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PATIENT EDUCATION AND FOOT DISABILITY IN JUVENILE IDIOPATHIC ARTHRITIS

A PHYSIOTHERAPY PERSPECTIVE

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To Sofie, Anna, Jocke and all other
children/adolescents with JIA

ABSTRACT

Introduction: Juvenile idiopathic arthritis (JIA) influences the child in different aspects and needs multi-professional treatment. Physical therapists' major goal is to maintain or increase physical activity, e.g. with education programs and custom-made foot orthoses. Assessment of treatment effects requires valid and reliable measurement that is sensitive to change.

Aims: The major aim of the present work was to assess the effects of an education program among parents of children with JIA and among adolescent with JIA, and the effects of custom-made foot orthoses among children/adolescents with JIA. A further aim was to develop and test measurement methods for assessing treatment.

Methods: Two new questionnaires were developed to cover relevant aspects concerning perception about managing the disease JIA, and concerning foot related disability. The validity and test-retest reliability of these methods were tested. The effects of an eight-hour multi-professional education program were assessed. An intervention group with 55 parents of children with JIA, and 11 adolescents, completed a questionnaire before, directly after and four months after the program. A comparison group with 18 parents and five adolescents completed the questionnaire twice times with a four-month interval. The effects of foot orthoses were studied in 48 children/adolescents with JIA with standardized tests of daily activities: here pain and/or capacity were assessed. The tests were performed with and without foot orthoses in two sets, respectively, in a randomized order. Analyses were also calculated for the different diagnostic subgroups and different foot malalignments.

Results: The questionnaire on perception of managing the disease had acceptable content validity, as was partly indicated in a factor analysis of medical, exercise, pain and social-support factors. This medical, exercise, pain and social support (MEPS) questionnaire seemed to be stable over time and sensitive to change to detect effects of the education program. The questionnaire on foot-related disability appeared to cover relevant aspects of domain of interest, appeared valid in terms of the underlying theoretical constructs and was stable over time. The education program had a significantly positive outcome in the groups of parents, most expressed in the medical and pain aspects. The effects were also significantly improved in relation to the comparison group. In the adolescents group, only minor improvements were found after the education program. Using foot orthoses resulted in significantly reduction of pain after standing, stair-climbing, walking and running, improved balance capacities and faster self-estimated walking speed than without foot orthoses.

Conclusions: In summary, the present investigation, including the special developed measurement instruments, demonstrated beneficial effects of the education program as well as of the use of foot orthoses. It is hoped that these methods will come to be included as a self-evident part of the treatment of JIA, in this way contributing to enhanced physical activity and decreased disability.

Keywords: Clinical trials, education program, foot orthoses, juvenile idiopathic arthritis, physical therapy, reliability, validity, questionnaire

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SAMMANFATTNING

Introduktion: Juvenil idiopatisk artrit (JIA) kan påverka barnet ur många olika aspekter och kräver ett multiprofessionellt omhändertagande. Sjukgymnastens mål är att öka förutsättning för fysisk aktivitet, t.ex. med patientutbildning och specialanpassade fotortoser. För att utvärdera effekten av behandlingar, krävs det valida, reliabla och sensitiva mätmetoder.

Syfte: Huvudsyftet med avhandlingen var att utvärdera effekten av patientutbildning hos föräldrar till barn med JIA och hos ungdomar med JIA, samt effekten av specialanpassade fotortoser hos barn/ungdomar med JIA. Ytterligare ett syfte var att utveckla och testa metodik för att utvärdera ovan nämnda behandlingar.

Metod: Två nya frågeformulär utvecklades för att täcka relevanta aspekter inom dels uppfattning om förmåga att hantera sjukdomen JIA, dels förekomst av fotrelaterade funktionshinder. Mätmetodernas validitet och test-retest reliabilitet prövades. Effekten av ett åttatimmars multiprofessionellt patientutbildningsprogram studerades. En interventionsgrupp med 55 föräldrar till barn med JIA, och 11 ungdomar besvarade ett frågeformulär före, direkt efter och efter fyra månader. En jämförelsegrupp med 18 föräldrar och fem ungdomar besvarade frågeformuläret två gånger med fyra månaders intervall. Effekten av fotortoser studerades hos 48 barn/ungdomar med JIA med hjälp av standardiserade tester av vardagsaktiviteter, då smärta skattades och/eller kapacitet mättes. Testerna utfördes med respektive utan fotortoser i två randomiserade set. Analyser genomfördes även avseende diagnostisk undergruppering respektive typ av fotfelställning.

