

Pain in Juvenile Idiopathic Arthritis: Parents and children as agents of disease management

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Abstract

Juvenile idiopathic arthritis (JIA) is a severe childhood disease usually characterized by long-term morbidity, unpredictable course, pain, and limitations in daily activities and social participation. The disease affects not only the child but also the whole family. The family is expected to adhere to an often very laborious regimen over a long period of time. However, the parental role is incoherently conceptualized in the research field.

Pain in JIA is of somatic origin, but psychosocial factors, such as mood and self-efficacy, are critical in the perception of pain and in its impact on functioning. This study examined the factors correlating and possibly explaining pain in JIA, with a special emphasis on the mutual relations between parent- and patient-driven variables.

In this patient series pain was not associated with the disease activity. The degree of pain was on average fairly low in children with JIA. When the children were clustered according to age, anxiety and depression, four distinguishable cluster groups significantly associated with pain emerged. One of the groups was described by concept vulnerability because of unfavorable variable associations. Parental depressive and anxiety symptoms accompanied by illness management had a predictive power in discriminating groups of children with varying distress levels. The parent's and child's perception of a child's functional capability, distress, and somatic self-efficacy had independent explanatory power predicting the child's pain.

Of special interest in the current study was self-efficacy, which refers to the belief of an individual that he/she has the ability to engage in the behavior required for tackling the disease. In children with JIA, strong self-efficacy was related to lower levels of pain, depressive symptoms and trait anxiety. This suggests strengthening a child's sense of self-efficacy, when helping the child to cope with his or her disease.

Pain experienced by a child with JIA needs to be viewed in a multidimensional bio-psycho-social context that covers biological, environmental and cognitive behavioral mechanisms. The relations between the parent-child variables are complex and affect pain both directly and indirectly. Developing pain-treatment modalities that recognize the family as a system is also warranted.

Tiivistelmä

Lastenreuma on pitkäaikainen ja vakava lapsuus- ja nuoruusiän sairaus, jonka kulkua on lapsen sairastuessa vaikea ennustaa. Lastenreuma voi vaikuttaa lapsen ja nuoren toimintakykyyn, se voi rajoittaa lapsen osallistumista arkipäivän toimintoihin sekä lapsen ja perheen sosiaalisia suhteita. Myös kipu liittyy lastenreumaan. Sairauden hoidossa ja kuntoutuksessa koko perheeltä, erityisesti vanhemmilta edellytetään pitkäaikaista sitoutumista. Kuitenkin vanhempien rooli on kuntoutuksessa ja hoidossa toistaiseksi jäänyt melko vähälle huomiolle. Lastenreumaan liittyvässä kivussa on usein tautiin liittyvä alkuperä. Kuitenkin psyykkis-sosiaaliset tekijät, kuten masentuneisuus, pystyvyyden tunne ja vanhempien kyky tukea lasta sairauteen liittyvissä haasteissa vaikuttavat ratkaisevasti, millaisen merkityksen kipu saa lapsen ja perheen elämässä, ja muodostuuko kivusta haittaava pitkittyvä oire. Näin ollen psyykkis-sosiaalisten tekijöiden tutkiminen lastenreumassa on teoreettisesti ja kliinisesti tärkeää. Tässä väitöskirjassa selvitetään, mikä selittää lastenreumaa sairastavien lasten ja nuorten kipua, sekä kuinka vanhemmuuteen ja lapseen liittyvät tekijät ovat keskenään yhteydessä lastenreumaan liittyvän kivun selittäjinä.

Tutkitussa aineistossa kipuoireita oli melko vähän, eikä kipu ollut yhteydessä tautiaktiivisuuteen. Lastenreumaa sairastavissa lapsissa ja nuorissa on kuitenkin iän, masentuneisuus- ja ahdistuneisuusoireiden perusteella toisistaan eroavia alaryhmiä. Erityisesti yhdellä näistä ryhmistä havaittiin sellaisia muuttujien välisiä yhteyksiä, joita kuvataan haavoittuvuuden käsitteellä. Tällä ryhmällä myös kipuoireita oli muita ryhmiä enemmän. Lisäksi vanhempien hyvinvointiin ja sairauden kanssa selviämiseen liittyvät tekijät olivat yhteydessä lapsen haavoittuvuuteen. Itsenäisesti lapsen kipua ennustivat tekijät ”lapsen ja vanhemman arvio lapsen toimintakyvystä”, ”lapsen psyykinen pärjäävyys” sekä ”lapsen pystyvyyskäsitys taudin somaattisten tekijöiden osalta”. Pystyvyyskäsitys tarkoittaa henkilön luottamusta pystyvyyteensä sairauteen liittyvissä somaattisissa, psyykkisissä ja sosiaalisissa haasteissa.

Yhteydet lapsen kivun sekä vanhemman ja lapsen psyykkisen voinnin ja pystyvyyskäsityksen välillä ovat suoria ja epäsuoria. Lastenreumaan liittyvää kipua voidaan ymmärtää käyttäen apuna bio-psyko-sosiaalista mallia, jossa kivun luonne tulee huomioitua somaattisena, psyykkisenä ja ympäröiviin sosiaalisiin suhteisiin liittyvänä tekijänä. Kokonaisuudessaan tutkimus osoitti sekä lapsen että vanhemman psyykkisen voinnin ja pystyvyyden tunteen olevan merkityksellisiä tekijöitä kuntoutuksen suunnittelun ja kipuoireiden kroonistumisen kannalta. Kuntoutuksessa perhe ja sen sisäiset vuorovaikutussuhteet tulee ottaa huomioon.

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On Mother's Day

9.5.2010

List of original publications

- I Vuorimaa, H., Honkanen, V., Konttinen, Y. T. , Komulainen, E., & Santavirta, N. (2007). Improved Factor Structure for Self-efficacy Scales for Children with JIA (CASE) and Their Parents (PASE). *Clinical and Experimental Rheumatology*, 25, 494–501.
- II Vuorimaa, H., Tamm, K., Honkanen, V., Konttinen, Y. T., Komulainen, E., & Santavirta, N. (2008). Empirical classification of children with JIA: a multidimensional approach to pain and well-being. *Clinical and Experimental Rheumatology*, 26, 954–961.
- III Vuorimaa, H., Tamm, K., Honkanen, V., Komulainen, E., Konttinen, Y. T., & Santavirta, N. (2009). Parents and children as agents of disease management in JIA. *Child: Care Health and Development*, 35, 578–585.
- IV Vuorimaa, H., Tamm, K., Honkanen, V., Komulainen, E., Konttinen, Y. T., & Santavirta, N. Pain in JIA: a family matter. *Children`s Health Care*. In print.

Some unpublished results are presented (V).

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Abbreviations

ANOVA	one way analyses of variance
BDI	Beck's depression inventory
CASE	arthritis self-efficacy scale for children
CASEsom	self-efficacy with somatic symptoms
CASEpsy	self-efficacy in psychological functioning
CASEsoc	self-efficacy in social functioning
CBCL	child behaviour checklist
CDI	child depression inventory
CHAQ	childhood health assessment questionnaire
CFA	confirmatory factor analyses
CFI	comparative fit index
CSQ	coping self-efficacy questionnaire
DMAR	disease modifying antirheumatic drug
EFA	exploratory factor analyses
EM	expectation maximization method
HADS	hospital anxiety and depression scale
HADS-A	hospital anxiety and depression scale-anxiety subscale
HADS-D	hospital anxiety and depression scale-depression subscale
ILAR	International League Against Rheumatism
JIA	Juvenile idiopathic arthritis
JRA	juvenile rheumatoid arthritis
LDA	descriptive linear discriminant analyses
MANOVA	multivariate analyses of variance
MAR	missing at random
MASC	multidimensional anxiety scale for children
VAS	visual analogue scale
PASE	parental self-efficacy scale
PASEsom	parental self-efficacy with somatic symptoms
PASEpsy	parental self-efficacy in psychological functioning
PASEsoc	parental self-efficacy in social functioning
RASE	rheumatoid arthritis self-efficacy scale

RCADS	revised child anxiety and depression scale
RCMAS	revised children's manifest anxiety scale
RF	rheumatoid factor
RMSEA	root mean square error of approximation
STAI	state-trait anxiety inventory
STAIC	state-trait anxiety inventory for children
SPQ	structured pain questionnaire
VAS	visual analogue scale

1 Introduction

The main purpose of this study is to identify the factors that explain the variance of pain among children with juvenile idiopathic arthritis (JIA). JIA is a chronic rheumatic disease. Its incidence in Finland is 20–23/100,000 of the child population (Berntson et al., 2003; Kaipianen-Seppänen & Savolainen, 2001). Its medical management has become more aggressive in recent years, which has reduced the disability associated with it (Goldmuntz & White, 2006; Minden et al., 2002). Despite the major advances in the treatment however, JIA is still a severe disease because of its long-term morbidity, unpredictable course, chronic pain, and potential limitations in daily activities and social participation. Pain in JIA can be attributed partly to organic causes and partly to psychosocial factors. However, in addition to asking whether the pain is organic or functional, it would be useful to identify the factors that are likely to maintain or increase it. Psychosocial factors play an important role in chronic pain (Casey et al., 2008). According to several sources, they are critical in terms of the perception of pain and of its impact on functioning (Anthony & Gil, 2003; Anthony & Schanberg, 2003; Logan & Scharff, 2005; Malleson & Clinch, 2003; Packham et al., 2002; Schanberg et al., 1997; Schanberg et al., 2003; Schanberg et al., 2000; Thastum et al., 1997; Thompson et al., 1987; Varni et al., 1988).

In clinical practice, an obvious question is, how to distinguish children and adolescents that are more vulnerable than others to pain symptoms, and those who manage aspects of the illness with less resilience than others. Vulnerability is multifactorial in a sense that many disease-related, but also psychological, and social factors take the child into lower level of functioning or more pain symptoms. In the current thesis these questions are tackled by investigating critical variables reflecting this vulnerability. In pain research bio-psycho-social -definition of pain is generally applied (Turk, 1988; Waddell et al., 1984; Waddell, 2004). It explains how biological, environmental, and cognitive behavioural mechanism develop and maintain pain. Pain has also social aspects. Thus, the parents as the most important agents in the environment of the child were included in the analyses of the current thesis. Another important background for the study was the family system theory (Kerr, 1988) that underlines interdependency of child and his/her parent. Of special interest in the current study is the concept of arthritis self-efficacy (Barlow et al., 2000; Barlow et al., 2001), which refers to the belief of an individual that he/she has the capability to manage JIA. Self-efficacy beliefs are considered essential, as they predict current behaviour and can be used as a mechanism of change or improvement in rehabilitation (Rhee et al., 2000; Turner et al., 2007). So far, research on arthritis self-efficacy in children has been scarce, and parents have not usually been included in the studies.

1.1 Juvenile idiopathic arthritis

Juvenile idiopathic arthritis (JIA) is one of the most common rheumatic diseases of childhood (Goldmuntz et al., 2006). JIA is defined as the presence of objective signs of arthritis (swelling of the joint or two or more of the following: limitation of motion, tenderness, pain with motion or joint warmth) in at least one joint for more than six weeks in a child younger than the age of 16 after other types of childhood arthritis have been excluded. A new classification (Table 1) of juvenile arthritis was developed by the International League Against Rheumatism (ILAR) and is currently used worldwide, replacing the previous American classification of juvenile rheumatoid arthritis (JRA).

Table 1. Subtypes of juvenile idiopathic arthritis (JIA) based on the second revision of International League of Associations for Rheumatology (ILAR) criteria.

Subtype and subcategory	Inclusion criteria
1. Oligoarthritis • persistent • extended	<ul style="list-style-type: none"> Arthritis in 1–4 joints during the first six months 1a. affects less than 5 joints throughout disease 1b. affects 5 or more joints after the first six months
2. Polyarthritis 2. Seronegative 3. Seropositive	<ul style="list-style-type: none"> Arthritis in five or more 5 joints during the first six months RF-negative RF-positive
4. Systemic arthritis	<ul style="list-style-type: none"> Arthritis in one or more 1 joints with or prior to 2-week fever and at least one of following: Erythematous nonfixed rash Generalized lymph node enlargement Hepato- and/or splenomegaly Serositis
5. Entesitis related arthritis (ERA)	<ul style="list-style-type: none"> Arthritis and entesitis, or arthritis or entesitis with at least 2 of following: Present or history of SI tenderness and/or inflammatory lumbosacral pain Presence of HLA-B27 Onset of arthritis in males aged at least 6 years Acute symptomatic anterior uveitis History of AS, ERA, sacroiliitis with IBD, Reiter’s syndrome, or acute anterior uveitis in 1st degree relative
6. Psoriatic arthritis	<ul style="list-style-type: none"> Arthritis and psoriasis, or arthritis and at least 2 of following: Dactylitis Nail pitting or onycholysis Psoriasis in 1st degree relative
7. Undifferentiated arthritis	<ul style="list-style-type: none"> Arthritis which does not fulfill the criteria for any of the other categories or fulfills the criteria for more than one of the other categories

Modified from Petty et al. 2004.

Abbreviations: SI=Sacroiliac, IBD=inflammatory bowel disease, HLAB27=human leukocyte antigen.

The cause of JIA is unknown, but there is substantial evidence to suggest that the pathogenesis is autoimmune (Anthony & Schanberg, 2003). Diagnosis is a matter of exclusion, and is made on the basis of a combination of clinical and laboratory data. The course of the disease is highly variable, and in the past it has been viewed as a more benign illness than current perceptions suggest. Recent data reveal that a substantial number of children diagnosed with JIA have an active disease that persists into adulthood and may result in functioning limitation. The course of arthritis is unpredictable regardless of the type, but it most commonly fluctuates and is characterized by periods of flare and quiescence.

Because JIA is a chronic condition, it is important to make distinction between the concept “disease” and “illness”. “Disease” is defined as an objective biological event involving the disruption of specific body structures or organ system caused by either anatomical, pathological or physiological changes. “Illness”, in contrast refers to subjective experience that a disease is present, and how a sick person and the members of his/her family live, and respond to, symptoms of disability (Gatchel et al., 2007). Due to the fact that both disease- and illness- aspects are present in JIA, it is particularly important to have an integrative view when studying and treating it.

Due to chronic nature of JIA with the potential for significant morbidity, the pharmacological treatment of articular manifestations has become more aggressive and is initiated at an earlier stage than before. The goal in the Finnish JIA treatment algorithm is always remission. Once the diagnosis has been confirmed the patient receives intra-articular glucocorticoid injections to the active joints. In cases of polyarthritis, or if the intra-articular injection-induced remission tails off in those with oligoarticular disease, the patient is started on a disease-modifying anti-rheumatic drug, usually methotrexate. If remission is not achieved in six months despite repeated intra-articular injections and the disease-modifying drug, a biological drug is considered. Methotrexate and, more recently, biologics have produced substantial improvements in the disease control, particularly in patients with polyarticular JIA (Lovell, 2006; Tynjälä et al., 2006).

Considering long-term outcome, half of the JIA patients had distinctive changes in body function or structure after disease duration of less than 15 years (Minden et al., 2002). Oen et al. (2002) showed that the probability of active disease continuing into the late twenties or early thirties was high for patients who were not in remission by the age of sixteen. The functional outcome has improved, but in the patients with systemic and RF+ polyarticular onset, the disability may still develop in a significant number of patients (Oen et al. 2002). Hyrich et al. (2010) showed recently that although the majority of children have a significant improvement in pain due to steroid injections or DMARDs, one-third of children continued to have moderate to severe levels of disability and joint inflammation

and disability after one year (Hyrich et al., 2010). In Foster et al. (2003) the effect of JIA on quality of life in young adults was remarkable with poor self-perceived health status, and despite excellent educational attainment there were high rates of unemployment.

A Finnish prospective study (Arkela-Kautiainen et al., 2005) on childhood-onset arthritis showed that the health-related quality of life and social functioning of early-adult JIA patients was as good as in matched controls. Nor was there a significant difference in educational level between the groups. However, physical functioning in the patient group was significantly poorer than in the control group at the mean age of 23 years. 37 per cent of the patients were in remission. Although the outcome of the cohort was good, the extended oligoarthritis group exhibited a pattern of low health-related quality of life, and low physical and mental functioning.

1.2 Psychological aspects of juvenile idiopathic arthritis

Children with a chronic illness such as JIA face an increased risk of behavioural and emotional difficulties. According to Packham (2004), the major psychological difference between adult-onset inflammatory arthritis and JIA is that coping strategies are not fully developed in childhood, and adolescence and the chronic disease have to be negotiated at the same time. Children with rheumatic disease and their parents are usually asked to adhere consistently and over a long period of time to a variety of therapeutic regimens, including medication, therapeutic exercises and splinting the joints. Some may fail to understand the delayed beneficial effects of these regimens (Rapoff, 2001), and others believe that the side-effects of the medication outweigh the potential benefits and therefore are reluctant to take the medicine. Adherence to the regime is one of the major issues that may lead to a favourable outcome with JIA (Feldman et al., 2007), and it is particularly important with the new expensive pharmacological treatments. April et al. (2006) and Feldman et al. (2007) found that the factor most highly associated with perceived adherence was belief that the treatment was helpful.

Horne (2006) proposes that patients respond to illness in a dynamic way based on their evaluation of it. According to him, people do not blindly follow health advice, but rather people tend to evaluate whether the advice makes “common sense” in light of their own perceptions of the illness. First, this evaluation takes into account health threats, which can be considered as a first response to build a mental map, an illness representation that enables a person to make sense of the threat. Illness representation answers questions about illness: what it is, how long it will last, what causes it, how it will affect me, and can it be controlled. It has a cognitive and an emotional component, also, for instance believing that a flare is untreatable may cause anxiety etc. Second, symptom experiences

and labels are associated with the illness evaluation of a person. For instance failure to achieve a convincing diagnostic label to match symptom experiences may increase anxiety and lead to repeated consultations. Third, when constructing illness representation, a person tries to achieve common-sense coherence. For instance, an initial belief that the cause of pain is flare-up of the disease may prevent perceptions of the best way to cure it. Accepting that there may be psychological aspects in the condition may be difficult. The illness representations influence action. This fourth argument in Horne's (2006) theory can be illustrated in such a manner that actions that are not congruent with patients' illness-representations are not easily adopted. For instance, cutting of antirheumatic medication in case of pain symptoms may be unacceptable for patients with an illness-representation that he/she has a flare-up of the disease. Following this, Horne (2006) claims that beliefs and behaviour interact in a dynamic process, and that emotional process occurs in parallel with cognitive process. For instance, an appraisal of a family that the child has a flare, although this cannot be proven by laboratory tests, might induce anxiety. Moreover, seeking for second opinion in order to have symptoms and findings diagnosed in such terms that the family can accept may take place.

Juvenile idiopathic arthritis may force the child, and also the family, into phases of disturbed family rhythms (e.g., pain variation), dependency (e.g., the need for parents due to joint stiffness), feelings of ambivalence (e.g., participation restriction, difficulties with school attendance) and uncertainty about the future (Sällfors et al., 2002). Participation in school and leisure activities may be limited, and the children face barriers (e.g., lack of understanding by teachers, high steps into buildings) that may limit choice, opportunities for social interaction, and the process of becoming independent of parents (LeBovidge et al., 2003; Packham, 2004).

Adolescence as a period of change may be more challenging to individuals with JIA than to healthy children. Physical limitations may restrict the ability to gain independence from parents and develop a self-concept. Participation in socialising activities may be limited, which in turn may inhibit independence and social maturation (Barlow et al., 1997). Most children and adolescents with JIA perceive themselves as socially competent (Huygen et al., 2000; Noll et al., 2000), although some studies show that children have difficulties adapting to JIA and have a strong sense that their emotional state is negatively affected by the disease (Packham, 2004; Packham et al., 2002).

JIA may have significant implications for the family structure and dynamics (Packham et al., 2002). A stable family environment and effective family support for a child coping with the disease greatly influences child's adaptation to it. Parents need to develop resilient traits such as balancing the illness and family needs, maintaining flexibility in the family unit, remaining socially integrated, maintaining clear family boundaries, and developing collaborative rather than dependent relationships with health

professionals (Packham et al. 2002). According to Barlow et al.(2002) parents express concern and anxiety about the impact of JIA upon their child’s development and future. Another dilemma facing parents is the medication. Although it is considered necessary for treating the symptoms and the disease, it may cause parental concern because of the side-effects (Barlow et al., 2002) (e.g., stunted growth and gastrointestinal irritation).

