegewerkt aan het 'post-cambodja klachten onderzoek' bij de afdeling Medische Psychologie van de Vrije Universiteit Amsterdam.

Van april 1998 tot april 2002 was zij als Assistent in Opleiding verbonden aan de afdeling Medische Psychologie en Psychotherapie en de afdeling Huisartsgeneeskunde van het Erasmus MC Rotterdam. Van april 2002 tot februari 2004 heeft zij als junior onderzoeker de gelegenheid gekregen een gerandomiseerd onderzoek naar het effect van de in dit proefschrift beschreven cognitief gedragsmatige training voor adolescenten met chronische pijn uit te voeren. Deze aanstelling omvatte eveneens het doceren en ontwikkelen van onderwijs voor geneeskunde studenten. Sinds februari 2004 zijn haar onderwijstaken bij de afdeling Medische Psychologie en Psychotherapie verder uitgebreid.

Vivian is lid van de Bestuurswerkgroep Richtlijnen van de sectie Revalidatie (NIP). Deze werkgroep zet zich in voor het ontwikkelen van richtlijnen op een aantal verschillende gebieden van de revalidatiepsychologie. Vanaf januari 2005 zal zij een klinische rol vervullen bij de afdeling Revalidatie en Psychologie van het Jan van Breemen Instituut te Amsterdam. Deze aanstelling is een onderdeel van de 2-jarige post-doctorale opleiding tot Gezondheidszorg (GZ) Psycholoog, centrale RINO groep - Amsterdam.

´Hm´, mompelde Tom Poes. Hij begon zo langzamerhand een idee te krijgen, en hij besloot nog een beetje verder te praten. ´Maar als iemand nu eens álles wist wat er te weten is´, zei hij. ´Wat zou er dan gebeuren?´ ´Dan zou de arme stakker exploderen´, zei de geleerde met een mekkerend lachje. Álles weten is onmogelijk geworden – en heeft trouwens ook geen zin´.
´Daar gaat het hem nou net om´, zie Tom Poes.
´Iemand, die álles weet wat er te weten is, heeft veel kennis´, vervolgde professor Sickbock, die schik in het onderwerp begon te krijgen. ´Maar waarom zou hij alles willen weten? Kennis zonder doelstelling is eigenlijk niet-kennis. Kunt ge me volgen?´

De Weetmuts, Maarten Toonder (In: Daar zit iets achter, 1980)