Resultat: Frågeformuläret om förmåga att hantera sjukdomen hade god innehållsvaliditet, vilket bl.a. påvisades i en faktoranalys där fyra aspekter; en medicinsk, en träningsrelaterad, en om smärta och en om socialt stöd, identifierades. Formuläret benämndes MEPS (medical, exercise, pain and social support) och befanns även vara stabilt över tid samt sensitivt för att fånga effekter av patientutbildning. Frågeformuläret om fotrelaterade funktionshinder befanns täcka relevanta aspekter av aktuella egenskaper, vara valitt i relation till det underliggande teoretiska begreppet och vara stabilt över tid. Patientutbildningen hade en signifikant positiv effekt i föräldragruppen, mest uttalat inom den medicinska och smärtaspekten. Effekten var även signifikant i relation till jämförelsegruppen. I ungdomsgruppen påvisades endast enstaka positiva effekter av patientutbildningen. Vid användning av fotortoser påvisades signifikant reducering av smärta vid stående, trappgång, gång och löpning samt förbättrad balanskapacitet och högre självvald gånghastighet jämfört med då fotortoser inte användes..

Konklusion: Sammantaget har föreliggande arbete, med hjälp av specialutvecklad mätmetodik, påvisat goda effekter av såväl patientutbildning som fotortoser. Förhoppningen är att dessa metoder ska ingå som en självklar del i behandlingen vid JIA och därigenom bidra till fortsatt fysisk aktivitet och minskat funktionshinder.

Nyckelord: Fotortos, frågeformulär, juvenil idiopatisk artrit, klinisk studie, patientutbildning, reliabilitet, sjukgymnastik, validitet

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LIST OF PUBLICATIONS

This thesis is based on the following studies, which will be referred to by their Roman numerals.

- I. André M, Hedengren E, Hagelberg S, Stenström CH. Perceived ability to manage juvenile chronic arthritis among adolescents and parents: Development of a questionnaire to assess medical issues, exercise, pain and social support. *Arthritis Care Res* 1999;12:229-237.
- II. André M, Hagelberg S, Stenström CH. Education in the management of juvenile chronic arthritis. Changes in self-reported competencies among adolescents and parents of young children. *Scand J Rheumatol* 2001;30:323-327.
- III. André M, Hagelberg S, Stenström CH. The juvenile arthritis foot disability index: development and evaluation of measurement properties. *J Rheumatol* 2004;31:2488-2493.
- IV. André M, Hagelberg S, Stenström CH. Immediate effects of custom made foot orthoses in children with juvenile idiopathic arthritis. A clinical trial. Submitted