The recent trend in JIA rehabilitation and care is to involve parents and patients fully in the decisions about treatment. There is also a growing interest in coaching the parents and the child to be “experts” in this active patient /parent role (Barlow & Ellard, 2004).

1.3 A definition of pain

The International Association for the Study of Pain offers the following definition: pain is an unpleasant sensory and emotional experience associated with actual or potential tissue damage or described in terms of such damage. The statement includes a note that pain is always subjective, and each individual learns the application of the word through experiences related to injury in early life (Merskey, 1986). In the revised pain definition (Merskey, 1994) the following content have been included: “many people report pain in the absence of tissue damage or any likely pathophysiological cause; usually this happens for psychological reasons. There is usually no way to distinguish their experience from that due to tissue damage if we take the subjective report. If they regard their experience as pain and if they report it in the same ways as pain caused by tissue damage, it should be accepted as pain. This definition avoids tying pain to the stimulus. Activity induced in the nociceptor and nociceptive pathways by a noxious stimulus is not pain, which is always a psychological state, even though we may well appreciate that pain most often has a proximate physical cause” (Merskey, 1994). Pain is also the sensory modality that alerts patients so that it leads to protective responses. It can be described in terms of its intensity, affective component and location. It could also be described in terms of its duration, in other words as acute, sub-acute or chronic (Linton, 2004). Acute pain is defined as pain lasting up to three weeks. Pain is called sub-acute when it is considered to last three to 12 weeks, and chronic or persistent when lasting over three months (Linton, 2004).

Chronic pain can also be defined as prolonged and persistent pain of at least three months and chronic recurrent pain as recurrent episodes of pain. Acute pain is of variable duration, typically characterised by sudden onset: such as tissue-damaging stimulation produced by disease and a limited and predictable course (American Academy of Paediatrics and American Pain Society, 2001). Chronic pain is defined as repeated or continuous episodes of pain that are experienced either as

a component of a well-characterised medical disorder or by an otherwise healthy individual in the absence of a well-defined organic aetiology (American Pain Society, 2001).

In general, pain symptoms during childhood are fairly common. In a population-based study El-Metwally et al. (2004) showed recently that of Finnish schoolchildren 32 per cent had musculoskeletal pain, and at one year-follow-up 54 per cent of them had still musculoskeletal pain. Of those, 64 per cent still had pain symptoms at the 4-year follow-up. The authors discussed that pain symptoms are not as transient and self-limiting as previously assumed and that persistence of musculoskeletal pain in preadolescence was strongly associated with pain recurrence in adolescence (El-Metwally et al., 2004). Hakala et al. (2002) have suggested in a Finnish biennial nationwide survey and annual classroom surveys, that pain in neck, shoulder and lower back are becoming more common in Finland in age group of 12–18 years.

1.4 Pain in JIA

Several studies suggest that pain is more prevalent in JIA than previously recognised, and may persist in some patients into adulthood (Flato et al., 2003). Schanberg et al. (2003), who used daily-diary methodology, showed that children with chronic arthritis reported pain on an average of 73 per cent of days. Various studies on children with JIA report pain intensity in moderate range, averaging about 3 on a 0–10 mm VAS-scale (Schanberg et al., 2000; Schanberg et al., 2003; Sällfors et al., 2004; Malleon et al., 2004; Shaw et al., 2006a, 2006b).

No single unifying etiological explanation of why pain changes from acute to sub-acute, to sub-chronic and to chronic in children with JIA has been found. Inherent in the disease are precipitants that characterise acute pain, such as joint inflammation. However, there is a consensus that such physical variables do not explain all the variance in pain experiences in children with JIA (e.g. Anthony et al., 2003). Even when there are organic causes (such as arthritis), psychosocial factors are critical in the perception of pain and its impact on functioning.

Gatchel et al. (2007) provides a distinction between nociception and pain. Nociception involves the stimulation of nerves that convey information about potential tissue damage to the brain, whereas pain is a subjective perception that results from the transduction, transmission, and modulation of sensory information (Gatchel et al., 2007). Thus pain is the subjective response of an individual to noxious stimuli. According to the psychobiological argument put forward by Zeltzer et al. (1997), the noxious stimulus is sensed by a peripheral receptor, and a noxious sensory message is then transmitted via afferent fibres to the spinal cord. After entering the central nervous system the signal is passed up ascending tracts in the spinal cord to synapse in the specific areas of the midbrain, pons

and diencephalon. At any point along the path reflex arcs might be stimulated to initiate aversion responses to the noxious event. From these lower brain centres, the nociceptive information might or might not be forwarded to the limbic and somatosensory areas of the telencephalon, where the subjective, conscious experience of pain originates (Zeltzer et al., 1997). The great variability in pain perception could be attributed to two mitigating categories of factors: physiological and psychological differences in individuals and differences in cross-talk between and among these systems (Zeltzer et al., 1997).

Some researchers have attempted to explain arthritis pain in children with physiological factors such as disease severity and pain threshold, whereas others have focused on psychosocial characteristics, such as anxiety of the child and/or illness management of the child by the parent. The scope of the research on juvenile pain could also be categorised as intrapersonal or interpersonal. The intrapersonal level includes investigation of physiological disease-related and demographic variables, whereas the interpersonal scope incorporates psychosocial aspects such as family, community and cultural contexts, and the social and psychological variables that affect pain experience and expression. The interpersonal differences between individuals are strongly influential in the trajectory from acute inflammation-related pain to chronic pain.

Disease severity or activity as physical intrapersonal factors is associated with pain symptoms in JIA, as evidenced by Malleson et al. (2004), Schanberg et al. (2003), Schanberg et al. (1997) and Sällfors et al. (2003). However, studies on the relationship between pain intensity and disease activity have also produced mixed results. Ilowite et al. (1992) found that parents' and doctors' ratings of pain in children correlated with joint inflammation, but this correlation was observed only for self-reported pain in young children. On the other hand, researchers have found that disease severity predicts only a small-to-medium proportion of the variance in children's pain reports. Ilowite et al. (1992) demonstrated that joint inflammation explained only 10 per cent of the variance in pain scores, and found no significant relationship between the scores and disease subtype or the number of joints affected. Even smaller explanatory power was evidenced in Malleson's (2004) study, in which disease activity accounted only 6,5 per cent of pain variance.

Another physical intrapersonal factor that may alter the child's expression of pain is the pain threshold. The results of experimental studies have shown that patients with JIA have reduced pain tolerance and a lower pain threshold than healthy controls (Hogeweg et al., 1995a, 1995b), and that patients with JIA had a lower pressure-pain threshold in inflamed joints and in non-inflamed paraspinal areas, especially those associated with inflammation, when compared to other joints (Hogeweg et al., 1995a, 1995b). A lower pain threshold has also been detected in children with

JIA in remission, which means that the change in threshold may persist (Hogeweg et al., 1995a, 1995b; Kuis et al., 1997). Receptors sensitive to noxious stimuli generated as a consequence of inflammation have an important role in the pathogenesis of inflammatory pain becoming chronic. These receptors, called nociceptors, activate predominantly unmyelinated, small-diameter sensory nerves, mediating nociceptive information to higher-order neurones in the dorsal horn of the spinal medulla (Kuis et al., 1997).

The role of intrapersonal demographic variables, particularly age and gender, have also been examined as factors that discriminate between children with a higher risk of developing chronic or recurrent pain. It has been suggested that subjective pain reports depend, in part, on the child's cognitive capabilities, such that as children mature cognitively the meaning attributed to sensations in their inflamed joints changes, thereby influencing their perception (Beales et al., 1983). Children's expressions of pain are influenced by both age and the social context, as shown in experimental research conducted by Zeman & Garber (1996): the presence of peers reduced pain expression, and that of mothers increased it. Some studies have shown that age is a statistically significant predictor of pain in JIA (Hagglund et al., 1995), and others that it is not (Schanberg et al., 1997). Adolescents tend to report more pain in JIA (Shaw et al., 2006). Yet, age has not been a major element in pain research in general in recent years, or in pain research within JIA. There are also conflicting results concerning the impact of gender on the pain reports of children with JIA, with some studies showing no differences between boys and girls and others indicating more severe daily pain in girls (Schanberg et al., 2001; Sällfors et al., 2003).

Interpersonal factors may discriminate between children with a higher risk of developing chronic pain. Neither prospective nor experimental studies have confirmed that psychosocial factors, such as depression of the parent, directly influence pain in JIA. However, when there is no medical explanation, social and psychological variables that affect the child's experience and expression of pain assume importance. Craig et al. (1997) and Craig (2002) put forward a model suggesting that any given childhood pain is likely to involve a sequence of events including tissue trauma, the experience and expression of pain, caregivers' observations and assessment, and subsequent interventions, all of which are potential targets for social influence. Craig (2002) suggests that the role of social factors is significant in children, specifically young children because they have fewer self-management skills than adults.

The aim in the field of pain research since the 1970s has been to integrate biomedical, psychosocial and behavioural data into one coherent model (Turk et al., 1988; Waddell et al., 1984, 2004) (Figure 1). It is now well recognized that pain is multidimensional, and that it is best understood within the

context of a bio-psycho-social model that covers biological, environmental and cognitive behavioural mechanisms in the development and maintenance of pain. Several such models have been proposed with the idea of including nociception within a set of concentric “shells” incorporating suffering, pain behaviour, and social context. According to Gatchel (2007) any model that focuses on only one of dimensions such as nociception, pain suffering, and pain behaviour, will be incomplete and inadequate.

The biopsychosocial conceptualisation of pain is applicable in a population of JIA patients in that the pain could be viewed as acute and chronic. There are few prospective studies focusing on the transition from acute to chronic pain and disability. One recent study conducted by Casey et al. (2008) on adult-patients with acute neck or back pain showed that baseline depressive symptoms and belief in pain permanence were the most powerful predictors of chronic disability. Baseline pain did not predict follow-up chronic pain, and baseline acute pain intensity did not directly predict three-month disability, but it was indirectly related to disability through belief in pain permanence. It was concluded that the transition to chronic pain syndromes was a function of serious life stressors and cognitive factors more than medical factors.

In sum, it seems that pain in children and adolescents with JIA should be viewed as a dynamic, non-static factor on which developmental factors and the social contexts in which children live their lives have a substantial impact. Individual variation in terms of physiological, demographic and psychosocial factors produces different pain outcomes.

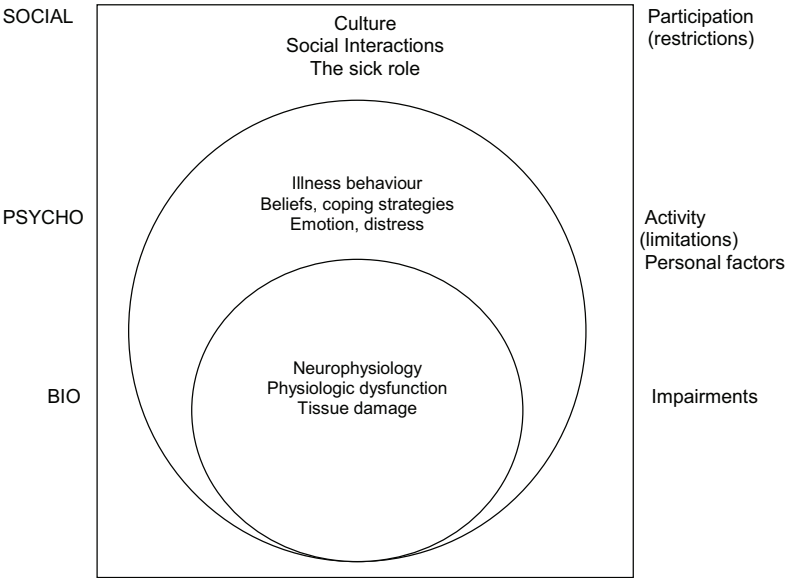


Figure 1. A biopsychosocial model of pain. Modified from Waddell (2004).

1.5 Pain-associated variables

Several studies have demonstrated the role of many psychosocial variables in children's pain reporting. Pain in children with JIA has been studied from several perspectives, and the emphasis has been shifting from single child variables towards interpersonal variables such as parental well-being as possible influencing factors. The biopsychosocial model of pain defines aspect that might affect on the child's and the parents beliefs, emotions etc. that restrict participation of the child, might enhance a sick role, and might make the family system vulnerable.

1.5.1 Depressive symptoms in the child

The term depression could refer to a single symptom, a symptom cluster, or a disorder. Features distinguishing normal sadness from abnormal depression include severity, persistence, and quality of the mood. Depression is a clinical symptom characterised by emotional distress, negative thinking, motivational deficits and vegetative symptoms (Main et al., 2008), reduction in or loss of the ability to experience pleasure, low self-esteem, and guilt, feelings of helplessness and hopelessness, suicidal thoughts, loss of energy, poor concentration, restlessness, and changes in appetite, weight and sleep. It could be considered a normal human emotion of variable intensity, but if sufficiently severe it is better understood as a major mental illness (Main et al., 2008). A depressed mood could also be considered a normal reaction to events that are characterised by prolonged stress or loss of independence. Of Finnish schoolchildren aged 14–16 years 22 per cent was demonstrated to have some symptoms of depression, and three percent severe depression (Ellonen et al., 2008).

Studies on psychological problems in children with JIA give conflicting results. Some report more adjustment difficulties in JIA patients than in healthy comparisons (Aasland et al., 1997; LeBovidge et. al, 2003; Sällfors et al., 2004; VanDyke et al., 2004) whereas others find no signs of psychosocial disturbance and no difference between children with JIA and healthy peers (Evers et al., 2002; Huygen et al., 2000; Schanberg et al., 2000; Schanberg et al., 2003). Schanberg (2000) investigated daily variation in mood and stressful events among children with rheumatic diseases, finding that none of them scored in the clinically significant range on the Children's Depression Inventory, but there was variability in daily mood: a negative mood predicted increased reports of pain. Packham et al. (2002) found in a long-term follow-up study that of the patients as adults with childhood onset JIA 38 per cent felt that their emotional state had been negatively affected by the disease and 26 per cent felt that arthritis had had a severe detrimental effect on their emotional state. Furthermore, depression was most commonly manifest at an adult age when the age at the onset of

JIA was between six and 12 years, compared to earlier or later onset. Shaw et al. (2006a) showed that in a JIA sample, of the age group of 17-year-olds two-thirds reported depressive symptoms, which was clearly more than in the younger age groups.

Several studies on pain in JIA include depressive and anxiety symptoms in the core set of explanatory variables. Hoff et al. (2005) studied longitudinal relationships between depressive symptoms and both pain intensity and functional disability among children with disease-related pain. He found that the extent of the association depended partly on the individual's history of pain and disability. Evers et al. (2002) further proposed that early rheumatoid arthritis did not in itself predict anxiety/depression at an adult age, but that a lack of health-enhancing behaviour and perceptions of control accounted for the relationship between the physical symptoms and future distress. Thastum et al. (2005) found that psychological factors were strongly influential in explaining pain in JIA, and that being in control of the pain despite the condition was the key factor in its prediction. Schanberg et al. (1997) found that children who rated highly their ability to control pain had lower pain intensity. Malleson et al. (2004) claimed that pain cannot be explained simply in terms of disease-related variables.

A fundamental question remains. Do depressive symptoms in JIA lead to depressive actions such as changes in the child's management of the disease and in its life in general? Some studies suggest that this is the interactive mechanism that predicts pain. Kashikar-Zuck et al. (2000, 2001) found in their studies on chronic pain in juvenile patients that maladaptive coping was associated with depressive symptoms, which in turn were associated with the child's disability. However, maladaptive coping was not independently associated with functional disability. This suggests that depressive symptoms function as a mediator between disability and maladaptive coping. Interestingly there was no relation between pain intensity and depression, but the association between depression and functional disability was strong. Depressive symptoms as such did not predict pain, although children with high levels of depressive symptoms tended to cope poorly (e.g. internalising and catastrophizing) and to be the most disabled by their symptoms (Kashikar-Zuck et al., 2001). Furthermore, a recent study conducted by Casey et al. (2008) indicates that depressive symptoms and low controllability beliefs could lead to passive coping and avoidance, thereby exacerbating the disability. Depression and pain are mutual risk factors, although research results support the notion that chronic pain is a precursor of depression more strongly than vice versa (Main et al. 2008).

In conclusion, it seems that depression is not a central feature of JIA in general. Some children do have depressive symptoms, which persist in some of them until adulthood. Psychopathology is not a good enough explanation for altered mood in children with JIA, rather it might be a question of

comorbidity of symptoms due to often complex medical treatment regimen. LeBovidge et al. (2003) have demonstrated in a meta-analytic review that adjustment difficulties, particularly internalising symptoms are common in children with JIA. It should also be borne in mind that depressive symptoms are not separate from the cognitive functioning of children or parents: children with JIA, who demonstrate psychological comorbidity, may be at long-term risk for cognitive dysfunction with respect to perception of functional ability (Peterson et al., 1997). After all, according to a population based study (Härmä et al., 2002) recurrent pain symptoms are associated with depression among adolescents.

1.5.2 Anxiety symptoms in the child

Anxiety falls into two categories in terms of content. State-anxiety reflects transitory anxiety states, subjective, consciously perceived feelings of apprehension, tension and worry that vary in intensity and fluctuate over time (Spielberger et al., 1983; Spielberger et al., 1973). Trait-anxiety, on the other hand, refers to relatively stable individual differences in anxiety proneness and the tendency to experience anxiety states. Generalized anxiety disorder is characterized by excessive and uncontrollable worry about a number of life events or activities, and by its extension over a long period of time (Main et al., 2008). Anxiety is a common feature among patients with pain, particularly when they have not been given a clear explanation for it (Main et al., 2008). In JIA, anxiety may be better viewed as a pain-associated psychological dysfunction rather than as a primary psychiatric disorder (Main et al., 2008). However, it may be advantageous to identify children with JIA and a high level of trait anxiety given the frequently observed temporal sequence of anxiety followed by depression (van Dyke et al., 2004).

David et al. (1994) reported that anxious and helpless responses were more common in patients whose arthritis started in adolescence than among those with later or earlier onset, whereas Aasland et al. (1997) found that 17 per cent of 52 adult JIA patients had a psychiatric disorder, often anxiety, but none had a depressive disorder. Packham et al. (2002) reported an incidence of 32 per cent for anxiety and five per cent for depression in 246 adult JIA patients. Those in the late-onset group, at over 12 years of age, were at the highest risk of developing anxiety-related problems compared with the mid- and early-onset groups. Packham (2002) posited that early onset of the disease could have a later effect on the effectiveness of learned coping strategies.

Schanberg et al. (2000, 2003) conducted studies in which children with JIA filled in daily diaries, and found that day-to-day fluctuations in mood and stressful life events were related to daily symptoms. A worse mood and more stressful events were significantly related to increased daily pain, fatigue

and stiffness, and a lower ability to control and decrease pain. Anxiety rather than depression may be a clinically significant problem for many children with JIA, given its significant association with increased pain frequency, pain intensity and fatigue.

There may be substantial overlap between anxiety and depression in JIA and in other chronic childhood illnesses. For instance, van Dyke et al. (2004) found in their study that anxiety and depression shared 69 per cent of the variance, whereas Seligman et al. (2004) reported a 25-per-cent overlap on the STAIC trait-anxiety scale with the Children's Depression Inventory CDI scale (Kovacs, 1985). An important aspect often studied in association with anxiety is catastrophizing. (Sullivan et al., 2001). Pain catastrophizing is defined by Sullivan (2001) as "an exaggerated negative mental set brought to bear during actual or anticipated pain experience". Recently, Veervoort et al. (2010) studied schoolchildren's catastrophic thinking about their pain by a hierarchical regression analysis. One of the special interests was, whether trait anxiety accounts for the effect of catastrophizing. It was found that the child's pain catastrophizing at the baseline had a small contribution to the prediction of pain at the six-month follow-up. Trait-anxiety did not explain the variance of pain. Instead, anxious disposition was a precursor of catastrophizing in children in such a manner that catastrophizing children with higher levels of trait anxiety at baseline were more inclined to report higher levels of catastrophizing at the follow-up. It was discussed that catastrophic thinking may arise as a functions of predispositional factors such as trait-anxiety, and that children with high levels of trait anxiety might be particularly vulnerable to catastrophizing. Hermann et al. (2007) found positive correlation between pain-related catastrophizing and the amount of depressive symptoms in a sample comprised of juvenile patients and healthy controls. Moreover, the level of pain related catastrophizing was significantly positively correlated with trait anxiety and the frequency and intensity of pain episodes in their study. Interestingly, no significant correlation was distinguished between pain-related catastrophizing and other behavioural problems (Hermann et al., 2007).