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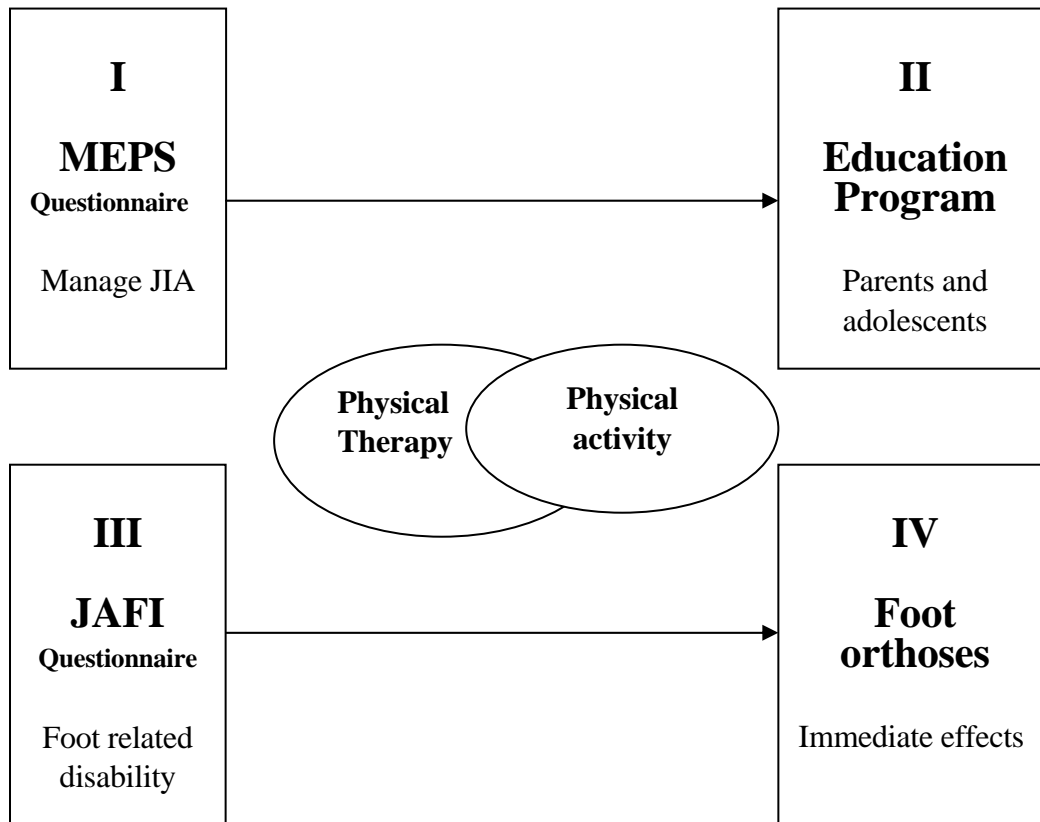
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LIST OF ABBREVIATIONS

ACR	American College of Rheumatology
ANOVA	Analysis of variance
CHAQ	Childhood Health Assessment Questionnaire
EULAR	European League Against Rheumatism
IBD	Inflammatory Bowel Disease
ICC	Intra-class correlation coefficient
ICF	International Classification of Functioning, disability and health
ILAR	International League of Associations for Rheumatology
JAFI	The Juvenile Arthritis Foot disability Index
JAS	Juvenile Ankylosing Spondylitis
JCA	Juvenile Chronic Arthritis
JIA	Juvenile Idiopathic Arthritis
JpsA	Juvenile Psoriatic Arthropathy
JRA	Juvenile Rheumatoid Arthritis
LSR	Legitimerade Sjukgymnasters Riksförbund
K_w	Weighted Kappa coefficients
MEPS	Medical Exercise Pain and Social Support
MTX	Methotrexat
NSAID	Nonsteroidal-anti-inflammatory drugs
RF	Rheumatoid factor
SEM	Standard error of measurements
TNF	Tumor necrosis factor
VAS	Visual Analog Scale
WCPT	The World Confederation of Physical Therapy
WHO	World Health Organization

THE PRESENT WORK



DEFINITIONS OF CONCEPTS

Physical activity is defined as any bodily movement produced by skeletal muscles that result in energy expenditure (Caspersen et al., 1985). Physical activity can be seen as an umbrella term for human behavior with many dimensions and sub-categories such as exercise, sport, leisure activities, dance, transportation etc (Corbin et al., 2000).

Physical fitness is defined as a set of attributes that individuals possess or achieve and that relate to their ability to perform physical activity (Caspersen et al., 1985).

In the present work *children* are people up to twelve years of age and *adolescents* are people aged 13-19.

1 INTRODUCTION

Physical therapy is based on the knowledge and study of human movement. It is an important part of the multidisciplinary treatment of children with juvenile idiopathic arthritis (JIA). Movement in JIA may be limited due to the inflammatory process and to various components of disability. The physical therapist's responsibility is to identify and improve the children's possibilities for movement. Pediatric physical therapists have specialized knowledge of children in general and of movement restriction in particular. This offers unique opportunities to prevent and reduce disability in children with JIA. Physical activity is one important aspect of movement often targeted in physical therapy. Two important interventions, aiming at improving physical activity, are patient education and the prescription of foot orthoses. However, their effects have not yet been proven, partly because of a lack of appropriate outcome measures in this area.

1.1 PHYSICAL THERAPY

Movement is the main concept of physical therapy, which has its root in medical science. Hislop (1975) defined physical therapy as: "A health profession that emphasizes the science of pathokinesiology and the application of therapeutic exercise for the prevention, evaluation, and treatment of disorders of human motion".