Several new self-report instruments measuring symptoms of childhood anxiety have been generated. The Revised Child Anxiety and Depression Scale RCADS (Chorpita et al., 2000); the Revised Children's Manifest Anxiety Scale (RCMAS) (Reynolds & Richmond, 1985); the Multidimensional Anxiety Scale for Children (MASC) (March & Albano, 1997); and the Hospital anxiety and depression scale HADS-scale (Zigmond & Snaith, 1983) designed for use among adult patients with physical illness. There has been discussion about the discriminant validity of older measures such as the State-Trait Anxiety Inventory for Children (STAIC; Spielberger, 1973) and the State-Trait Anxiety Inventory (STAI; Spielberger, 1983) (Winters et al, 2002). Seligman, et al. (2004) found that the anxiety scales STAIC and STAI, discriminated between youth with

an anxiety disorder and those without. HADS-scale was used to measure parental anxiety state. Based on literature review of validity of HADS-scale, Bjelland et al. (2002) conclude that HADS perform well in assessing the symptom severity of anxiety and depressive disorders in both patient populations as well as in general population.

1.5.3 The role of the parents

Several determinants of childhood living conditions may have an impact on child's health later in life. Among these determinants parental mental problems are strongly associated with psychological distress in early adulthood (Kestilä et al. 2005). Mechanisms of effect and transition are complex. Two dominant theoretical models try to explain how child's pain and family factors may be considered as being connected to each other. One of them is an operant-behavioural theory, which is focused on the role of social reinforcement in maintaining maladaptive pain behaviours. The other is a family system theory, focused on family's overall functioning and role assignment (Palermo & Chambers, 2005). According to the family system theory (Charles, 2001; Cox & Paley, 1997; Kerr & Bowen, 1988; Kerr, 2010; VanEcke et al., 2006), which is the theoretical background for this thesis, families function as a system that makes the members interdependent. According to Bowen (Kerr & Bowen, 1988), people within family ought to connect emotionally to it, it is the very nature of a family. People solicit each other's attention, approval, and support and react to each other's needs, expectations and distress. The connectedness and reactivity makes the functioning of family members interdependent. The interdependency promotes cohesiveness and cooperation, which are evolved to protect, shelter, and feed the family members. According to Bowen theory (Kerr, 2010), heightened tension can intensify the processes that promote unity and teamwork, and this can lead to problems. For instance anxiety of one family member can escalate by spreading among the family. As anxiety goes up, the emotional connectedness of family members may become more stressful than comforting. As an example, Goubert et al. (2009) found that parents estimate their child's pain as higher when they catastrophize more about their child's pain. Chambers et al. (2002) showed that maternal behavior can have direct impact on their daughter's subjective reports of pain.

Parents are important agents of disease management, and they could take an active part in the rehabilitation process, but the main focus at the clinics is on the health state of the child. Thus, differences in parental coping ability and disease self-efficacy are not necessarily recognized, and vulnerable families may become overwhelmed when the parents are unable to successfully manage their child's disease. Research has produced conflicting results regarding parental degree of

psychosocial dysfunction due to JIA. According to some studies, only a minority of parents with a JIA child experience a significant degree of psychosocial disturbance. Gerhardt et al. (2003) found that families with experience of JIA were resilient and showed no severe signs of psychosocial dysfunction. Noll et al. (2000) and Barlow et al. (2000), on the other hand, found that mothers of children with JIA had more anxiety/depressive symptoms than mothers in the control group. Timko et al. (1992) reported that mothers were more depressed than fathers, and that families with sufficient resources were less prone to depression.

Both parent and child psychosocial factors may serve as a risk or supportive factor in either exacerbating or minimizing the child's functional disability (Peterson & Palermo, 2004). Some parents become distressed and depressed and their ability to cope with the disease is impaired for a number of reasons, including parental mood disorder, parenting stress (Eccleston et al., 2003) and the parents' own pain history (Schanberg et al., 2001). Parental solicitousness (Peterson et al., 2004) and over-protectiveness (Huygen et al., 2000; Simons et al., 2008), family cohesion (Scharff et al., 2005), and the family environment (Logan et al., 2005) and supportiveness (Gerhardt et al., 2003; Sällfors et al., 2001) have been shown to affect child pain and functioning with JIA.

Some studies have focused on parent-child interaction in an attempt to understand child well-being and pain. Wagner et al. (2003) found that increased parental distress was associated with greater depressive symptomatology in the child, and with high child-reported illness intrusiveness. Reid et al. (2005) explained how parent-child-interaction patterns influenced the child's adaptation to pain during an experimental task. Gauntlett-Gilbert & Eccleston (2007) found that pain-associated disability was associated with parental relationships. Parents also experience worries. Specifically, the inability to relieve the child's pain (Jordan et al., 2007), and the side-effects of medication and worries about the child's future (Barlow et al., 2002) have been mentioned as risk factors for maternal psychosocial well-being.

It is particularly important to address the role of family as a system with respect to chronic illness, such as JIA. Bowen (Kerr & Bowen, 1988) has introduced a family projection process, which illustrates how a parent may transmit his/her emotional problem to a child. According to Bowen, this projection process has three steps: first the parent focuses on a child out of fear that something is wrong with the child, second the parent may interpret the child's behaviour as confirming the fear, and finally the parent treats child as something is really wrong with the child. A recent study by Lopez et al. (2008) gave understanding of the relation between parental psychological distress and adolescent anxiety in youth with chronic illnesses. It was shown that both higher levels of perceived child vulnerability and parental psychological distress significantly predicted adolescent

anxiety, and specifically that perceived child vulnerability mediated parental distress and adolescent anxiety. According to the authors, this transaction could be understood in a way that parents who have psychological distress may be more likely to perceive their child as a vulnerable, which could result in higher levels of anxiety in the adolescent. It was conversely discussed that adolescent anxiety leads to parental distress and subsequent perceptions of their child as vulnerable.

Research on childhood illnesses that are especially characterized by chronic pain commonly reports parental depressive and anxiety symptoms. Eccleston et al. (2003) investigated children with chronic pain symptoms and their parents. Forty per cent of the parents scored above the cut-off point on the anxiety or depressive subscales, and 60 per cent scored above the cut-off on anxiety symptoms. The child's depressive or anxiety symptoms did not significantly predict parental depressive or anxiety symptoms. However, parent-child dysfunctional interaction was best predicted by the child's young age, long pain duration and high depression.

Communication as interaction between parent and child has also been a focus of interest in pain research. Among adolescent JIA patients, Shaw et al. (2006a, 2006b) recently studied parent-child agreement regarding the child's physical health and health-related quality of life, including pain. There was a higher level of agreement when the disease-related variables were recognizably absent, mild or severe, but the parents had difficulties interpreting mid-range symptoms. The authors concluded that agreement was likely to be influenced by communicational or other individual differences between the child and the parent. Palermo et al. (2004) also found that individual differences such as depressive symptoms in the child predicted parent-child discrepancy in understanding the symptoms of pain.

The role of learning the experience and expression of pain has also been questioned when trying to explain how child's pain and family factors may be considered as being connected to each other. Theoretically this focus of interest emerge from operant-behavioural theory, centred on the role of social reinforcement in maintaining maladaptive pain behaviours. Children may learn to interpret symptoms and to determine what kind of response is appropriate by observing how their parents respond to their own episodes of pain. Further, Thastum et al. (1997) compared the responses of healthy children with those of juvenile arthritis patients and their parents to the cold pressor task, and found remarkable similarities in terms of pain threshold, intensity and tolerance between the parents and the children with juvenile arthritis. Social learning may be a significant factor in explaining children's pain.

In sum, several aspects of parent-child interaction patterns may predict child functioning. Indirect (e.g. mediating) effects tend to emerge in analyses of interaction between a parent and the child and the effect of this interaction on any disease-related domain, such as pain. Parent-child interaction that

is characterized by discrepancy, overt solicitousness, or conflicting and unexpected responses from the child or parent may reveal essential elements of this interaction, such as depressive symptoms in the parent or the child. Even if the symptoms fall below the cut-off limit of clinical pathology, these parent-child interaction patterns may form a vicious circle that affects the children and their coping with and management of the disease. Parent-child interactions are transactional in nature and child-to-parent and reciprocating effects often occur over time along many pathways of influence.

1.5.4 Somatic complaints

Somatic complaints in children are fairly common. In children with a severe disease, such as JIA, somatic complaints as comorbid symptoms may be related to the disease or to the treatment of the disease. There may be many explanations for the development of somatic complaints in children with JIA, such as the disease itself (e.g., tiredness) or its medical treatment (e.g. nausea). The following variables were used to reflect somatic complaints in the current study: nightmares, constipation, dizziness, tiredness, pain, headaches, nausea, eye problems, skin problems, stomach problems and vomiting (Achenbach & Edelbrock, 1983). The Child Behaviour Checklist (CBCL: Achenbach et al., 1983), which is the most widely used international scale for assessing child behaviour, includes the somatic complaints discussed above in the factor “internalizing symptoms”, referring to anxiety, depressive symptoms and social withdrawal. Externalizing symptoms include hyperactivity, oppositional behaviour and aggression. Barlow et al. (2006) also used CBCL in their meta-analyses and found, overall, more adjustment problems and symptom internalization among children/adolescents with arthritis than among the controls.

The issue of somatic complaints has recently been argued by Crombez et al. (2009) and Merskey (2009). Both criticize the use of term somatisation used for numerous physical complaints for which there is no physical cause. Eminson (2007) suggests the use of term “medically unexplained symptoms” for its descriptive qualities in an area where it is tendency to dichotomise psyche and soma. In this thesis clustering of symptoms is the motive to include somatic complaints in the analysis, because the presence of pain tends to elevate somatic symptoms. This clustering is typically referred to as somatisation, and psychological factors are considered to have primary causal or contributory roles (Unruh & Campbell, 1999). In JIA clustering of somatic complaints and emotional symptoms can be understood in the bio-psycho-social framework (Waddell, 1984; 2004) of pain.

It is not unusual for a child to have or to develop pain in more than one bodily system. Zeltzer et al. (1997) developed the concept “pain-vulnerable child” in an attempt to enhance understanding of the clustering of somatic symptoms in children with chronic pain. Chronic pain does not develop in

all children who have significant acute pain, and not all children with chronic pain are incapacitated (Zeltzer et al., 1997). According to Zeltzer, the three key elements involved in pain perception are the child's ability to regulate the focus of attention, the rapidity and perhaps magnitude of arousal development in the face of the perceived threat, and the memory of past noxious events. Each of these elements individually and together plays a role in the child's ability to cope with pain and the perception of that sensory experience as potentially controllable or as something to be feared. Thus, a poor ability to regulate the focus of attention will be disadvantage to a child who has acute pain because the pain itself will be the major stimulus that focuses the attention. As the child focuses on the pain, anxiety grows, and a fear of pain may develop. This fear may magnify the noxious sensory experience and thus rivet the child's attention on the pain. Arousal, rumination (i.e. excessive worry about pain) and catastrophizing contribute to the child's sense of helplessness in the face of pain. Enhanced perception of pain could lead to pain-related fear and arousal, and to a reduction in activity and social isolation, which in turn will foster the focus on the pain because there are fewer distractions. Pain-fear and pain-focus become a self-perpetuating cycle. Thus, the concept of the pain-vulnerable child refers to the interacting factor that makes the child incapacitated by his/her symptoms. "Intrinsic" (e.g. a low pain threshold, poor perceived control over pain) and "extrinsic" (e.g. previous pain experiences) factors interact in this cycle to predispose the child to developing more pain than her/his peers would under similar conditions.

According to Garralda (2004) emotional disorders in children with unexplained medical symptoms are common. There are specific psychological aspects in the patients with unexplained medical symptoms that differentiate children with such medical symptoms from those with emotional disorders. One contributing factor is disease beliefs and illness behaviour. When children that have previous history of medical problems have symptoms with uncertain nature, the children and their parents may have a tendency to think that the symptoms have a biological medical cause. These beliefs may hold disease conviction in the absence of medical evidence. These beliefs, in turn may make the child more impaired by the physical symptoms. This illness-related impairment is more prominent in chronic fatigue syndrome than in patients with JIA (Garralda, 2004).

Zeltzer et al. (1997) also studied the psychological profiles of children with extreme chronic pain and pain-related disability. They found more somatic symptoms but also severe perceptual distortions, extremely poor problem solving, excessive use of denial to cope with stressful life events, and active avoidance of strong and aversive emotions in them. They concluded that it was time to change the paradigm of thinking about pain from an organ-system perspective towards

understanding pain as a complex web of interacting cognitive, emotional and neuro-physiological processes that are related to each other.

Several epidemiological studies have examined the prevalence rates of multiple pain in conjunction with other physical symptoms. A Finnish epidemiological study it was reported that children who seldom or never experienced pain had fewer diseases and symptoms than children with more frequent pain symptoms (Mikkelsen et al., 1997). Specifically, those with widespread or neck pain had a higher mean score on somatic complaints than the healthy controls. Conte et al (2003) found that children with arthritis had fewer somatic complaints than children with juvenile fibromyalgia, but more than healthy children.

A large cross-sectional study reported somatic complaints in children aged 2–17 years (Bentson et al., 2001). Scores of somatic complaints, including stomach complaints, headaches, sleeplessness, etc., increased with age and specifically in girls. Specifically in middle childhood and adolescence the incidence of somatic symptoms increases (Nunn et al., 2007). In a review article, Eminson (2007) concluded that medically unexplained symptoms, referable to term somatic complaints used in the current thesis, are extremely common in childhood and adolescence. Such symptoms were most significantly associated with later anxiety and depression, and increased medical consultation.

1.6 Resilience and vulnerability

Pain conditions are largely shaped by an individual learning history either through direct experience, modelling, or information from others (Gatchel et. al, 2007). Certain developmental history in the family system may predispose some children and their parents to make maladaptive appraisals of pain, leading the child more vulnerable to pain suffering. There are also protective factors in living with a chronic childhood illness, such as optimism or self-efficacy, a concept used in the current study, which could be called resilience factors. However, there has been relatively little research on protective factors particularly in pain research. A significant milestone in explaining chronic childhood illness in terms of the social context, and particularly including aspects of resilience, was Wallander and Varni's (1998) conceptual model of child adjustment to paediatric chronic physical disorder. This chronic illness is conceptualised as an ongoing chronic strain for both children and their parents. According to this model, a major principle is that modifiable risk and resistance factors can be identified empirically, which in turn may give guidance for the development of interventions. Resistance factors in the original model were delineated in three categories: intrapersonal, social-ecological and stress-processing. The risk factors included disease/disability parameters, functional dependency in the activities of

daily living, and psychosocial stressors. It is important that the model provides a developmental perspective to chronic childhood illness in showing that both the child and the family experience continuous development, which in turn may have positive effects on child adjustment to paediatric chronic physical disorder. The model also considers the disease related factors, both the child and parental factors, as being in interrelation and in process affecting on each other. Since Wallander and Varni's (1998) model the theory of variables affecting child adjustment to paediatric chronic physical disorders has not remarkably developed. However, multivariate models of explicit mutual associations between the variables have given new information in such situations.

The parents of a child with a chronic disorder may function much like other people who experience stressful circumstances: some develop maladjustment and others show resilience. Family influences play a central role in risk- and resistance-based research on chronic childhood illnesses. Parental empowerment, a process whereby individuals gain control over their own lives and become proactive in matters of social change benefits the child.

There is a need for prospective designs that address the complex interplay of the risk and resistance factors that influence adaptation over time, because a straightforward search for pathology in children or in their social context falls short in explaining variation in adaptation and in pain. As in Wallander and Varni's model, it is important to identify resilience factors within the whole family context, because the family plays an active role in JIA care and rehabilitation. The success of care and rehabilitation depends on the dynamic balance between the parents' ability to cope and the child's often rapidly changing somatic symptoms (e.g. inflammatory pain), psychological factors (mood) and social situation (age-dependent social roles).

1.7 Cognitive influences on pain experience

Recent pain rehabilitation is based on the recognition that individuals can influence their own health and well-being in a wide variety of situations. This theoretical perspective could be simplified as follows: patients who believe that they can control their pain, who avoid catastrophising their condition, and who believe that they are not severely disabled appear to function better than those who do not. Beliefs may mediate some of the relationships between pain severity and adjustment (Jensen et al., 1991). Thus, children who are able to deal with their pain or illness are less likely to experience feelings of helplessness with regard to their symptoms.

Variables regarding cognitive processes (i.e. self-efficacy) may facilitate psychological and functional adjustment in children with chronic illnesses (Kaminsky et al., 2006; Varni, et al., 1988). The results of research on children with physical illnesses support the existence of a relationship

between self-efficacy, adjustment, and symptoms of depression and anxiety in children with pain symptoms. Kaminsky et al's (2006) study on the psychological correlates of depression in children with recurrent abdominal pain showed a relationship between higher self-efficacy and lower levels of depressive symptoms. These findings suggest that coping style, self-efficacy, and social support are correlates of depressive symptoms in the presence of abdominal pain.

1.7.1 Social cognitive theory and the locus of control

JIA as a chronic illness challenges the families usually over a longer period of time. A relevant question is what their interpretation of their own possibilities to have control over the demanding situation is. An influential theoretical construct related to the current study is the locus of control (Rotter, 1966), according to which people tend to believe that control of the events in their lives is either primarily internal or primarily external. The locus of control refers to beliefs about whether one's actions affect outcomes. According to the theory, behaviour is influenced by generalized expectations that outcomes are determined either by one's actions or by external forces beyond one's control (Bandura, 1997). Thus a person with an internal locus of control tends to believe in his/her own ability to control events, whereas someone with external control believes that other people or events are the primary influences on their own circumstances or chances.

The results of studies on the locus of control in childhood chronic illnesses suggest that external control is related to a greater tendency to internalize problems (Thompson et al., 1993). Someone facing a challenging situation will take action based on the conviction that he or she has the skill to execute the behaviour required in order to produce the desired outcome (efficacy beliefs), and on an estimation that the chosen behaviour will lead to such an outcome (outcome expectation/ locus of control) (Main et al., 2008). Strong efficacy beliefs and positive outcome expectations are characterized as productive behaviour affording personal satisfaction, whereas a low level of efficacy beliefs and negative outcome expectations are characterized as behaviour reflecting apathy or resignation. However, although closely related, self-efficacy beliefs and outcome expectations represent different phenomena (Bandura, 1997): self-efficacy is a good predictor of behaviour, whereas locus of control (outcome expectation) is an inconsistent predictor of the same behaviour.

It has since been posited that the locus-of-control theory simplifies the complex reality, and that the concept is ambiguous. However, the term self-efficacy, which is of interest in the current study, arose from this theory. It is assumed that beliefs that personal actions determine outcomes give rise to a sense of efficacy and power, whereas beliefs that outcomes occur regardless of what one does create apathy (Bandura, 1997). It is therefore important to distinguish beliefs about the

locus of the outcome from beliefs about personal efficacy, because the locus of control represents a judgement about the likely consequence of actions whereas self-efficacy refers to perceived ability to carry them out (Bandura, 1997) However, the belief that outcomes are determined by one's own behaviour may be demoralising or empowering, depending on whether or not one believes one can produce the required behaviour. Thus, self-efficacy and locus of control together constitute a theoretical framework within which a person's beliefs and actions can be better understood. People, who regard outcomes as personally determined, but who lack the required skills, experience a low sense of efficacy and view activities with a sense of futility.

1.7.2 Self-efficacy

According to Bandura (1977) who was the first to use the term, self-efficacy refers to an individual's belief about being capable of executing the behaviour required to produce a particular outcome. Beliefs of personal efficacy constitute the key factor in human agency. If people believe they have no power to produce results they will not attempt to make things happen (Bandura, 1997). Beliefs in one's ability to exercise control constitute a pathway through which psychosocial influences affect health functioning (Bandura, 2004). In general, self-efficacy beliefs determine how obstacles are viewed and attributed. The stronger the self-efficacy, the higher the goals people set for themselves and the firmer is their commitment to overcoming obstacles.

The original theory of self-efficacy was based on social cognitive theory, which posits that behaviour is a result of interaction between both personal and environmental variables (Holloway, & Watson, 2002). Thus, most behaviour is determined by many interacting factors: people are contributors to rather than sole determiners of what happens to them and their behaviour is shaped through learning from the environmental conditions, and in turn, individuals shape the environment (Bandura, 1997). According to Bandura (1997), people are both producers and products of social systems and they can exercise influence over what they do. In terms of social cognitive theory, a person's adaptation or change is rooted in social systems in that human behaviour cannot be understood solely in terms of social-structural or psychological factors.

In the context of JIA self-efficacy refers to the belief of an individual that he/she has the ability to execute the behaviour required for tackling the condition (Barlow et al., 2000, 2001). It refers not only to the courses of action pursued but also to the effort expended, perseverance in the face of difficulties, the nature of thoughts (encouraging/self-deprecating), and affective reactions (Barlow et al. 2000). According to the preliminary validation of the first arthritis self-efficacy scale for children with JIA developed by Barlow (2001), higher scores were associated with better physical

and psychosocial well-being among children and their mothers. As the founder of the self-efficacy concept, Bandura (2004) states that people guide their lives by their beliefs of personal capabilities. Perceived self-efficacy refers to beliefs in one's capabilities to organize and execute the course of action to produce given attainments (Bandura, 2004). In daily life individuals are both agents and objects, capable of self-reflect and self-influence by analyzing the situation that confront them, considering alternative courses of action, judging their ability to carry them out successfully, and estimating the results the actions are likely to produce. People act on their judgments, reflect on how well their thoughts have served them in managing the events at hand, and change their thinking and strategies accordingly (Bandura, 2004). A triadic reciprocal causation model (Bandura, 2004) illustrates the theoretical framework by which it can be understood, why and how an individual acts upon and how this functioning is socially affected in a reciprocal manner. In this model, human agency operates within an interdependent causal structure involving triadic reciprocal causation. Internal personal factors in the form of cognitive, affective, and biological events, behaviour and environmental events all operate as interacting determinants that influence on one another birectionally. Human adaptation and change are rooted in social systems in such a manner that social structures (which are devised to organize guide, and regulate human affairs) do not arise by immaculate conception. They are created by human activity and behaviour is partly regulated by the social reactions it evokes (Bandura, 2004). Bandura states that human behaviour cannot be understood solely by either social structural factors or psychological factors: a full understanding requires an integrated perspective. Human agency operates generatively and proactively rather than just reactively for instance through self-efficacy beliefs. Performances do not just happen to a person; a person does a lot to bring them about (Bandura, 2004).

Measures of self-efficacy are based on Bandura's social cognitive theory (Bandura, 1997). Bandura posits assessment on three levels of generality: general, intermediate and specific self-efficacy beliefs, of which intermediate beliefs refer to a performance class within one domain under a class of conditions sharing common characteristics (Barlow et al. 2001). The items in the measures are phrased in terms of "can do" rather than "will do" (Bandura, 1997). In terms of research on pain, several single-item self-efficacy scales have been developed (Brady et al. 2003) to assess self-efficacy in relation to specific aspects of experimental paradigms (Main et al. 2008). Of such scales developed within arthritis research five target specific beliefs and three target generalized beliefs (Brady, 2003). The chronic-pain self-efficacy scales in Rosenstiel's and Keefe's (1983) CSQ (Coping Self-efficacy Questionnaire), amended more recently by Anderson et al. (1995), are most widely used in assessing coping efficacy in relation to pain. However, they were designed for adults. RASE (Rheumatoid

Arthritis Self-efficacy), the most recent adult scale, was developed by Hewlett et al. (2001). Barlow et al. (2001) generated the first arthritis self-efficacy scale for children (CASE). It measures their perceived ability to control or manage salient aspects of life with JIA. Three subscales cover the aspect (items) of arthritis-related self-efficacy: the symptom subscale (pain, joint stiffness, tiredness, swollen joints), the emotional subscale (sadness, loneliness, annoyance) and the activity subscale (school attendance, physical education and school games, friends and family). The purpose in creating a new scale was twofold: it was thought that the perceived ability to carry out courses of action that produced desired attainments in the domain of life with JIA would enhance understanding of the variations in adjustment (Barlow et al., 2001), and that children's arthritis self-efficacy might be a useful outcome measure in the evaluation of psycho-educational interventions.

Julie Barlow (2000) was also the first to develop a self-efficacy measure for use among parents of children with JIA. The rationale behind the development was that the perceived ability to carry out courses of action that would produce the desired attainments in caring for children with JIA would enhance understanding of the variations in parental adjustment (Barlow et al., 2000). Parental self-efficacy was defined as the perceived ability to control or manage salient aspects of their child's JIA. The aspect of arthritis-related self-efficacy in parents comprised pain, joint stiffness, joint swelling, sleep, non-medical control of pain, fatigue, activity, sadness, loneliness, frustration, pleasure, school activities, and friends and family (Barlow et al., 2000).

2 Aims of the study

The overall aim of the current study was to identify the factors that explain the variance in pain among children with JIA by combining inter- and intrapersonal, and bio-psycho-social variables chosen in the analyses. Parental and child variables were included as potential explanatory variables. First, associations and interrelationship between the variables were sought, and following that, pain as the outcome variable was included in the analysis. Specifically, the focus was on a set of variables including the depressive and anxiety symptoms of the child and the parents, cognitive child and parental factors such as self-efficacy, and the interaction between child and parent variables that might be affected by the above-mentioned variables and thus increase or reduce the risk to the affected child of having pain.

The specific aims of the study were:

1. to characterize the frequency of pain in children with JIA (at baseline and in the one-year follow-up) (IV, V);
2. to investigate whether homogenous subgroups of juvenile patients could be identified based on trait anxiety, depression and age, and to compare the differences in clinical parameters among the subgroups found (II);
3. to identify which parental variables (distress, arthritis self-efficacy, sense of illness management) would discriminate between the four cluster groups of children with JIA (III);
4. to predict parent-child interaction patterns related to pain in JIA, and to investigate to what extent these patterns explain the variance in levels of pain experienced by the child (IV);
5. to test the factor structures and validate the CASE (Barlow et al. 2001) and PASE (Barlow et al. 2000) self-efficacy scales in a Finnish sample of patients with JIA and their parents, to investigate the association between the self-efficacy of the child and disease-related parameters, pain and distress, and to describe how parental self-efficacy is associated with the child's functioning (I, II, III, IV)

3 Methods

3.1 Outline of the study

General outline of the study and statistical methods used in each sub-study are shown in Figure 2. First, descriptive statistics were used to characterise the frequency of pain in children with JIA. Secondly, preliminary screening of the relationships between the variables pain and depression, trait-anxiety and age produced scant results in terms of linear relationships: associations based on combinatory elements rather than additive components seemed to have predominance in the material. These findings motivated the selection of an exploratory study design, with a view to achieving the second aim of the study – to identify the associations between the cluster grouping and the child and parental variables. Of the clustering methods both hierarchical and K-means cluster analysis were applied (see Santavirta et al., 1996). K-means clustering was considered the most applicable algorithm, given the fairly large sample (Metsämuuronen, 2006).

Thirdly, Descriptive linear discriminant analysis (Silva & Stam, 2004) was applied in order to follow the methodological track. Discriminant analysis allows a set of variables to be identified as forming a discriminant function that can predict the group membership of the outcome variable. The independent variables used included parental wellbeing, illness management and parental self-efficacy. The dependent categorical variable was the cluster grouping of the child.

The fourth aim was to assess parental and child self-efficacy. The first step was to validate the self-efficacy scales by means of factor analysis. Exploratory factor analysis of a Finnish sample identified no meaningful factor structure from the two-factor solution, as in the original study conducted by Barlow et al. (2000). The next step was to conduct confirmatory factor analyses (Gonzalez & Griffin, 2001). The association between the self-efficacy of the child and the disease-related parameters pain and distress was investigated in order to find out how parental self-efficacy is associated with the child's functioning.

The final question to be addressed concerned whether parent-child interaction patterns exist, and if so to what extent they explain the variance in levels of pain experienced by the child. Exploratory factor analysis was again used in order to see if any such patterns emerged. The extent to which these common factors might account for the variance in pain levels experienced by children diagnosed with JIA was assessed by means of linear multiple regression analysis.

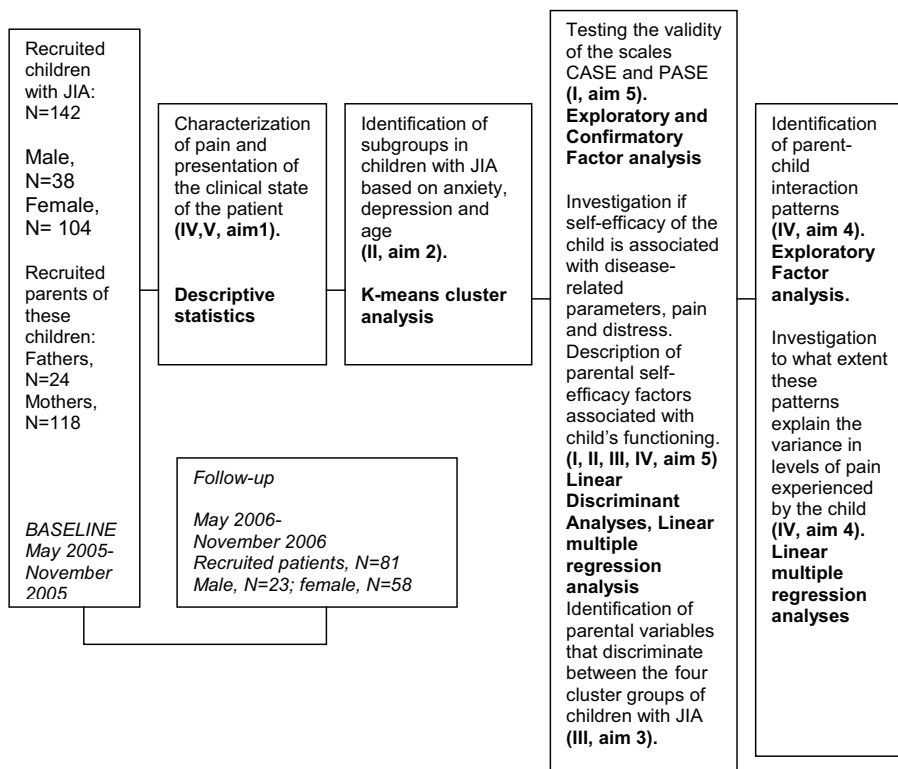


Figure 2. General outline of the study and methods used in each study. Roman numeral refer to individual substudies that have allowed examinations of specific research questions. Aims refer to the specific aims of the thesis.

3.2 Participants

3.2.1 Patients

The patients included had JIA diagnosis (Petty et al., 2004) at least one year prior to the study, and were aged eight and 15 years at the start of the study. One hundred and forty-two consecutive patients during routine clinical visits were recruited over a six-month period. 99 patients were recruited from the Rheumatism Foundation Hospital in Heinola, with a catchment area covering the whole country except for the Helsinki Metropolitan area, which participated by recruiting 43 patients from the Pediatric Rheumatology Clinic of the Helsinki University Hospital. The parent attending with the child was also invited to participate. Only nine parents and/or patients refused. All the patients and parents signed informed consent forms. The mean age of the patients was 11.92 ($SD = 2.25$) years. JIA was diagnosed between one and 14 ($M = 6.15$) years before enrollment. Of the patients, 104 (73.2 per cent) were girls and 38 (26.8 per cent) boys. All of

the participants were Caucasians. Fifty percent of the patients had a polyarticular, 16.9 percent an extended oligoarticular, and 33.1 percent an oligoarticular disease course.

3.2.2 Parents

There were 24 (17 per cent) fathers and 118 (83 per cent) mothers in the study. The mean age of the fathers was 38.5 years ($SD = 6.56$) and of the mothers 36.5 ($SD = 6.06$) years.

3.3 Measures

The selection of the instrument set used in the study was based on validity. The aim was to use well-documented methodology in the context of research on pain and on childhood illnesses and parental experiences and reactions to it. One new scale, Parental illness management, was created. Additionally, three scales, PASE (Barlow et al., 2000), CASE (Barlow et al., 2001) and STAIC (Spielberger et al., 1973, 1983) were translated into Finnish and then validated. The translation was done in three steps (Beaton et al., 2000). An interdisciplinary team comprising a certified translator, a psychologist, a physiotherapist, a paediatric rheumatologist and a professional, senior researcher translated the original questionnaires from English into Finnish. This translated version was back-translated into English by an independent certified translator, who did not participate in the first translation session. The final consensus version in Finnish was produced in a joint session in which all of the above participated, with access to the original, translated and back-translated versions of the questionnaires.

Well-documented scales were used to measure the responses of both parents and children. They were all reliability tested for the current study. Cronbach's α s were calculated in order to test the internal consistency of all the scales included except for the separate items measuring Parental illness management, for which reliability was estimated from the communalities in a three-factor Principal Axis Factor analysis.

3.3.1 Scales for children with JIA

3.3.1.1 Trait anxiety among the children

Trait anxiety ("anxiety") in the children was measured by means of the State-Trait Anxiety Inventory for adolescents and adults (STAI, Spielberger et al., 1983) and the State-Trait Anxiety Inventory for Children below thirteen years of age (STAIC) (Spielberger et al., 1973). STAIC was used for 96 children and STAI for 46 adolescents. Both inventories consist of two subscales:

the State-anxiety scale (S-scale) and the Trait-anxiety scale (T-scale). The S-scale is designed to measure subjective, consciously perceived feelings of apprehension, tension and worry, which fluctuate over time and are influenced by the immediate environment. The T-scale is an indicator of the level of trait anxiety experienced by children, and represents how they generally feel. It is relatively impervious to the conditions under which it is given. Therefore, only the T-scales were used in this study. The internal consistencies of the STAIC ($\alpha=0.89$) and STAI ($\alpha=0.88$) T-scales were good. Since the scale intended for children (STAIC) differs slightly from that meant for adolescents (STAI), the two T-scales were combined and adjusted to be scale invariant so that the minimum value was zero and the maximum value was 30, the higher values indicating stronger anxiety.

3.3.1.2 Depression/mood of the children

The Finnish version the Child Depression Inventory (CDI, Kovacs, 1985) was used to measure the depression/mood disturbance of the patients. The scale has previously been validated in a Finnish sample (Almqvist et al., 1991). The Finnish version includes 26 of the 27 items in the English version: the question about suicide was excluded for ethical reasons. The score for each item varies from zero to two, and the total score thus varies from zero to 52. Higher values indicate increasingly severe depression. In the current study the internal consistency of the scale was good ($\alpha = 0.85$).

3.3.1.3 Functional disability

The Childhood Health Assessment Questionnaire (CHAQ, Ruperto et al., 2001) was used to measure the children's functional status. It assesses performance in eight areas (dressing and grooming, rising, eating, walking, hygiene, reaching, gripping and other activities), providing an overall disability score within the range from zero to three. The higher scores indicate greater functional impairment. The CHAQ has been reported to be reliable and sensitive, and it has been validated in a Finnish sample (Pelkonen et al., 2001). In the current study the internal consistency of the scale was good ($\alpha = 0.84$).

3.3.1.4 Pain

Pain was measured by means of the Structured Pain Questionnaire (SPQ, King & Wold, 1996; Mikkelsen et al., 1998). The questionnaire uses a five-level frequency classification of pain over the previous three months (seldom or never, once a month, once a week, more than once

a week, almost daily). Each of the seven pain areas (neck, upper and lower extremities, chest, upper back, lower back and buttocks) was scored from zero to four, the total (frequency and area combined) score ranging from zero to 28. The body area concerned was marked on a picture beside the question to help the child to recognise it. In this study the internal consistency of the scale was good ($\alpha = 0.75$).

3.3.1.5 Somatic complaints

The somatic-problems subscale of the Child Behaviour Checklist (CBCL, Achenbach & Edelbrock, 1983) was used to measure the general somatic symptoms of the children. The reliability and validity of the scale have been documented in Finland (Almqvist, 1988) as well as in many other countries. In total, the CBCL consists of 118 questions, each scored from zero to two, from which one total somatic-problem score is summed. The somatic-complaints subscale includes the following items: nightmares, constipation, dizziness, tiredness, pain, headaches, nausea, eye problems, skin problems, stomach problems and vomiting. The alpha coefficient for this subscale was $\alpha = 0.82$ in the current study.

3.3.1.6 Self-efficacy

The Self-efficacy Scale for Children (CASE, Barlow et al., 2001), for children aged 7–17 years with JIA was used to measure belief in one's efficacy in exercising control over arthritis-related problems. The original CASE questionnaire includes 11 items concerning the child's own JIA management (Barlow et al., 2001). The children were asked to rate how certain, on a five-point scale ranging from 1 (= very uncertain) to 5 (= very certain), they are of being able to manage physical and psychosocial issues related to their disease. According to Barlow and her co-workers (Barlow et al. 2001), these 11 items measure three different aspects of the child's self-efficacy – the “symptom subscale” (items 1–4), the “emotion subscale” (items 5–7) and the “activity subscale” (items 8–11).

3.3.1.7 Active joint count

An active joint was defined as a joint with swelling, or a tender joint with a limited range of motion (Giannini et al. 1997). A paediatric rheumatologist assessed the active joint count.

3.3.2 Scales for the parents

3.3.2.1 Parental depression

Beck's Depression Inventory II (BDI II, Beck et al., 2004) was used to assess parental depression and mood disturbance. The inventory comprises 21 items, the scores ranging from zero to three, and the total score from zero to 63. The higher scores indicate more severe depression. In the current study the internal consistency of the scale was good ($\alpha=0.90$).

3.3.2.2 Parental depressive and anxiety symptoms

Parental depressive and anxiety symptoms were assessed on the Hospital Anxiety and Depression Scale (HADS, Zigmond & Snaith, 1983), which contains 14 items rated on a four-point scale (0–3), and includes anxiety (HADS-A) and depressive (HADS-D) symptom subscales. Seven of the items focus on depressive and the other seven on anxiety symptoms. HADS is a self-rating instrument, the aim of which is to measure anxiety and depression in a context related to disease and illness, whereas BDI (Beck et al. 2004) measures clinical depression. The total score ranges from zero to 42 and each of the subscales from zero to 21, the higher scores indicating greater levels of anxiety and/or depression. The internal consistency of the scale was good ($\alpha=0.88$) in the current study. HADS performs well in assessing symptom severity in adult depressive patients and in the general healthy population (Bjelland et al., 2002).

3.3.2.3 Parental self-efficacy

The Self-efficacy Scale was used to measure parental self-efficacy (PASE, Barlow et al., 2000). It is meant for parents who have a child with JIA, and measures belief in parental efficacy in exercising control over the child's JIA-related problems. The original PASE scale includes 14 questions about parental management (Barlow et al., 2000). The parents are asked to rate how confident they are of their ability to control their child's adjustment (e.g., pain, sadness and joint stiffness) on a seven-point scale ranging from 1 (= very uncertain) to 7 (= very certain). According to Barlow and her co-workers (2000), these 14 items measure two different aspects of parental self-efficacy in this context, which they called the "symptom subscale" (items 1–7) and the "psychosocial subscale" (items 8–14).