The World Confederation of Physical Therapy (WCPT, 1999) characterized physical therapy as "concerned with identifying and maximizing movement potential, within the spheres of promotion, prevention, treatment and rehabilitation involving the interaction between physical therapist, patients or clients, families and caregivers, in a process of assessing movement potential and in establishing agreed upon goals and objectives using knowledge and skills unique to physical therapists".

The Swedish Association of Registered Physiotherapists (Broberg, 1997) defined physical therapy as "a field of practice concerned with prevention, examination, treatment and rehabilitation of movement disorders that limit or threaten to limit the movement capacity of the individual." Development of methods and quality aspects, and evaluation of outcomes were acknowledged as integral parts of the field of practice.

1.2 INTERNATIONAL CLASSIFICATION OF FUNCTIONING, DISABILITY AND HEALTH

To provide a unified and standard language and framework for the description of health and health-related conditions, the World Health Organization has developed the International Classification of Functioning, disability and health (ICF) (WHO, 2001). Its structure offers the possibility to conceptualize several health status components, either in positive aspects under the umbrella term "functioning" including "body functions and body structures", "activities and participation", or negatively under "disability". The latter and its components "impairment", "activity limitation and participation restriction" will be used in the present work. All health conditions may also be associated with interacting "environmental factors" and "personal factors"

(Figure 1). The ICF model is useful in physical therapy to identify the components aimed at in different treatments or evaluations.

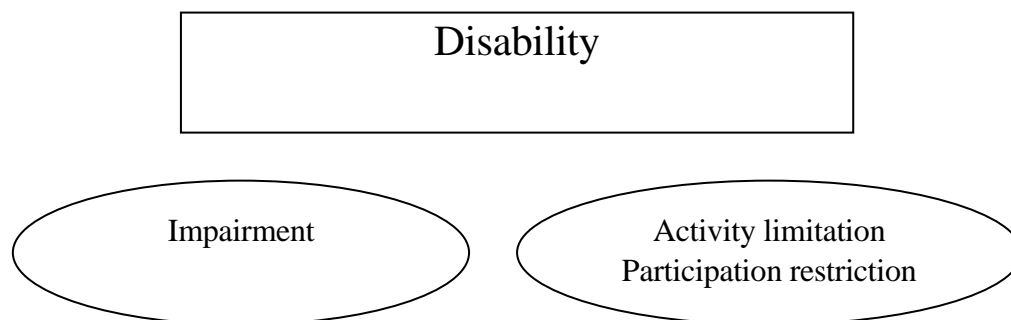


Figure 1. Components of the International Classification of Functioning, disability and health related to negative health aspects.

1.3 JUVENILE IDIOPATHIC ARTHRITIS (JIA)

Juvenile idiopathic arthritis includes a heterogeneous group of diseases. Their etiology is not yet fully established (Cassidy and Petty, 2002).

1.3.1 Terminology, Criteria and Classification

Diagnostic criteria are necessary, in research as well as in daily clinical work, especially when there are no specific laboratory tests available to ascertain a diagnosis. Different classifications of and sets of criteria for childhood arthritis have been used in different parts of the world. The term Juvenile Rheumatoid Arthritis (JRA) was established and classified by the American College of Rheumatology (ACR) in 1972 (Brewer, 1973). The European League Against Rheumatism (EULAR) established another classification system in 1977 under the term Juvenile Chronic Arthritis (JCA) (Wood, 1978). The heterogeneity of the disease and disagreement about diagnostic criteria have made interpretation and comparison of studies difficult. The first attempt to reach an international consensus came in 1995. The Pediatric Standing Committee of the International League of Associations for Rheumatology (ILAR) proposed a new set of criteria with the umbrella term Juvenile Idiopathic Arthritis (JIA) (Fink, 1995). The criteria were revised in 1997 and are known as the 'Durban criteria' (Petty et al., 1998). Seven onset types were classified and specified as to inclusion and exclusion criteria (Table I).