3.3.2.4 Parental illness management

The parents were asked to express their views with regard to seven items on a visual analogue scale (0–100) (Table 2). An interdisciplinary team comprising a paediatrician, a psychologist, a physiotherapist and a social worker chose the items. They were generated based on four domains typically targeted in the treatment of chronic childhood illnesses: disease management, emotional problems, health prevention and health promotion (Barlow & Ellard, 2004). The reliability of these seven separate variables varied from 0.40 to 0.63, estimated from the communalities in a three-factor Principal Axis Factor analysis (Child, 2006). Communality (h^2) is a measure of the unique or error variance of an item (or vice versa of the common variance), and it is the absolute lowest bound of reliability. The higher the communality the more reliable the item is. The scale was named Parental illness management.

Table 2. Descriptive data of the items measuring parental illness management.

Parental illness management dimensions	h^2	min	max	M
Influence-disease Reflecting parents' current perceptions of their influence on the child's disease	0.52	0	100	50.1
Influence-mood Reflecting parents' current perceptions of their influence on the child's depressive symptoms related to the disease	0.58	12	100	68.1
Influence-treatment Reflecting parents' current perceptions of their influence on the treatment of the disease	0.44	0	100	62.7
Severity Reflecting parents' current perceptions of the severity of the disease	0.59	0	100	59.6
Coping Reflecting parents' current perceptions of how well the child was coping with the disease	0.40	21	100	76.4
Pain intensity Reflecting parents' current perceptions of child's pain intensity	0.52	0	100	17.6
General well-being Reflecting parents' current perceptions of the child's level of general well-being	0.63	0	96	17.0

3.4 Statistical analyses

SPSS 14.0 for Windows (Norusis, 2005) was used in the data analysis in studies I-IV. The descriptive values of the variables were expressed as means and standard deviations or percentages. It was assumed that missing data was missing at random (MAR). The few MAR values were multiply imputed prior to the analyses in accordance with the Normprogram data-augmentation

procedure (Schafer, Retrieved August 1, 2007, from <http://www.sat.psu.edu/~jls/misoftwa.html>). Iteration was carried out in 2,000 cycles; each 500th cycle produced one imputed matrix. The expectation-maximization (EM) method was used to obtain the initial estimates (Tabachnick et al., 2007; Hill et al., 1977). Some distributions were non-normal in some of the analyses and the interval assumption could not always be defended. In these cases analysis were run in parametric, but also with non-parametric statistics by Muthén's Mplus, version 4.1 (Muthén & Muthén, 2006) using the variables as ordered categorical (ordinal) and censored below (variables having a floor effect). The descriptive values of the pain variables were expressed as means, standard deviations and percentages. Both hierarchical and *k*-means clustering were used in the empirical classification of the children. Since the trait anxiety and depression correlated, squared Mahalanobis distances were used in the calculations. The most stable classification produced four clusters using the *k*-means procedure with running means. Multivariate analysis of variance (MANOVA) was used to evaluate the overall differences between the four cluster groups in disease duration, pain and somatic complaints. Differences between the cluster groups were tested separately for each variable (disease duration, pain, functional disability and somatic complaints) by means of a one-way-analysis of variance (ANOVA). The contingencies between the diagnoses and the cluster group were chi-square tested.

The first step in identifying which parental variables (distress, arthritis self-efficacy, sense of illness management) discriminated between the four cluster groups was to test the skewness of the continuous variables on the Q-plots and the Shapiro-Wilks test. Descriptive linear discriminant analysis (LDA) was used to examine the differences among the children's cluster groups in the parental variables (Silva & Stam, 2004). Discriminant analysis is a method that allows a set of variables to be identified as forming a discriminant function, which could predict the group membership of the outcome variable. The independent variables in this study were parental wellbeing, illness management and parental self-efficacy. The dependent categorical variable was the cluster grouping. There are two steps in discriminant analysis: (1) forming the discriminant functions (the linear composites of the original variables) and (2) testing how correctly they reclassify the cases. Several pieces of information are reported: (1) the equality of the group means (ANOVA); (2) the equality of the covariance matrices of the dependent variables across the groups (Box's *M*); (3) the canonical discriminant-function structure coefficients; (4) the significance of the discriminant functions; (5) the function values at the group centroids, representing the average weighted discriminant scores for each criterion group; and (6) the classification results. The Cronbach's alpha coefficient was used

to assess the reliability of the subscales, and to derive the lowest bound of reliability for the single items from the communalities in a Principal Axis Factor Analysis.

The factor structure of the PASE and CASE scales was verified by means of Confirmatory Factor Analysis (CFA) and the use of AMOS software (Arbuckle & Wothke, 1999). Prelis 2 was used for computing polychoric correlations (Jöreskog & Sörbom, 2002). The Principal Axis Factoring method was used for the Exploratory Factor Analysis (EFA). The rotation was oblique. CFA was conducted without gender distinction, and the latent factors were free to co-vary in the model. AMOS software provides a number of goodness-of-fit statistics and experts generally recommend the use of a variety of fit indices so that the weakness of one index is offset by the strength of another (Gonzalez & Griffin, 2001). The goodness of fit of the models in the current study was evaluated using (1) the χ^2 -test, (2) the relative χ^2 (χ^2/df), (3) the CFI, and (4) the RMSEA. Cronbach's alpha was used to test the reliability or the internal consistency of the subscales derived by means of CFA. Descriptive statistics were used to describe the central tendency and the variability of the subscales. Student's t-test was applied to test the differences between groups (mothers and fathers). Pearson's correlation coefficient was used to measure the construct validity. Statistical significance was set at $\alpha < 0.05$. MANOVAs evaluated the associations between the cluster groups vs. children's self-efficacy, and between parental self-efficacy and the child parameters. An exploratory factor analysis was conducted in order to identify the interactions between the parent and patient variables. The extraction method was Principal Axis Factoring with Direct Oblimin oblique rotation. The percentage of variance accounted for by each factor was used to determine the number of factors included in the rotation. Kaiser-Meyer-Olkin was used as the measure of sampling adequacy. Because the factor scores were multidimensional and did not have equal covariance, and the scale was a weighted sum, Cronbach's alpha was not an appropriate measure of reliability, and the weighted factor scores minimising the measurement error were therefore taken into account. A multiple regression analysis with the created factor scores as predictors was used to determine how well the scores explained pain in children with JIA.

3.5 Ethics

The principles of the Declaration of Helsinki (World Medical Association Declaration of Helsinki. Ethical Principles for Medical Research involving Human Subjects) were followed (Retrieved June 9, 2009 from <http://www.wma.net/e/policy/b3.htm>). All of the patients and their parents received both oral and written information about the study and gave their written informed consent. The study protocol and procedures were accepted by the Ethics Committee of the Päijät-Häme Hospital District.

4 Results

The main results of the four separate sub-studies are explained in detail in the original articles (referred to in the title of the results). Figure 3 shows the main results of each sub-study, and Table 3 presents the descriptive data covering all the variables included in the study, and the correlation coefficients.

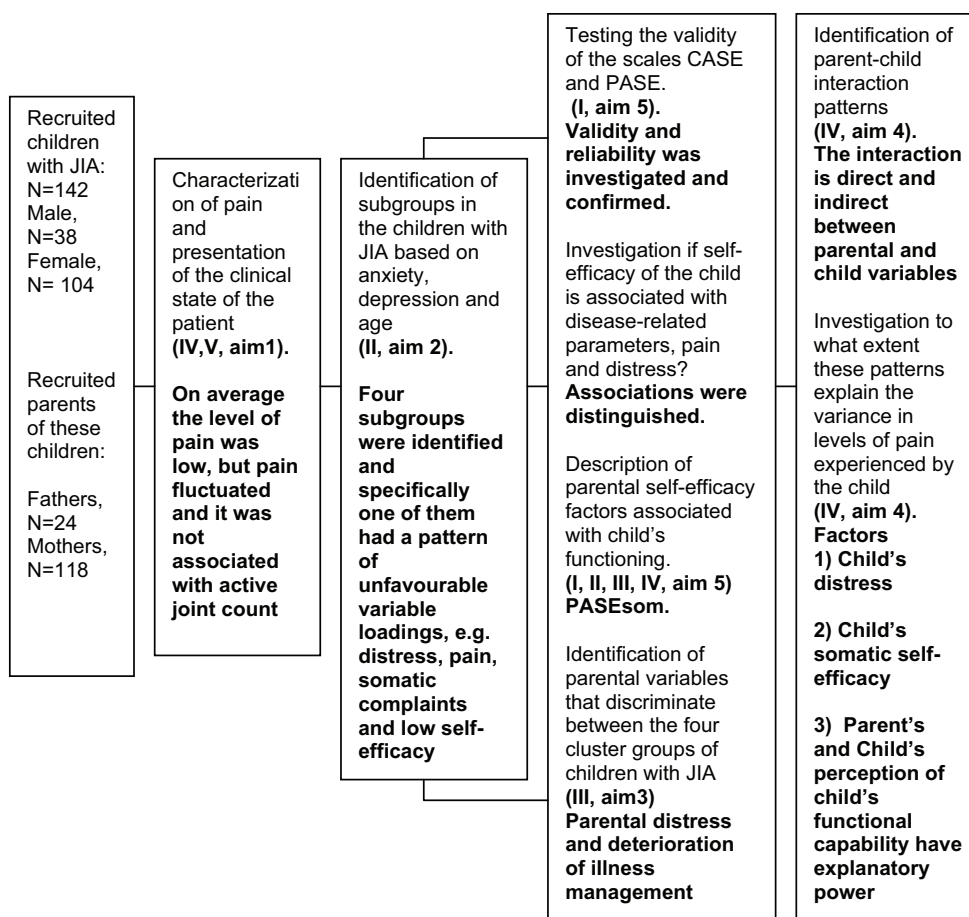


Figure 3. Results of the studies 1-IV and aims 1-5.

4.1 Pain and disease activity (IV, V)

The total score on the structured pain questionnaire (including five-level frequency classification in seven body areas) indicated that children with JIA reported fairly low levels of pain. The average score measured over the previous three months was 3.7 ($SD = 4.3$, range 0–19, on a 0–28 scale). Nine percent of the children reported neck pain and nine percent reported pain in

Table 3. Descriptive statistics of the parent variables and intercorrelations between parent variables and children variables

Parent variables	Children variables										
	M(SD)	Pain (SPQ)	Anxiety	CDI	CHAQ	CASEsom	CASEpsy	CASEsoc	Som Compl	Active joints	Pain-VAS
M(SD)	3.77(4.30)	11.03(6.55)	4.87(4.62)	0.23(0.39)	4.30(0.74)	3.74(0.91)	4.30(0.74)	3.80(2.63)	1.7(3.2)	15.56(23.3)	
BDI	6.20(6.18)	0.26 **	0.39 ***	0.46 ***	0.24 **	-0.16	-0.24 **	-0.31 **	0.37 ***	0.18 *	0.21 *
HADS-A	4.4(3.01)	0.12	0.27 **	0.29 **	0.16	-0.16	-0.26 **	-0.17 *	0.29 **	0.14	0.08
HADS-D	2.48(2.81)	0.19 *	0.37 ***	0.38 ***	0.18	-0.17	-0.21 **	-0.26 **	0.30 **	0.15	0.14
PASEsom	4.37(1.10)	-0.17 *	-0.11	-0.15	-0.07	0.23 **	0.27 **	0.33 ***	-0.12	-0.20 *	-0.16
PASEpsy	4.81(1.17)	-0.10	-0.10	-0.26 **	-0.08	0.24 **	0.30 **	0.26 **	-0.24 **	-0.17 *	-0.08
PASEsoc	5.42(1.13)	-0.20 *	-0.11	-0.19 *	-0.12	0.31 **	0.24 **	0.37 ***	-0.21 *	-0.13	-0.22 **
Inf-disease	50.15(24.24)	0.04	-0.26 **	-0.28 **	0.09	0.15	0.21 *	0.14	-0.09	-0.23 **	0.01
Inf-mood	68.10(18.74)	-0.09	-0.20 *	-0.38 ***	-0.09	0.22 **	0.27 **	0.30 **	-0.24 **	-0.16	-0.19 *
Inf-treatment	62.80(19.09)	0.07	-0.02	-0.11	-0.03	0.20 **	0.12	0.04	-0.17 *	-0.15	-0.53
Severity	59.56(20.54)	0.11	0.01	0.12	0.07	0.08	0.00	-0.25 **	-0.04	0.28 **	0.20 *
Coping	76.42(17.96)	-0.24 *	-0.02	-0.16	-0.11	0.14	0.20 *	0.18 *	-0.12	-0.05	-0.20 *
Parental Pain-VAS	17.05(22.0)	0.67 ***	0.15	0.20 *	0.41 **	-0.18 *	-0.22 *	-0.31 **	0.25 **	0.14	0.64 ***
General well-being	17.00(19.73)	0.46 ***	0.09	0.11	0.47 ***	-0.22 **	-0.21 *	-0.30 **	0.16	0.13	0.51 **

*p<0.05, **p<0.01, ***p<0.001

BDI=Beck's depression inventory , HADS-A=Hospital anxiety-depression scale (anxiety) , HADS-D=Hospital anxiety-depression scale (depressive symptoms)
PASEsom=parental self-efficacy with somatic symptom, PASEpsy=parental self-efficacy in psychological functioning, PASEsoc=parental self-efficacy in social functioning,
Inf-disease=parental perception of their influence on child's disease, Inf-mood=parental perception of their influence on child's mood,
Inf-treatment=parental perception of their influence on child's treatment, Severity=parents' perception of the severity of the disease,
Coping=parents' perception of how well the child is coping with the disease, parental pain-VAS= parent's estimate of the intensity of child's pain,
General well-being=parent's estimate of the level of child's general well-being , Pain= pain frequency, CDI= child depression inventory, CHAQ=functional disability
CASEsom=child's self-efficacy with somatic symptoms, CASEpsy=child's self-efficacy in psychological functioning, CASEsoc=child's self-efficacy in social functioning
Som. Compl=somatic complaints

the lower extremities almost daily during that time (Fig 4). Only 25 per cent of them had been pain-free, and 46 per cent had pain at least once a week in at least one of the body areas.

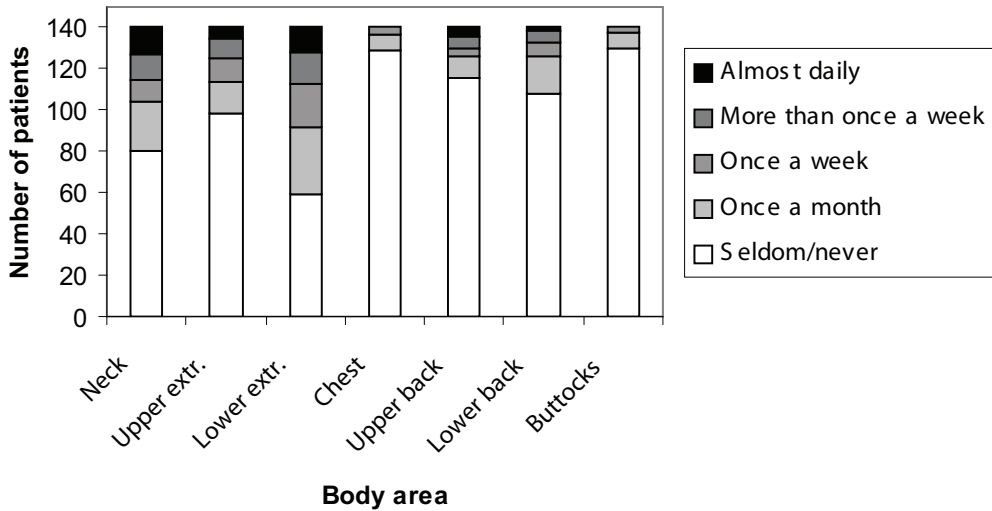


Fig. 4. Frequencies of pain in different body areas in patients with JIA.

According to the parental Pain-intensity score (VAS) and the Child self-reported Pain-intensity score (VAS) the parents estimated their child's current pain more highly than the child (Table 3). The bivariate correlation between the parental ($M = 17.05$, $SD = 22$) and the child scores (VAS) ($M = 15.56$, $SD = 23.3$) was significant ($r = .64$, $p < .001$), as were the correlations between current child-reported pain intensity (VAS) and the structured pain questionnaire ($r = .64$, $p < .01$). The follow-up results of the sample indicate persistence in pain: crude correlation between time 1 and time 2 in the structured pain questionnaire was significant ($r = .63$, $p < .001$) (Fig 5). In conclusion, almost half of the children with JIA had pain symptoms at least once a week, and the children and their parents shared corresponding views of the pain intensity. Disease activity ($M = 1.7$, $SD = 3.2$) assessed as of the number of active joints was fairly low (Table 3), and 50 per cent of the children had no joint activity at the time of the measurement.

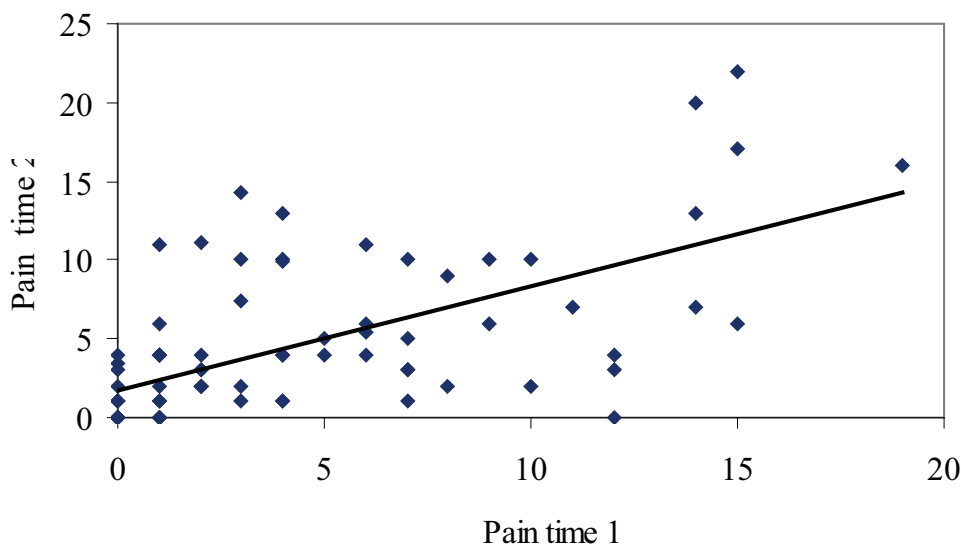


Fig. 5. Pain measured by SPQ at baseline (time 1) and at one-year follow-up (time 2).

4.2 Classification results (II)

Given that the relationship between disease parameters and pain is not straightforward, it is essential to construct a framework that will enhance understanding of the moderating and mediating factors. Trait-anxiety, depression and age could theoretically be considered mediators/moderators that influence the relationship between clinical activity and severity, and pain. The children were classified based on their emotional wellbeing and age (age, trait-anxiety and depression). The associations between the subgroups (similarly based) and disease-related parameters such as pain, somatic complaints and self-efficacy were analysed.

4.2.1 Homogenous subgroups of children based on trait-anxiety, depression and age

The correlations between pain, trait anxiety, depressive symptoms, functional disability, somatic complaints and self-efficacy in social functioning were significant (Table 3). Joint activity (disease activity) was not associated with pain. In terms of depressive symptoms 9.6 per cent of the children reached the cut-off point (13 points). Trait anxiety was measured on two corresponding scales designed for use among children between the ages of nine and 12 (STAIC) and adolescents (STAI). The STAIC mean was 30.91 ($SD = 6.48$, range = 20–46) and the STAI mean was 35.74 ($SD = 8.68$, range = 23–60). *k*-means clustering was used in order to determine whether there

were any distinguishable groupings (Table 4). The categorising variables chosen were age, trait anxiety, depression. Following exploration of several options a four-cluster solution was reached in which the cluster centroids differed (Wilk's lambda = 0.071, $F = 73.87$, $df1 = 9$, $df2 = 338.44$, $p < 0.001$).

The cluster means of the categorising variables were age ($F = 84.50$, $df1 = 3$, $df2 = 141$, $p < 0.001$), trait anxiety ($F = 66.39$, $df1 = 3$, $df2 = 141$, $p < 0.001$) and depression ($F = 80.08$, $df1 = 3$, $df2 = 141$, $p < 0.001$) (Table 4). The number of patients in each cluster was as follows: Cluster 1, $n = 20$, Cluster 2, $n = 43$, Cluster 3, $n = 28$ and Cluster 4, $n = 54$. The cluster groups were named: (1) teenagers with high scores (compared to the other cluster groups) for trait anxiety and depression, (2) children scoring highly on trait anxiety and low on depression, (3) children and (4) teenagers with low scores for trait anxiety and depression (Table 4).

Table 4. Children's cluster solution

	Cluster 1	Cluster 2	Cluster 3	Cluster 4
Age				
<i>M(SD)</i>	13.2(2.0)	10.1(1.5)	10.2(1.2)	13.9(1.0)
<i>Min</i>	8.4	7.5	8.0	11.9
<i>Max</i>	15.7	13.9	12.3	15.7
Anxiety				
<i>M(SD)</i>	19.5(4.5)	15.0(4.4)	5.4(3.7)	7.6(4.3)
<i>Min</i>	11.7	8.0	0.0	0.0
<i>Max</i>	30.1	26.0	13.0	16.7
Depression				
<i>M(SD)</i>	14.0(3.8)	4.3(2.8)	3.6(3.4)	2.7(2.1)
<i>Min</i>	8.6	0.0	0.0	0.0
<i>Max</i>	22.0	12.0	11.0	7.0

4.2.2 Association of pain and clinical parameters to children's cluster grouping (II)

Preliminary analysis of the cluster groups revealed equal diagnosis rates for oligoarthritis, oligoextended and polyarthritis in all four. In terms of clinical parameters there were significant overall differences between them (Wilk's lambda = 0.691; $F = 3.61$, $df1 = 15$, $df2 = 378.60$, $p < 0.001$). When investigated separately for each clinical parameter the groups differed significantly in disease duration ($F = 3.77$, $df1 = 3$, $df2 = 141$, $p = 0.012$), pain ($F = 5.60$, $df1 = 3$, $df2 = 141$, $p = 0.002$), functional disability ($F = 5.00$, $df1 = 3$, $df2 = 141$, $p = 0.003$) and somatic complaints ($F = 6.35$, $df1 = 3$, $df2 = 141$, $p < 0.001$) (Table 5). According to Wilk's test there

were significant differences between the groups in disease duration, pain, somatic complaints and functional disability, but not in diagnosis or active joint count. With regard to active joint count the differences were not significant, but a trend emerged when clusters 2, 3 and 4 were combined. Cluster 1 showed significantly higher scores on joint inflammation than the other groups combined ($t = 2.17$, $df = 141$, $p = 0.032$).

4.2.3 Relationship between the parental variables and the children cluster grouping (III)

Table 3 presents the descriptive statistics of the parental variables. A cut-off score of eight or more on the anxiety (HADS-A) or depression (HADS-D) subscale indicates an anxiety or a depressive disorder, respectively (Zigmond & Snaith, 1983). Seven percent of the parents scored above the cut-off for anxiety disorder, and twelve per cent for a depressive disorder. The mean BDI II rating for depressed mood was 6.20 ($SD = 6.18$, range 0–28). According to the cut-off points given in the BDI II (Beck, 2004) manual, 14.8 per cent of the parents had mild and 3.5 per cent had moderate depression.

On average the parents of the current sample were not depressed and their anxiety symptoms were not particularly high. However, some parents showed a pattern of distress symptoms and illness-management varied in the parents' sample (Table 3). Also the bivariate correlations between the parental and child variables raised the question, which parental variables discriminate between the four cluster groups of children.

The 13 parental well-being- and coping-related variables were used in the discriminant function analysis as predictors of the division of the children among the four cluster groups, and transformed into z-scores. Cluster 1 again differed from the other groups (Fig. 6).

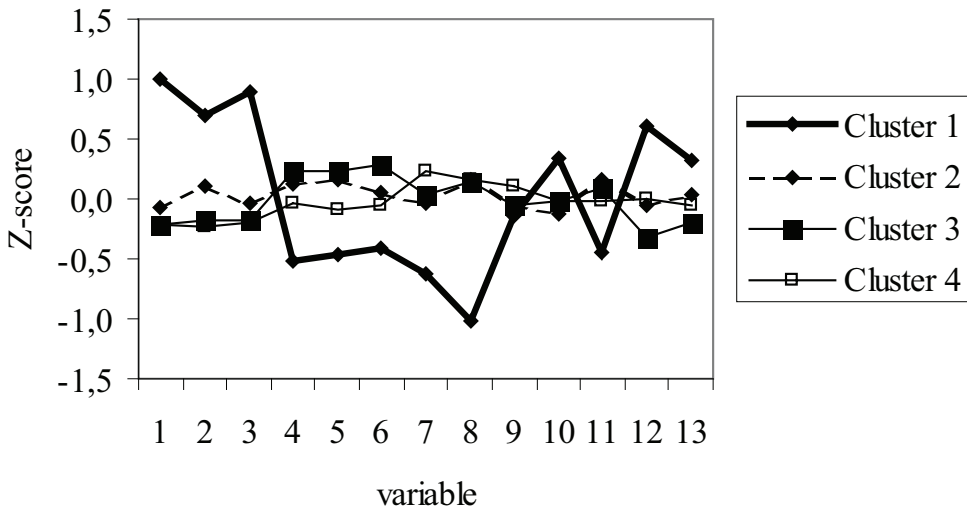


Fig. 6. Anxiety, depression and age in the four cluster groups according to 13 parental indicators.

Equality of the group means was tested by means of ANOVA, which revealed significant between-group differences in six of the 13 variables: BDI, HADS-A, HADS-D, Inf-disease, Inf-mood and Pain intensity (VAS) (Table 5).

Box’s M-test was significant ($p < .001$), indicating that the covariances between the predictive variables were not equal across the groups. This appeared to be attributable to two variables, influence-disease and influence-mood. When the analyses were repeated without them, the Box M was non-significant. However, the main result of the discriminant analysis did not change after removal of these two variables. Furthermore, all the variables in the discriminant analysis were normalised using four different methods, and this did not change the results.

The first discriminant function significantly separated the cluster group termed “teenagers scoring highly on trait anxiety and depression” from the other three groups. The other discriminant functions were not statistically significant. The high-loading variables were parental depressive symptoms (according to the BDI total score and the HADS D-score), parental anxiety symptoms (HADS A-score), parental self-efficacy with regard to somatic symptoms in the child, and the following illness-management items: parental perception of their influence on the child’s mood and disease, parental pain-intensity score (VAS), and disease severity. The first function was named “parental distress and deterioration in illness management”. Total discrimination was only modest. The first canonical correlation was 0.54 and the first function accounted for about 71 per cent of the variance among the groups: 42.3 per cent of the parents were correctly reclassified in their respective groups.

The cross-validated grouping was 30.3 per cent showing some over fitting in the linear discriminant analyses. The parents of the children in cluster group 1 achieved the largest mean score in the first discriminant function.

In conclusion, a set of parental variables was identified that discriminated between the cluster groups of children with JIA. Of these, parental depression, measured in accordance with the Beck Depression Inventory II (Beck et al., 2004), showed the best discriminant validity. The set was named “parental distress and deterioration of illness management” and was related to the wellbeing of the JIA children. Specifically, adolescent distress, somatic complaint and pain were associated with parental deterioration in illness management. A vulnerable group of children was identified.

Table 5. Description of parameters in the different cluster groups.

a) Disease related parameters

b) Variables related to the parental well-being in the cluster grouping of children.

	Cluster1	Cluster2	Cluster3	Cluster4	<i>p</i> *
	M(SD)	M(SD)	M(SD)	M(SD)	
a					
Disease duration (0-14)	5.8(4.1)	5.3(2.5)	5.4(3.0)	7.4(4.1)	0.012
Pain (0-19)	6.5(4.6)	3.5(3.8)	1.7(2.8)	4.0(4.7)	0.002
CHAQ (0-1.5)	0.5(0.5)	0.3(0.5)	0.2(0.3)	0.1(0.3)	0.003
Active joint count (0-20)	3.3(4.6)	1.1(1.8)	1.9(4.1)	1.6(1.6)	0.114
Somatic complaints (0-13)	5.8(3.0)	4.4(2.7)	2.9(2.7)	3.3(2.3)	0.000
CASEsom (1-5)	3.3(0.9)	3.4(0.8)	4.1(0.8)	3.9(0.8)	
CASEpsy (1.3 -5)	3.0(0.9)	3.5(0.9)	4.1(0.8)	4.9(0.8)	
CASEsoc (1.3 - 5)	3.6(0.7)	4.3(0.7)	4.7(0.5)	4.4(0.7)	
b					
BDI (0-28)	12.6(7.8)	7.8(5.0)	4.9(5.3)	4.9(5.6)	0.000
HADS-A (0-18)	6.4(3.6)	4.7(2.9)	3.9(2.5)	3.7(8.9)	0.005
HADS-D (0-15)	4.9(3.8)	2.3(2.5)	2.0(2.4)	1.9(2.4)	0.000
PASEsom (1.8 - 7)	3.8(1.2)	4.5(0.9)	4.6(1.0)	4.3(2.0)	0.056
PASEpsy (1.2 - 7)	4.3(1.3)	5.0(2.0)	5.1(1.0)	4.7(1.3)	0.064
PASEsoc (1.8 - 7)	5.0(1.4)	5.5(0.9)	5.8(1.1)	5.3(1.2)	0.121
Inf-disease (1-10)	3.5(2.0)	5.0(2.5)	5.1(2.0)	5.6(2.4)	0.016
Inf-mood (1-10)	4.9(2.3)	7.1(1.5)	7.1(1.7)	7.1(1.7)	0.000
Inf-treatment (1-10)	6.0(2.1)	6.2(1.8)	6.2(1.9)	6.5(2.0)	0.763
Pain-intensity (0-100)	31.1(23.8)	16.3(23.2)	10.3(13.2)	17.6(22.9)	0.017
Severity (1-10)	6.7(2.5)	5.7(1.8)	5.9(2.1)	5.9(2.0)	0.398
Coping (1-10)	6.8(1.8)	7.9(1.3)	7.8(2.0)	7.6(2.0)	0.164
General well-being (1-10)	23.4(19.3)	17.6(21.6)	13.2(14.5)	16.0(20.6)	0.368

p<.05*, p<.01**, p<.001***. The range of the scales are presented in brackets. Abbreviations in Table 3.

4.3 Self-efficacy (I, II, III, IV)

4.3.1 Validation of the scales

4.3.1.1 Parental self-efficacy scale (PASE)

Factor analyses were conducted in order to investigate whether the original two-factor loading pattern also fitted a sample from the Finnish population. No meaningful factor structure by EFA emerged from a two-factor solution, the same as in Barlow's original study (Barlow et al., 2000). In particular, items 8–14, which reflect the psychosocial subscale, did not emerge as one coherent factor. The items measuring somatic symptoms (items 1–7) loaded quite well onto one coherent factor, with the exception of item 7, which did not load onto it at all. The next step was to carry out CFA. First, in order to confirm the somatic dimension identified in the EFA we tested its fit with item 6 (fatigue): the statistics showed a poorer fit with than without this item. Secondly, tests were carried out on various factor models consisting of two or three latent dimensions, somatic, psychological and social. Finally, three three-factor models were tested in order to confirm their clustering into three domains, one somatically, one psychologically and one socially oriented. The fit statistics did not support the first of the three three-factor models (somatic dimension 1–6, psychological 7–10, and social 11–14), but they did in the second ($df/\chi^2 = 1.6$, RMSEA = 0.078, CFI = 0.96). However, item number 7 was excluded and did not load onto any factor. All the items were included in the third model, and the fit was good ($df/\chi^2 = 1.9$, RMSEA = 0.089, CFI = 0.94). This model was then used for further analysis of the data. Two cross-loadings were included in this final PASE-scale structure solution. First, item 6 (fatigue) loaded onto the psychological ($\lambda = 0.5$) and the somatic ($\lambda = 0.24$) dimension and was therefore included in the former. Secondly, item 11 (pleasure) loaded onto the somatic and social dimensions with an equally strong coefficient. The decision to include this item in the socially-oriented dimension was based on its semantic properties in the Finnish version of the PASE scale. In the final solution the latent dimensions were named "PASEsom, self efficacy with somatic symptoms" (items 1–5), "PASEpsy, self-efficacy in psychological functioning" (items 6–10) and "PASEsoc self-efficacy in social functioning" (items 11–14). The naming was based on the original qualities of the items loaded onto these dimensions. The reliability coefficients for these sum variables (dimensions), measured in terms of internal consistency, were $\alpha = 0.84$ for PASEsom, $\alpha = 0.88$ for PASEpsy, and $\alpha = 0.93$ for PASEsoc.

The final three-factor model comprising somatic, psychological and social dimensions was qualitatively and quantitatively satisfactory. It confirmed the existence of three separate domains

in parental self-efficacy. These factors were fairly strongly inter-correlated $\phi = 0.56\text{--}0.74$, however indicating that the theoretical limits for factor inter-correlation $\phi = 0.90$ were not exceeded (Aroian, & Norris, 2005). Two error-term correlations were found: items 12 and 13, included in the social dimension, correlated in terms of measurement error with item 8, included in the psychological dimension ($r = -0.22$ and $r = 0.20$, respectively).

4.3.1.2 Children`s self-efficacy scale (CASE)

In line with previous studies (Barlow et al., 2001), the first step in the CFA of the CASE scale was to test whether a three-factor model would fit the data. The final three-factor model with dimensions corresponding to the PASE scale reflecting self-efficacy in somatic, psychological and social functioning fitted the data quite well. However, item 4 (swollen joints/relief) had a stronger loading with self-efficacy in psychological functioning ($\lambda = 0.60$) than with self-efficacy in somatic functioning ($\lambda = 0.24$), and was therefore included in the psychological dimension. This differs from Barlow`s solution (Barlow 2000). The difference may be attributable to the final wording of this particular item in the Finnish questionnaire.

The latent dimensions were termed “CASEsom, self-efficacy with somatic symptoms” (items 1–3), “CASEpsy self-efficacy in psychological functioning” (items 4–7), and “CASEsoc self-efficacy in social functioning” (items 8–11). The reliability coefficients for the sum variables (dimensions), measured in terms of internal consistency, were $\alpha = 0.77$ for CASEsom, $\alpha = 0.80$ for CASEpsy and $\alpha = 0.79$ for CASEsoc. The CASE factors inter-correlated $\phi = 0.54\text{--}0.66$, but the inter-correlations were acceptable and below 0.90 (Aroian & Norris, 2005).

Univariate statistical analysis showed that the mean scores on all the subscales of the child and parental self-efficacy scales were fairly high, in particular with regard to social self-efficacy (Table 3). The patterns among parents and children corresponded. The mean scores (SD), measured on a scale from one to seven, for parental self-efficacy with somatic symptoms, in psychological functioning and social functioning were 4.7(1.26), 4.9(1.28) and 5.3(1.14), respectively. Measured on a scale from one to five for child self-efficacy the mean scores were 3.7(0.80), with somatic symptoms in psychological functioning 3.7(0.87) and in social functioning 4.2(0.74). The mothers showed stronger self-efficacy than the fathers on the following dimensions: somatic symptoms $M_m = 4.6$, $M_f = 3.9$, psychological functioning $M_m = 4.9$, $M_f = 4.8$ and social functioning $M_m = 5.4$, $M_f = 5.2(0.87)$. The difference was significant ($df = 117$, $t = 2.29$, $p < 0.05$) on the somatic dimension, but not on the others.

In conclusion, the refined three-factor structure of the PASE scale and the slightly modified three-dimensional CASE scale were found to be robust, thereby facilitating disease-specific analysis of somatic, psychological and social self-efficacy, and comparisons between patients and parents.

4.3.2 Summary

Self-efficacy among the children was investigated in the light of the cluster grouping. The overall level of difference between the groups was significant (Wilk's lambda = 0.708, $F = 5.73$, $df1 = 9$, $df2 = 338.44$, $p < 0.001$), as were the differences analysed separately ($p < .001$) in all the three variables (CASEsom, CASEpsy, CASEsoc). Cluster group 1 showed an unfavourable pattern: all the self-efficacy factor scores were the lowest (Table 5). The effect sizes ranged from 0.11 to 0.18 on the classical eta-squared measure, thus ranging from moderate to large according to Cohen's (Cohen, 1988) classification. The observed power varied between 0.97 and 0.99.

Self-efficacy was low in the cluster groups with the highest levels of anxiety and depression (groups 1 and 2), and high in those with lower levels (groups 3 and 4) (Table 5). Some of the medical parameters (pain, active joint count, somatic complaints) were also quite good in the groups in which self-efficacy was high and anxiety/depression was low.

The bivariate correlations between the parental-self-efficacy subscales and the child variables revealed interesting patterns (Table 3). Of the parental subscales PASEsom correlated significantly and negatively with child pain and active joint count, and positively with CASEsom, CASEpsy, CASEsoc. PASEpsy correlated significantly negatively with CDI, somatic complaints and active joint count, and significantly positively with CASEsom, CASEpsy, CASEsoc. PASEsoc correlated significantly negatively with child pain, CDI, somatic complaints and pain intensity (VAS), and significantly and positively with CASEsom, CASEpsy and CASEsoc. It was notable that there were no significant associations between parental self-efficacy, and anxiety and functional disability in the child, and that the bivariate correlations between the parental self-efficacy subscales and child pain were quite weak, although two of them were significant.

When parental variables were analysed in the discriminant analysis (Figure 6, Table 5), of the self-efficacy subscales PASEsom showed the best discriminant validity of the subscales. The first discriminant function significantly separated one cluster group, "teenagers scoring high on trait anxiety and depression", from the other three, and parental self-efficacy with regard to the child's somatic symptoms (PASEsom) was among the variables loading onto the first function. The parental BDI (Beck et al., 2004) proved to have the best discriminant validity. Furthermore, parental depressive symptoms were associated with PASEsom and these two with some other variables formed the first

function “Deterioration of illness management” in the discriminant analysis in the current sample. Parental deterioration in illness management was specifically characteristic of cluster group 1 in that the first discriminant function significantly separated it from the other clusters.

When factor analysis for all the scales used was done, parental self-efficacy subscales loaded into one coherent factor and child self-efficacy subscales into two separate factors. When regression analysis was done using these factor scores, parental self-efficacy did not emerge as a significant component in predicting the child’s pain, but the CASE subscales had explanatory power.

4.4 Parent and child interaction patterns and pain prediction (IV)

Following the identification of several indirect associations between the parent-reported variables, pain and the child-reported variables more specific analyses of these associations were carried out. The aim was to explore the characteristics of and the associations between both sets of variables (parental and child variables) in order to better understand how these might explain pain in JIA. An explorative design was chosen. The purpose was to investigate whether a higher-order factor solution would emerge comprising both parental (anxiety and depressive symptoms, illness management, and self-efficacy) and child (trait-anxiety, depression, functional disability, somatic complaints and self-efficacy) variables, and if so, to test whether such a factor (or factors) explained the variance in children’s pain. The specific aims were: (1) to study the interaction between the parent and child variables and (2) to investigate the extent to which these dimensions are related to the variance of pain in JIA.

The first step in addressing the research questions involved factor analysis. Both sets of variables were included as input variables in the EFA. Several factor models were tested and it was concluded that a six-factor model fitted the data best. The factor-score correlations ranged from 0.03 to 0.49. Table 6 shows the factor-loading matrix, the total variance explained being 52.5 per cent. Six subscales were created based on the factor scores, the internal consistency or reliability of which ranged from satisfactory (0.65) to very good (0.90).

The first subscale included items measuring parental depression and anxiety, and was named “Parental distress”. The second comprised parental items measuring self-efficacy and four items measuring illness management, and was named “Parental self-efficacy”. Thirdly, “Parental and child perceptions of the child’s functional capability” included both parent and child items and incorporated parental perceptions of the child’s general well-being and of how well he or she could influence the course of the disease, and the child’s estimate of his/her functional disability (CHAQ). The fourth

factor comprised parental perceptions of the severity of the child’s disease and of the number of active joints, and was named “Disease severity”. The fifth factor, which included the child variables somatic complaints, anxiety and depression, and the social and psychological self-efficacy subscales (loading negatively), was named “Child distress”, and the sixth, which included the child’s age and the CASEsom item, was named “Child somatic self-efficacy”.