Table I. The EULAR, ACR and ILAR classification criteria for juvenile arthritis

	ACR (1973)	EULAR (1977)	ILAR (1997)
Terminology	JRA	JCA	JIA
Basis of classification	Clinical	Clinical and serologic (RF)	Clinical and serologic (RF)
Duration of arthritis	≥ 6 weeks	≥ 3 months	≥ 6 weeks
Age at onset	≤ 16 years	≤ 16 years	≤ 16 years
Subgroup included	Systemic Polyarticular Oligoarticular - - - - -	Systemic Polyarticular Oligoarticular - - JAS ¹ JpsA ² IBD ³	Systemic Polyarticular, RF positive RF negative Oligoarticular Persistent Extended Enthesitis-related arthritis JpsA Other arthritis

¹JAS= Juvenile Ankylosing Spondylitis; ²JpsA=Juvenile Psoriatic Arthropathy; ³Arthropathy associated with inflammatory bowel disease

1.3.2 Epidemiology

The use of different classification criteria has rendered it difficult to ascertain the incidence and the prevalence of the various types of childhood arthritis. European incidence rates for chronic arthritis in childhood indicate great variation from a low 1.3 (Priour et al., 1987) to a high 22.1/100 000 (Rygg et al., 2000). A recent population-based epidemiological study of JIA in the Nordic countries reported an incidence from Sweden of 14/100 000, while the disease affects about 2 000 children in Sweden (Berntsson et al., 2003). This tallies with a previous Swedish study (Andersson et al., 1992; Andersson, 1999).

Onset is possible at any age during childhood (rarely before six months), but there are peaks at ages one to three years and at eight to twelve years. The disease is more common among girls than among boys in a ratio of 2-3:1 (Cassidy and Petty, 2002).

The present work considers mainly children with three types of onset. “Oligoarticular” onset is most common in children with JIA and is present in almost 60 % of cases. This onset is characterized by involvement of four or less affected joints; half of the children have only one affected joint, primarily the knee and secondly the ankle (Andersson and Fasth, 1995; Sharma and Sherry, 1999). “Polyarticular” onset is the second most common onset, involving five or more joints and present in about 25-30 % of the cases (Berntsson et al., 2003). “Enthesitis-related arthritis” onset is characterized by chronic inflammatory arthritis of the peripheral and axial skeleton, frequently accompanied by enthesitis. This onset is present in about 2-4% of children with JIA (Berntsson et al., 2003).

1.3.3 Clinical symptoms

Arthritis is unpredictable across all onset types of JIA, but most commonly follows a fluctuating course characterized by periods of flares and remissions. Impairments in JIA that are associated with articular inflammation are joint pain, swelling, stiffness, tenderness, and limited joint range of motion (Cassidy and Petty 2002; Schneider and Passo, 2002). Other, more general, impairments are fatigue and weight loss, muscle weakness, and reduced physical fitness. Leg length discrepancy, muscle contractures and joint destruction are other impairments that may be related to an actively hard-treated disease of long duration (Lindehammar and Backman, 1995; Fan et al., 1998; Takken et al., 2003; Klepper, 2003). Activity limitation and participation restriction, including difficulties in daily life, decreased physical activity, and consequences for health-related quality of life have also been reported in children with JIA (Flatö et al., 2002; Klepper, 2003; Foster et al., 2003).

1.3.3.1 Pain

Pain is a major symptom of childhood arthritis. It is regarded as chronic and mainly of nociceptive origin, and is an important target for therapy. Further, pain is more prevalent than previously recognized. In one study 86% of children with JIA reported pain during a routine clinic visit (Sherry et al., 1990). Another study indicated that 60 % reported pain at disease onset and 50 % at a one-year follow-up. Forty percent continued to report pain five years later (Lovell and Walco, 1989). In both the above studies the children reported pain intensity as mild to moderate, but it has later been found that 25 % - 31% of children with JIA have moderate to severe pain (Schanberg et al., 1997, 2003).

The relations between gender and reported pain in children with JIA are unclear. Some studies report no difference between boys and girls (Vandvik et al., 1990; Cassidy and Petty, 2002), while others have found that girls report more daily pain symptoms and seem to have poorer pain coping efficacy (Schanberg et al., 2001; Sällfors et al., 2003). It has thus been suggested that pain management and training of pain coping skills should be specifically targeted (Schanberg et al., 2001).

Pain is complexly related to psychological and social processes and may be associated either with the inflammatory activity or the degree of disability; or it is caused by investigation or treatment procedures (Anthony and Schanberg, 2003). It affects the health status of children with JIA negatively (Schanberg et al., 1997), over half the

variation in the children's well-being being explained in a recent study by self-reported pain, number of pain free days and participation in physical education classes (Sällfors et al., 2004). The family's relationship, pain history and environmental variables also influence the children's pain reports and their ability to carry out activities of daily living (Varni et al., 1988; Gragg et al., 1996; Schanberg et al., 2001). However, children who rated their ability to control and decrease pain as high had significantly lower pain intensity ratings (Schanberg et al., 1997).

1.3.3.2 Foot disability

The main task of the feet is to balance the body's weight on a small surface, to transfer it, and to compensate the unevenness of the ground. The task is performed by a number of the small and large joints, with the help of a strong capsule apparatus and numerous muscles (Truckenbrodt et al., 1994).

Foot involvement is frequently described as a common clinical manifestation of JIA. Inflammation may affect all joints in the feet (Spraul and Koenning, 1994; Tong and Sartoris, 1996; Ferrari 1998) and cause pain and malalignment. The terminology and definitions of foot position and alignments differ in the literature, and are confusing. In the present work the following definitions are used: the "ankle joint" is the talocrural joint, the "rear-foot" is the talocalcaneal joint, and the "mid-foot" consists of the talonavicular, calcaneocuboideal and the inter-cuneiform joints. The "forefoot" is the metatarsal joints and the proximal and distal interphalangeal joints. Foot positions are defined as "normal", "valgoplanus" (pronated rear-foot and/or mid-foot) with overload at the medial rim of the foot and a flattening of the longitudinal and transversal arches (Figure 2 a) or "cavovarus" (supinated rear-foot) with overload at the lateral rim and heightening of the longitudinal arch (Figure 2 b).

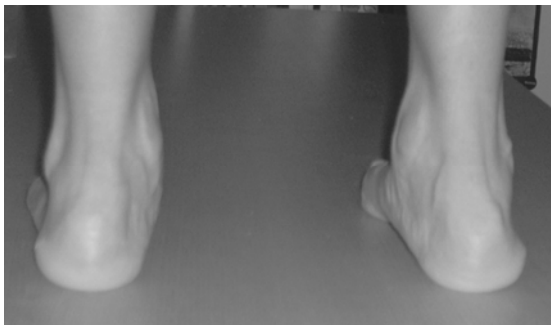


Figure 2 a. Valgoplanus foot position

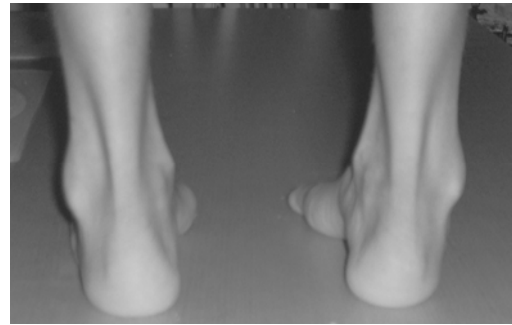


Figure 2 b. Cavovarus foot position

One study evaluated 144 children with JIA and categorized "foot problems" as inflammation, limitation of motion and malalignment. Ninety-four percent had one or more foot problems. The author also found that the pronated rear-foot (73 %) and mid-foot (72%) were the most common malalignments (Spraul and Koenning, 1994). It was early recognized that patterns of foot-joint involvement varied across different onset

types of JIA (Ansell, 1988). In oligoarticular onset the ankle is the most affected foot joint (Rana, 1982; Andersson and Fasth, 1995; Sharma and Sherry, 1999), while polyarticular onset most commonly affects the forefoot. Since inflammation and pain are the causes of all foot impairments, adequate pharmacological treatment is necessary, but the value of physical therapy has also been highlighted as equally important (Truckenbrodt et al., 1994).