The sample size in this study did not support the use of a Structural Equation Modeling framework in testing the parental variables as mediators of the child’s pain. Instead, the six factor scores were used in a multiple regression analysis to determine how well this set of factors predicted pain in children with JIA. The variance explained was 31 per cent ($R^2 = 0.31$, $R^2_{Adj} = 0.28$, $F = 10.13$, $df1 = 6$, $df2 = 133$, $p < .001$). Of the six, three factors predicted pain significantly: Parental and child perceptions of the child’s functional capability ($\beta = 0.42$, $p < .001$), Child distress ($\beta = 0.25$, $p < .01$), and Child somatic self-efficacy ($\beta = 0.19$, $p < .05$). The independent explanatory power for the other three was fairly weak.

Table 6. Factor Loading Matrix.

The items that were non-specific to several factors were included in the one with the highest loading.

	1	2	3	4	5	6	Pain(SPQ)total score
							r
HADS-A	0.87						0.13
BDI	0.83						0.26 **
HADS-D	0.83						0.19
PASEsoc		0.73					-0.20 *
PASEpsy		0.70					-0.10
PASEsom		0.68					-0.17 *
Inf-mood		0.56					-0.10
Coping		0.56					-0.24 **
Inf-treatment		0.53				0.33	0.07
Inf-disease		0.46	0.43				0.04
General well-being			0.69				-0.46 ***
CHAQ			0.58				0.31 **
Severity				0.57			0.11
Joint activity				0.29			0.16
Anxiety					0.89		0.23 **
CDI					0.79		0.29 **
CASEsoc				-0.31	-0.38		-0.34 ***
CASEpsy					-0.36	0.36	-0.13
SomComp(CBCL)					0.32		0.37 ***
CASEsom						0.59	-0.15
Age(child)						0.46	0.16

r= zero-order correlations between criterion variable pain and parental and child variables

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

The factors were named: 1=Parental distress, 2=Parental self-efficacy,

3=Parental and child perceptions of the child’s functional capability

4=Severity of the disease, 5=Child distress, 6=Child somatic self-efficacy

Investigation of the zero-order, bivariate correlations and the result of the regression analysis revealed an interesting pattern (Table 7). The factor score of Child somatic self-efficacy and the pain-outcome variable did not correlate significantly (zero-order correlation coefficient, $r = 0.08$), although this factor emerged as a significant predictor in the regression. Furthermore, a significant negative correlation was found between the factor scores reflecting Child somatic self-efficacy and Child distress ($r = -0.31, p < .001$). Moreover, the factor reflecting Child distress correlated significantly with the pain variable (zero-order correlation coefficient $r = 0.33, p < .001$), and it also appeared to be a significant predictor.

Table 7. The results of the multiple regression analyses of pain in children using the child and parental variables.

Criterion variable	Predictor	<i>Beta</i>	<i>Sig.</i>	Zero-order correlation
Pain	Parental distress	0.05	n.s.	0.19*
	Parental self-efficacy	-0.00	n.s.	-0.15
	Parental and child perception of child's functioning	0.42	0.001	0.49**
	Severity of the disease	0.00	n.s.	0.13
	Child distress	0.25	0.006	0.33**
	Child somatic self-efficacy	0.19	0.016	0.08

$$R^2=0.31, R^2_{Adj}=0.28$$

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

5 Discussion

On the whole the children in this study were found to cope quite well despite their disease. The level of pain was not alarmingly high, self-efficacy was good, and neither severe depression nor anxiety was detected. However, there were cases and groups showing some interesting patterns, which are worth addressing. There were four main findings. First, it seems that factors other than disease activity explain variance of pain in JIA. The pain is not intensive but the frequency is the critical aspect. Secondly, it seems that children, specifically adolescents, who are vulnerable in that they have distress symptoms, have parents with corresponding patterns of vulnerability. The third main finding concerns the role of self-efficacy, specifically that the child's self-efficacy has explanatory power in explaining pain. The overall role and mechanisms of self-efficacy in a family unit with a JIA child is complex and should be discussed. Finally, it seems that the family as a system plays a role in chronic childhood illnesses such as JIA. Most notably, parental and child perceptions of the child's functional capability, child's distress and child's somatic self-efficacy had an indirect effect on parental distress as an explanatory factor of the pain variation.

5.1 Pain

The treatment of JIA in Finland is centralized in a few hospitals or knowledge centres, it is intensive and the pharmacological treatment of articular manifestations is aggressive. This is a possible explanation for the fact that the intensity of patient pain in the current study was generally low. As a result of the effective treatment disease activity and pain levels have decreased, which indicates that factors other than disease activity explain the variance in pain. Other published Scandinavian studies on JIA concern children who were more severely affected by their arthritis and the disability indices were noticeably higher than in the current sample (Sällfors et al., 2003; Sällfors et al., 2004). According to earlier results, children with JIA tend to experience mild to moderate pain on the 0–10 VAS scale, ranging on average from 0.2 to 3.94 (Barlow et al., 2002; Hagglund et al., 1995; Malleson et al., 2004; Shaw et al., 2006a, 2006b; Thastum et al., 2005; Sällfors et al., 2002). The current sample falls within these boundaries. However, these children with JIA do have pain symptoms, which seem to persist – as shown by the follow-up results on pain. The intensity of the pain (measured on the VAS scale) is not very high, but other aspects turned out to be critical.

The pain score of the current JIA sample did not correlate with disease activity. According to the reasoning put forward by Gatchel et al. (2007), pain symptoms may relate more to illness, which

is a subjective experience, than to disease, a biological event. Anthony et al. (2003) suggest that joint inflammation may originally precipitate the pain, but for it to become more chronic in nature, other factors are involved. There are many organic reasons for pain chronification in JIA: cartilage damage (the thinning of the cartilage leading to distension of the joint capsule), misalignment of joints due to prolonged inflammation, a lower pain threshold causing pain in JIA remission, and pain with little or no connection with JIA. Moreover, as Hogeweg et al. (1995a, 1995b) and Kuis et al. (1997) postulate, altered pain processing in JIA patients due to prolonged activation of nociceptive systems remains a possible explanation to the results of this study. This suggestion also fits well with the bio-psycho-social model referred to (Waddell, 1984, 2004).

On the psychological level it is important to produce a precise description of a child's pain. How the child and the parents experience it, and how it is related to their lives and their cognitive attributions? As Home et al. (2006) argue, healthcare professionals need to advise children and parents on the evaluation of pain symptoms. They will benefit from being given a precise description of the pain that remains in spite of the fact that the disease might be in remission. Pain associated with the recurrence of joint inflammation may be predictable, and easier to comprehend and manage. With regard to acute nociceptive pain, clear diagnosis is often possible with little disagreement among the involved parties. However if there is no single cause, patients may be offered a variety of diagnoses. Lack of consistency and clarity may be distressing for families because they may worry that the "real" cause of the child's pain has not been established (Main et al. 2008; Kashikar-Zuck et al., 2006). Clinicians would thus do well in keeping in mind the fact that pain sensation in JIA patients may relate to factors other than tissue damage. Such factors are equally acceptable. They should simply lead to different treatment methods. Health care professionals should keep in mind that pain is a complex and subjective experience that occurs in a psychosocial context. Regardless of the etiology, pain symptoms should never be minimized or disregarded (Kashikar-Zuck, 2006). Both parents and children on a clinical visit related to arthritis of the child expect somatic evaluation and the exclusion of specific disorders. However, according to Koninjenberg et al. (2004), discussion of psychological investigations is best set in motion in the first meeting with the patient and his/her family, and should be initiated by paediatricians. This requires an interdisciplinary approach in which any explanation from the bio-psycho-social model of pain is equally appreciated and offered at an early stage of the disease. According to the results of this study, parental acceptance of a bio-psycho-social model of illness is crucial for the resolution of pain in JIA.

An important aspect in the current study is pain assessment: the picture of pain depends on how it is assessed. Huguet & Miro (2008) point out a problem regarding grading the severity of chronic

pain in children: such children do not constitute a homogenous group. They claim that published epidemiological studies have not provided information about the variation in the severity of pain conditions. They also propose a new classification system of pain in children based on persistence (the presence of chronic pain), intensity (average intensity in the three months before the interview) and impact (pain-related disability). Von Korff et al. (2000) also stress the importance of defining pain persistence, such as chronic pain, suggesting that assessment over a period may be more effective than measurement at a particular point of time. They recommend measuring both intensity and interference in conjunction with activities in clinical and healthcare services. In a recently validated pain experience questionnaire (PEQ), proven to be a valid and promising multidimensional pain assessment instrument, both pain intensity and pain frequency were included in pain severity subscale. Moreover, pain interference subscale covered suffering from pain and pain interference with regard to school work (Hermann, 2008). The scale, targeted at several juvenile pain patient groups including JIA, would have been an appropriate instrument for the purposes of the current study also.

Studies in the adult population (Elliott et al., 1999) and epidemiological studies on children (Mikkelsen et al., 1997) define chronic pain not only by duration but also by episode frequency. This classification and definition of pain persistence may shed new light on aspects that affect the severity of chronic pain.

In line with Huguet & Miro's reasoning (2008), it could be claimed that the results of this study regarding pain frequency, in terms of persistence for example, were significant and could be compared with the findings of epidemiological studies. Pain frequency was assessed by a structured pain questionnaire on five frequency levels over the previous three months. Interesting results emerged showing that fairly few children had been pain-free: 46 per cent had had pain at least once a week in at least one body area. This frequency assessment gives information about the location of the symptoms and the persistence of the pain, but not about the intensity. The structured pain questionnaire thus may not have been the best instrument for capturing pain in specific joints, which could be considered a limitation of the study. However, the questionnaire did reveal a new aspect of pain in the sample: children with JIA may have musculoskeletal pain other than that related to the condition. These children had more pain symptoms than Finnish schoolchildren in general (Mikkelsen et al., 1997). As Huguet & Miro (2008) suggest, this persistent musculoskeletal pain could be classified as "severe" in that it may interfere with the children's lives.

As reported earlier by others, pain symptoms during childhood are fairly common (El-Metwally et al., 2004). El Metwally et al. (2004) showed in a population-based study that 32 per cent of Finnish schoolchildren experience musculoskeletal pain, which persists, 54 per cent of them still

had pain at the one-year follow-up. McGhee et al. (2002) showed that musculoskeletal pain was the most common complaint for which children were referred to a paediatric rheumatologist, and it was present in approximately 50 per cent of new referrals.

Huguet et al. (2009) recently found a relatively high prevalence of pain in a community sample of schoolchildren between the ages of eight and 16. 37 per cent of them had chronic or recurrent pain. Chronic pain was defined as pain of three months or greater duration. In order for the pain to be defined as recurrent the children had to have reported at least one or two pain episodes at the same location every month during the three-month follow-up period. The definition was taken from Mikkelsen et al. (1997). The findings reported by Huguet et al. (2009) were comparable to the ones reported here on account of the same frequency definition of pain. On the other hand, the authors admit that their definition was relatively liberal, which possibly increased the number of children classified as having chronic or recurrent pain.

One important aspect related to chronic or recurrent pain and its impact on functioning concerns its cognitive appraisal. Studies conducted by Thastum et al. (2005) covering a sample of children with JIA, and by Huguet et al. (2009) on a sample of schoolchildren with pain symptoms, produced divergent results, particularly with regard to the cognitive appraisal of pain. Healthy schoolchildren were more convinced of their ability to control pain than the children in the clinical sample, and they were more likely to report that their emotions affected their pain. In the current sample, self-efficacy could be understood as a factor measuring cognitive appraisal, although it is not exactly the same theoretical concept. The current results complement those in Thastum et al. (2005) and Huguet et al. (2009) suggesting that conviction of being capable, in other words self-efficacy (Bandura, 2004), may be associated with low levels of pain, functional disability and somatic complaints.

In the current study, it was evident that pain in JIA may have multiple aetiologies. There are several possible interacting causes and modifiers, which may illustrate the condition more clearly. Identification of the interactive background factors might help the child and the parent involved in the pain management to comprehend, adhere to and master the pain situation. In JIA patients assessing both pain intensity, frequency and aspects regarding pain interference are warranted.

5.2 The vulnerable patient and the parent

The second major finding of the study was that children, and specifically adolescents, who are vulnerable when they have distress symptoms, may have parents with a corresponding pattern of vulnerability. A group of vulnerable adolescents was distinguished. Its members suffered from emotional distress that was not attributable to disease activity. Although the level of distress was

not notably high, it was related to unfavourable characteristics such as higher rates of pain and somatic complaints, and a higher disability level.

The characterisation of vulnerability is a major theme in this thesis in that it shows that pain is not a straightforward process. Explained in terms of bio-psycho-social mechanisms it seems to be much more complex. Pain also turned out to be related to parental factors. Projective processes among family members, particularly between children and parents, may enhance and maintain the experience of pain and of related behaviour patterns. According to family-system theory (Kerr, 1988; VanEcke et al., 2006; Charles, 2001; Cox & Paley, 1997), parents may transmit and project their emotional problems onto a child. As reported in this thesis, parental distress, parental self-efficacy with regard to the child's somatic symptoms, and the following illness-management items – parental perceptions of their influence on the child's mood and disease, parental estimate of child's pain intensity, and the disease severity of the child – relate to adolescent vulnerability.

In general the children showed no signs of severe depression or a severe level of anxiety symptoms, but some did show signs of trait anxiety and depressive symptoms. On average the CDI score for the current sample was lower than has been reported in migraine, chronic-fatigue (Smith et al., 2003), arthritis (Wagner et al., 2003) and pain-clinic patients (Kashikar-Zuck et al., 2001). Trait anxiety measured in accordance with STAIC/STAI (Spielberger et al., 1973, 1983) was comparable with samples in Barlow's (2001) study. On average the level of trait anxiety in this sample was lower than in juvenile primary fibromyalgia (Conte et al., 2003), migraine, or chronic fatigue syndrome (Smith et al., 2003). It could be argued that since chronic pain is often considered a precursor of depression more strongly than vice versa (Main et al. 2008), the fairly low level of pain, even though recurrent in the current sample, did not predispose the patients to depressive symptoms. However, there is strong evidence that to some extent depressive symptoms can explain pain-related functioning. It is possible that such symptoms in JIA patients may lead to depressive actions such as changing management of the disease (Gauntlett-Gilbert et al., 2007; Goubert et al., 2006; Kashikar-Zuck et al., 2000, 2001; Logan et al., 2006; Merlijin et al., 2003). Kashikar-Zuck et al. (2006) point out that underestimating child's pain often results in more severe expressions of distress, which often complicates the clinical picture. Thus, in the current sample of children with JIA and their parents it was relevant to proceed to a more comprehensive analysis of depressive and anxiety symptoms.

The classification results in the current study were in accordance with those reported in previous studies on JIA. Generally children manage quite well with their disease on psychological level, but some do have difficulties. When the four-cluster groups were analysed against self-efficacy, new information on their relationships emerged. It could be expected that the pain-vulnerable cluster

groups in the current sample were groups 1 and 2 because their self-efficacy was low. Disease activity was fairly high in cluster 3, with the highest self-efficacy, but the children in this group did not suffer pain or trait anxiety. Self-efficacy may regulate anxiety and depression so that good self-efficacy will help the patients to reduce their pain experience. Low self-efficacy may aggravate anxiety and depression and thus increase the pain. Some of the medical parameters (pain, active joint count, and somatic complaints) were quite good in the groups in which self-efficacy was high and anxiety and depression low. However, cluster group 1, which topped the levels of depressive and anxiety symptoms in this sample, also had high scores for pain and somatic complaints, and the lowest self-efficacy among these cluster groups. Interestingly, in the disease-activity parameters (active joint count) there were no significant differences between group one and other three groups.

The results related to distress are consistent with the results of previous studies in showing that slight depression or distress were associated with pain and vulnerability. Martin et al. (2004) pointed out that internalising symptoms in young people, particularly adolescents, are likely to be overlooked by adults. Sensitivity in distinguishing these features is required, and we cannot assume that a child's pain symptoms or somatic complaints could be eliminated purely through medical treatment of the disease. Indeed, children with JIA seem to fall into subgroups, and some of them are at risk of adapting unfavourably to their disease and life situation.

It is important to consider the age of the child when planning treatment and rehabilitation. The age range of the sample in the present study covers two different groups: children and adolescents. Recently, age has not been a central focus of interest in pain research, and on JIA-related pain in particular. Age was a good categorising variable in the *k*-means cluster analysis in that it identified a different psychological profile in one of the adolescent cluster groups (Cluster group 1). For instance, distress symptoms were stronger than they were in groups with a lower mean age, and this distress had unfavourable associations with other clinical parameters. However, another cluster group with approximately the same mean age (Cluster group 4) could be characterised as having a favourable variable profile. These results are strongly in accordance with clinical findings. Youth is a life period during which JIA patients are expected to learn to manage their illness themselves, usually with less parental support. They may also face challenges on the road to independence and identity consolidation (Barlow et al., 2004). Some show resilience and some find it difficult to take responsibility for their disease, e.g. to manage their own medication. According to Beales et al. (1983), growing adolescents become more aware of their internal sensations (e.g., pain in inflamed joints) and of the potentially serious consequences, and this may increase the distress evoked by the symptoms. Adolescents are better able than young children to conceptualise pain, and they have

a deeper understanding of the potential harmful effects of arthritis. They are also more willing to express pain in self-reports than younger children. In line with this reasoning, Malleeson et al. (2004) found a positive correlation between pain and age in juvenile patients with JIA. Further, when the children in the sample were stratified into two age groups, those aged 8–11 had lower pain scores than the 12–17-year-olds: the correlation between joint inflammation and pain was strong in children under the age of eight, and weak in older children (Malleeson et al., 2004). Moreover, according to Hagglund et al. (1995), age was not an independent predictor of pain, but simple correlations showed that children with JIA reported more pain as they grew. Age-related developmental challenges might produce an increase in depressive and anxiety symptoms in adolescence, which in turn might decrease overall wellbeing and adaptation to pain.

Hoff et al. (2006) studied longitudinal relationships between depressive symptoms and pain in JIA patients. She found that depressive symptoms predicted pain among children aged between eight and 17 years in six- and twelve-month follow-ups. However, she also found complex interrelationships between the disease and psychological factors associated with pain, and called for precise description of the level of depressive symptoms that influence current and longitudinal associations with pain. Other authors also recommend integrative approaches. Palermo & Chambers (2005) call for a comprehensive framework describing the role of parent and family factors, and encompassing consideration of both specific parent behaviour and broader aspects of family functioning in the face of children's chronic pain. Palermo & Eccleston (2009) stress the need for multiple approaches in order to enhance understanding of common parental reactions and the ways in which parent-child interaction changes in the face of pain.

The children's cluster grouping was investigated further by including parents in the analysis, the aim being to find out if parental characteristics could be identified as discriminant factors. "Parental distress and deterioration in illness management" turned out to be the discriminant function that was most strongly associated with the cluster grouping, with child distress and age as categorising variables. This factor characterises parental depressive and anxiety symptoms, and sense of helplessness regarding their child's functioning. Parental depression had the highest discriminant validity. This first discriminant function significantly separated cluster group 1, "Teenagers scoring high on trait anxiety and depression" from the other three groups. Thus, an aspect of childhood chronic illness in the family system, namely interdependence (Kerr, 2010; VanEcke et al., 2006; Charles, 2001; Cox & Paley, 1997) was distinguished. According to Bowen (Kerr & Bowen, 1988), family projection processes may impair the functioning of the children and make them more vulnerable to clinical

symptoms. The implication is that family system theory would provide a very suitable treatment framework in clinical practice.

It has been shown in previous studies that a high level of parental distress and a sense of helplessness may affect the adolescent's functioning in a corresponding manner, thereby creating a vicious circle within the family (Barlow et al., 2004; Eccleston et al., 2004; Wagner et al., 2003). Increased levels of pain in the child usually also provoke more distress in both parents and child. Not necessarily specifically the pain severity is the crucial aspect affecting on parental well-being. When parental distress was assessed in a sample of juvenile pain patients (Hermann et al., 2008), it was found to be independent of pain severity, but child's behavior problems and social behavior had greater impact on parental distress.

The current result regarding the vulnerability of both parties (child and parent) is significant given the overall very low level of distress symptoms in the sample compared to a corresponding study (Barlow et al., 2000). It can be concluded that distress in a child with JIA tends to be associated with distress in the parents. However, it is important to understand that the result regarding parental vulnerability, although interesting and promising, does not allow reporting of any causal associations. In terms of clinical practice, effort should focus on the key question of coping with and managing the disease, and on improving the often complicated and stressful life-situation with which families with JIA have to cope.

Compared to the study by Barlow et al. (2001), the parents in this study had considerably fewer depressive and anxiety symptoms. An interesting result was that only nine per cent of them suffered from depressive symptoms, and the majority had strong self-efficacy and a fairly strong sense of illness management. There may be several explanations for this. The fact that the majority of the parents and children participating in the study were not newcomers to rehabilitation might have had some effect. Most of the parents had participated in a variety of interdisciplinary treatment protocols that target disease management, adherence, self-efficacy and illness management, and it could be hypothesised that such interventions had been effective. Another contributing factor could be the fact that the pharmacological treatment of the articular manifestations of JIA has become more aggressive and is initiated earlier in the course of the disease, thus reducing its morbidity. This may also have psychological effects on preventing pain chronification at an early stage. However, improvements in pharmacological treatment do not necessarily ease the JIA-related strain on children and their parents. As shown in the current sample, a single factor such as the number of active joints falls short in explaining the child's pain, and even more so the distress of the parents. The clinical impression of the author is that parents still consider JIA as a severe illness.

5.3 The role of self-efficacy

With regard to the third main finding it was emphasised that the child's self-efficacy has predicting power in explaining his/her pain. Self-efficacy may thus have a role as a mediating variable in chronic childhood illnesses such as JIA. It was not possible to verify the influence of parental self-efficacy, although it was associated with corresponding aspects in the child. Its effect in predicting pain could be indirect.

Self-efficacy, a traditional psychological theory-based concept, did fit well in the theoretical framework of the current research project. It has not been studied intensively in samples comprising children, but it is more familiar in research on adult health (Brekke et al., 2003; Lorig et al., 2003; Saunders, 2004; Vancouver et al., 2001). I utilised the concept because I wanted to clarify the possible effects of the attributions of the child and his/her parent on pain, and to better understand the adherence and non-adherence. Given that the self-efficacy concept originated in social-cognitive theory, I hoped that it would encapsulate and explain the personal aspect of having a chronic illness and possibly pain, and also some of the social aspects in terms of how and under what conditions self-efficacy remains strong, and how parental and child self-efficacy are related.

The aim to understand the bio-psycho-social model of pain (Fig. 1) emphasised the need to clarify the factors and the processes underlying its dimensions, e.g., psychological and social aspects. Rhee et al. (2000) proposed a model suggesting that self-efficacy has an indirect effect on pain and depression, in other words it mediates between them in arthritis patients. They studied adult patients with rheumatoid arthritis and found that rehabilitation helped them to improve their coping strategies in terms of increased self-efficacy, and that the positive changes led to a decrease in pain and depressive symptoms. In a recent study Dahlbeck & Lightsey (2008) distinguished a relationship between anxiety and self-efficacy in children with a chronic illness. Turner et al. (2007) found that the perceived ability to control pain explained the greatest proportion of the effect of cognitive-behavioural therapy. However, self-efficacy had a unique mediating effect, independently of the other process variables, on pain in the therapy. The authors concluded that efforts to increase patient self-efficacy in managing pain might have unique additional benefits. They also clarified the pathways along which psychological interventions bring about this change. There are only a few published studies on this subject in the area of paediatric rheumatology (Barlow et al, 2000; Barlow et al. 2001; Porter et al., 2008). According to family system theory (Kerr, 2010; VanEcke, et al., 2006; Charles, 2001; Cox & Paley, 1997), parents and children may project emotional problems on each other, and this in turn increases the vulnerability of the family members. Strengthening self-efficacy might break this vicious circle and ease the projection, which, after all, is the objective. Barlow et al.

(2000) also showed that in the JIA context parental self-efficacy was associated with child wellbeing in many ways. Porter et al. (2008) recently introduced the term self-efficacy dyad referring to a parent and a child. This co-variation in parent and child self-efficacy over time could be a beneficial point of focus in that self-efficacy is a context-related concept in the development of which parents are very influential. The concept of the self-efficacy dyad fits into the family-system framework on which this thesis is based, and is worthy of further study.

Given the patient group addressed in this thesis, Bandura's (1997) theoretical claims may have many practical applications. A JIA patient who doubts his/her capabilities or has low self-efficacy beliefs in terms of managing aspects of JIA may shy away from any disease-related challenges and difficulties. He/she may experience difficulty in finding the motivation to engage in physiotherapy, back down in the face of obstacles, and have low aspirations and weak commitment to the goals of the rehabilitation. A taxing situation easily leads a person with low self-efficacy to dwell on his/her personal deficiencies and the difficulty of the task. This perturbed thinking may undermine any efforts to overcome personal deficiencies. On the other hand some patients are persistent: they manage well with their long-term disease, and in spite of active joint inflammation they keep up with their school routines, for example. This could reflect strong self-efficacy, which facilitates the setting up of personally adequate goals for dealing with the challenges. In this sense strong self-efficacy is strongly causal.

In the current sample distress symptoms were associated with lower self-efficacy, which is in accordance with the results of a fairly recent study in which a new pain-self-efficacy scale was validated (Bursch et al., 2006). This finding is important because it highlights the association between distress symptoms and cognitive attributes (e.g., self-efficacy). The regression analyses reported in this thesis identified three factors that predicted pain to a significant degree: Parental and child perceptions of the child's functional capability, Child distress, and Child somatic self-efficacy. Of these three, the self-efficacy subscales were included in Child distress and Child somatic self-efficacy. This result was expected due to the fact that the CASE scale proved to have good validity in previous stages of the project, and the associations with other variables were expected and logical. The variables CASEsoc and CASEpsy loaded onto the same factor as anxiety/depressive symptoms and somatic complaints, and these together explained pain. These results correspond with the findings reported by Bursch et al. (2006), and are in line with self-efficacy theory.

According to Bandura (1977), self-efficacy is a stress-processing factor and a change mechanism. Improved self-efficacy has also proved to be a sign of improvement in rehabilitation (Barlow et al., 2001; Kaminski et al., 2006). One implication of the present results is that child self-efficacy may

regulate anxiety, depression and other clinical parameters such that a high level could help the patients to deal with their pain and disease activity, thus lowering their level of anxiety and depression. This study supports the use of children's self-efficacy for such a purpose.

An interesting finding regarding self-efficacy relates to the PASE scale. Of the parental subscales (somatic, psychological and social), only the first was a significant discriminator in the allocation into different cluster groups. The other subscales were not significantly discriminative in the cluster grouping of children, which might relate to the low overall level of depressive and anxiety symptoms in this sample of parents. Correspondingly, the levels of the self-efficacy subscales were also high. These results do not correspond with those of Barlow et al. (2000) on parental self-efficacy in JIA: parental depression and anxiety were higher and self-efficacy was lower. It would be interesting to carry out discriminant analyses in that earlier sample, in particular to see if the discriminant function worked better if the level of parental distress symptoms was higher.

Parental self-efficacy did not predict child pain in the regression analysis. This is in line with the findings reported by Bursch (2006). Although in her study the parents assessed their child's self-efficacy, and in this thesis the parents assessed their own, Bursch's results and the present ones can be generalised. She argues that parents are forced to assess their child's self-efficacy based on the child's external behaviour or symptoms. A parent may be less sure of what the child "can do" if he/she does not go to school because of the illness or the pain, for instance. Children are more adept at assessing their own capability. Self-efficacy, which measures belief about what one is "capable of doing", is, after all, difficult to estimate. In line with this reasoning, for instance one of the questions on the parental self-efficacy scale in the current project, "How certain are you that you can do something to help your child to deal with the frustration of arthritis?" was possibly not specific enough, or not personal enough. Many intervening facts could affect the answer, such as how well the parent knows the child's emotional needs. Thus the current results showing little evidence of the predictive value of parental self-efficacy with regard to their child's pain, are understandable and acceptable.

Given the results discussed above, I suggest that rehabilitation programmes for children with JIA should focus on their sense of management. Individuals make efficacy judgements from past performance of a task, the performance of a similar task by others who are judged to be similar to oneself and verbal encouragement from others (Turk, 1996). Thus, self-efficacy-enhancing sessions in which children and parents receive information from other people, including professionals, would be beneficial (Bandura, 1977).

The present results could be of use in planning and targeting treatment. However, it should be noted that assessment of self-efficacy is closely related to actions on some occasions, and unrelated

on others (McCracken, 2005). Self-efficacy beliefs are rooted in personal development and the social systems, which the person belongs to, and are not necessarily easy to change. The concept could thus be seen as a variable that may change over time and under specific circumstances, such as rehabilitation. Understanding the contextual meaning of self-efficacy and its association with psychosocial development is important in the planning of treatment, rehabilitation and research.

Although parental self-efficacy proved not to predict child pain, the crude correlations between child and parental self-efficacy were significant and in the expected direction. I would therefore suggest that, given the observed interaction between the parental and child variables, we also need to focus on parental wellbeing and capability when managing the child's disease.

5.4 Predicting pain through interaction between the parent and the child

The fourth main finding of the study emphasises the role of the family system: parental and child perceptions of the child's functional capability, the child's distress and somatic self-efficacy, and an indirect effect of parental distress predicted pain variation in JIA. The bio-psycho-social conceptualisation (Gatchel, 2007; Waddell et al., 2004) of pain turned out to be adequate in this sample of JIA children. Factors other than disease activity, psychological and social aspects of pain and the illness, had stronger explanatory power.

The first step towards understanding the variance of pain in JIA was to ascertain whether there was any interaction between the parent and the child variables, and to investigate the extent to which such patterns explained pain. The factor structure did indicate interaction between the variables, but not very clearly. They did not load onto the same factors to the extent that was expected according to previous studies (Eccleston et al., 2004; Goubert et al., 2006; Logan et al., 2005). Nevertheless, both sets of variables were included in one factor score, "Parental and child perceptions of the child's functional capability". In the regression analysis this factor was the strongest predictor of the child's pain. Thus, in the current study the family as a system was a meaningful theoretical framework, although further studies are needed in order to shed light on reciprocal family and parent-child dyadic effects on pain. Another theoretical argument related to "Parental and child perceptions of the child's functional capability" concerns the locus of control. The factor included the following items: current parental perceptions of their influence on the child's disease, incorporated parental perceptions of the child's general well-being and of how well he or she could influence the course of the disease, and the child's estimate of his/her functional disability (CHAQ).

The zero-order correlations and regression analyses produced a very interesting result that was not related to the interaction between the parent and child variables, but which highlighted how complicated the interactions between somatic symptoms, distress and pain might be. The child's somatic self-efficacy appeared to be a predictor of pain, although there was no crude correlation between the two. The most likely cause was the significant crude correlation with the "Child's distress". Multicollinearity, which in one case concealed the effect of one variable on the others and in another case strengthened it, turned out to be an indicator of direct and indirect links between the variables predicting pain. Moreover, parental distress did not have independent explanatory power in predicting child pain, even though there was crude correlation between these variables. Thus, the results show that pain in JIA cannot be properly understood in terms of direct associations between the outcome variable and the potential explanatory variables.

Recent research has identified a very important aspect of research on pain in children. Palermo & Chambers (2005) and Eccleston & Chambers (2009) call for integrative approaches in which individual-level, dyadic and family-level variables are recognised and distinguished when testing pathways to pain and disability. The reciprocal influence of the pain and its accompanying levels of disability, child/parental factors in interaction on all levels of the individual dyad, and family variables as they relate to pain should all be investigated. The current study was aimed at such a direction, and the results gave support to the idea of understanding, studying and treating pain in JIA in a very integrative manner. Pain experienced by a child with JIA should be viewed as a multidimensional problem affected by both disease-related somatic factors and psychosocial factors of both the patient and parents. The relations between the parent-child variables are complex and they affect pain both directly and indirectly. Child's vulnerability variables, namely somatic complaints, anxiety and depression, and the social and psychological self-efficacy subscales (loading negatively) named as "Child's distress" did explain pain, as well as "Child somatic self-efficacy" did. JIA is still a severe disease because of its unpredictable course: it may expose the sufferer to chronic pain, and it may limit daily activities and social participation, and interfere in children's lives. Modern treatment should involve an interdisciplinary team equipped to address all areas of normal growth, social development and physical functioning, as well as to prescribe pharmacological treatment to limit chronic joint pain, inflammation and damage.

On the theoretical level, the finding of associations between parental and child variables support the idea of treating families with JIA as a systemic unit. It could be argued that family projection, as defined by Bowen (Kerr, 2010), could be a theoretical framework within which to conduct further studies on pain in JIA.

In sum, several aspects of the disease and its management were investigated in order to shed light on the direct and indirect links between the parental and child variables in relation to JIA, and specifically to pain. Further studies are needed in order to develop a model based on the variables. According to Baron & Kenny (1986), establishing mediation between two or more variables requires four steps: showing that the initial variable is correlated with the outcome (e.g., child distress with pain); showing that the initial variable is correlated with the mediator (e.g., child's distress with self-efficacy subscales); showing that the mediator affects the outcome variable (e.g., self-efficacy subscales affect pain); establishing that the mediator completely mediates between the initial and the outcome variable – the effect of the initial variable (e.g., child distress with regard to pain) controlling for the mediator should be zero. Partial mediation is indicated if the first three steps are accomplished and complete mediation following completion of all the steps. It would be interesting to find a model that was consistent with the findings of this study. The results with regard to the first two steps are discussed in this thesis.

5.5 Strengths and weaknesses of the study

There were several strengths in the study. The sample size was relatively high and the fact that the informants were recruited from the Rheumatism Foundation Hospital in Heinola, and the Paediatric Rheumatology Clinic of Helsinki University Hospital in the Helsinki Metropolitan area ensured its representativeness in that the catchment area covered the whole country. Both patients and one parent from each family were included, which was a further strength. JIA predominantly affects girls, but there were some boys in the sample. The statistical analyses were carried out with thoroughness.

An obvious limitation is the cross-sectional nature of the data. Thus, the causal relations should be interpreted with caution. Furthermore, the study could be categorised as descriptive, non-experimental and exploratory, and should thus be seen as a starting point for further research. The study design was theory-driven, based on the literature covering the bio-psycho-social nature of pain, the social-context model of child adjustment to paediatric chronic physical illness, and family-system theory. The explorative nature of the study gave the researcher the theory-driven flexibility to distinguish “characteristics” emerging in the sample, an advantage over experimental design in terms of allowing new perspectives to open up.

Nevertheless, several concerns and weaknesses should be acknowledged. First, attention should be given to the scales measuring self-efficacy (Barlow et al., 2000; Barlow et al., 2001 and Brady et al., 2003). The validity of the scales is carefully documented in the first sub-study. Of the available scales

measuring both child and parental self-efficacy, Barlow's scales were selected based on their good coverage and validation. These instruments were found to represent internally consistent measures of disease-specific self-efficacy in a Finnish sample of JIA patients and parents. The present findings support previous work indicating adequate psychometric strength of the CASE scale (Barlow et al., 2001). The factor structure of the parental PASE scale (Barlow et al., 2000) was reformulated as a three-dimensional solution analogous to the CASE scale. The CASE subscales and the CDI scale were correlated in order to test the construct validity of the translated CASE scale, and the correlations were in the expected direction. There was a significant correlation between the PASE subscales and adult depression measured on the BDI II, which demonstrated the construct validity of the translated PASE scale. It should be noted that the PASE and CASE self-efficacy scales were designed for use in juvenile arthritis populations and targeted at the general management of the disease. In this sense they contained too few items specifically on pain-related self-efficacy. There are several self-efficacy scales for use with adults, most of which target specific beliefs such as pain-related self-efficacy, although there are some general scales (Brady, 2003). There are no scales targeted on children with JIA and on their pain-specific beliefs. The first arthritis self-efficacy scale developed for children (CASE) is not pain-specific, and thus in terms of content validity could have been tailored to meet the needs of the current study. However, it served well as a screening method for arthritis-related self-efficacy beliefs. Self-efficacy was correlated with the clinical data (disease activity, functional disability, number of active joints, pain and somatic complaints) in order to assess criterion validity, and all the correlations were in the expected direction. The PASE scale is the first self-efficacy scale to be developed for use with parents who have a child with JIA. However, it was not sensitive enough to discriminate between parents with strong and low levels of self-efficacy. Another limitation of PASE is that it may be too general to capture the child's experience of pain and parental response to it (Bursch et al., 2006). Thus the content validity of the PASE scale would probably have improved had there been more pain-specific items.

A second limitation is the absence of a gender perspective. It would have been possible to identify pain factors that relate to gender, and this would be a useful focus of interest in future studies. Also, multiple informants, including both parents in the study design, would have provided unique information. Specifically fathers might have given valuable information regarding the child's and the family's functioning. Moreover, qualitative data collection complementing the quantitative measures might have shed light on the children's and their parent's experiences. Comments were made, such as when a mother said, "Finally someone asks us parents how we feel", but given the study design they were not taken into consideration.

The children in the sample had had JIA for approximately six years, meaning that most of them, and their parents, had already consulted a primary care physician and/or another specialist, and had received treatment. This clearly affected the results, inevitably enhancing patient and possibly parental wellbeing. Furthermore, the parent sample was biased, and their average level of management on several measures, such as depressive symptoms, was very good. It should also be noted that positive bias with regard to the participants could have been a factor. Given that the principal investigator was, in some cases, the same person who had been treating the patients, the responses may have been altered even though the data-collection procedure was carried out with care and thoroughness.

5.6 Clinical relevance and future aspects

The results shed light on an important aspect of pain in children and adolescents with JIA, and presented new information about the pain and its effect on parents. Pain other than that related to disease activity was distinguished, and there was more of such pain in the current sample than in samples in population-based studies. Mikkelsson et al. (1997) and Hakala et al. (2002) argue that musculoskeletal pain symptoms are becoming more common among adolescents in Finland. Thus, healthcare providers in general practice, but specifically in tertiary care regarding JIA, should start tailoring services to this group. This is crucial in the case of JIA, because the family is expected to adhere to a laborious regimen over a longer period of time. Along the way, originally a minor somatic problem may increase in severity. Developing pain-treatment modalities that recognize the family as a system are warranted.

Of the factors that were related to pain in the current sample, the vulnerability factors and their interrelations partly confirm and replicate earlier findings, given the significance of the integrative approach in explaining the pain of the child with JIA. These results extend earlier findings, specifically in showing several indirect and direct child-to-parent or parent-to-child reciprocal effects. Vulnerability was connected to distress in the adolescents, and there was a corresponding pattern among their parents. Interdependence between children and parents, which was shown in the associations between the parental and child variables, means in practice that parental wellbeing is an important matter in treatment and rehabilitation of children with JIA. Appropriate early intervention and the provision of intensive rehabilitation in the case of severe-pain-associated incapacity are warranted.

This study is the first in Finland focusing on pain in JIA which includes self-efficacy in an explorative design. This thesis emphasises the importance of measuring and assessing self-efficacy given the implication that children's self-efficacy predicts the variance of pain. The significance of self-efficacy as a mediating variable with regard to parent-child functioning with the pain symptoms of

the child is worth further investigation. The results indicate that methods based on such a theoretical framework might be useful in pain rehabilitation among JIA patients.

The starting point for the study was the need to understand differences in adjustment to JIA among children and their parents, and specifically why some children suffer from pain and some do not. Given that pain is multi-factorial, there is no sense in trying to find single factors that might “cure” it. At the clinic it is important to distinguish signs of vicious circles in the management of the disease, specifically in parent-child interaction. These features complicate treatment and rehabilitation, and they may adversely affect the roles of both parent and child as agents of disease management. In fact, aside from the medical care, JIA healthcare should incorporate measures that are sensitive enough to indicate eventual depressive symptoms among children and their parents and the potential interactions. Child self-efficacy is a potential and cost-effective mechanism for enhancing rehabilitation in the management of the chronic illness.

6 References

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