1.3.4 Prognosis and Outcomes

The prognosis for children with JIA has improved during the past decade, but minorities of children still have an unremitting disease with severe disease activity (Andersson and Fasth; 1995, Minden, 2000; Milojevic and Ilowite, 2002; Flatö et al., 2002). However, the results vary between studies, maybe depending on differing diagnostic criteria and study designs. A recent review of health outcome in children with JIA suggested that 30-35% had gone into remission 10 years after onset, but this varied considerably with disease onset type (Duffy, 2004). One earlier study estimated that 50% of all children with JIA will recover completely (Andersson and Fasth, 1995). Children with oligoarticular onset have the best prognosis, with about 50 % remitting (Andersson and Fasth, 1992; Flatö et al., 2002), while a less favorable prognosis has been reported for polyarticular and systemic onsets (Andersson and Fasth, 1995; Lomater et al., 2000). Factors such as gender, disease duration and different joint involvements also influence prognosis and outcome (Andersson and Fasth, 1995; Flatö et al., 2002).

Outcome may be described (in adult rheumatology) in terms of death, disability, dollars, drug side effects and discomfort (Fries et al., 1980). Disability (activity limitation) and discomfort are the only sections that have been adapted for children and used in a questionnaire included in a suggested core set of outcome measures (Giannini et al., 1997). A long-term follow up (14.9 years), showed poorer physical health among participants with active JIA than among controls, but also in participants with disease in remission (Minden et al., 2000). Reports of children's activity limitation reflect varying results: 36-60 % of the children investigated reported activity limitations (Andersson and Fasth, 1995; Flatö et al., 2002). In one study despite relatively good physical health, poorer general health and health-related quality of life were found compared with controls (Foster et al., 2003). However, another study reported higher health-related quality of life for children with JIA versus controls (Ruperto et al., 1997). Maybe the conflicting findings reflect both the use of different measurements and the heterogeneity of the disease.

Symmetric arthritis, hip joint involvement and female gender predict disability (Flatö et al., 2002), but note that present-day treatment can, it is hoped, improve outcomes in the future (Bowyer et al., 2003).

1.3.5 Treatment

1.3.5.1 Medical treatment

The treatment in JIA is directed at minimizing inflammation and disability. Major advances have been achieved in medical treatment during the past few decades

(Milojevic and Ilowite, 2002). More effective use of available drugs and the application of new ones have undoubtedly improved the outcome of JIA (Murray and Lovell, 2002). The new therapeutic approach with anti-tumor necrosis factor (anti-TNF) agents has reportedly shown excellent results for children with polyarticular onset of JIA (Lovell et al., 2003). However non-steroidal anti-inflammatory drugs (NSAID) are still in use for most children with JIA (Cassidy and Petty, 2002) and Methotrexate (MTX) is the most common second-line agent (Giannini et al., 1992, 1993; Singsen and Goldbach-Mansky, 1997; Woo et al., 2000; Ruperto et al., 2004). Intra-articular corticosteroid injection is also commonly used for children with JIA to supplement other drugs. It is reportedly safe and effective (Huppertz et al., 1995; Padeh and Passweel, 1998; Sherry et al., 1999; Hagelberg et al., 2000; Ravelli et al., 2001; Broström et al., 2004). Oral corticosteroids have side effects such as growth retardation and osteoporosis if used long-term (Bowyer et al., 2003; Murray and Lowell, 2002), but are still employed in very active disease.

1.3.5.2 Physical therapy

Physical therapy in JIA has developed much during the past few decades. Previous emphasis on rest, passive hands-on treatment and restriction of physical activity has given way to the present considerably more active approach. This approach was developed along with improved medical treatment and better outcome in pediatric rheumatology (Flatö et al., 2002), but was also based on the emerging evidence of the safety and benefit of physical activity in adult rheumatoid arthritis (Stenström and Minor, 2003), and more recent evidence on the benefits of physical activity in children with JIA (Klepper, 2003). Modern physical therapy includes a wide variety of treatment approaches such as education, exercise, and manual techniques. All have the goal of maintaining or improving the children's physical activity and reduce disability.

Children with JIA are less physically fit than healthy children. Lower aerobic capacity (Takken et al., 2002), significant muscular deficits (Lindehammar and Backman, 1995; Lindehammar and Sandstedt, 1998; Hedengren et al., 2001) and poorer scores on standardized fitness tests (Fan et al., 1998; Wessel et al., 1999) have been described. These impairments are possibly related to low levels of physical activity as well as to pain, fatigue and stiffness (Singsen, 1995; Takken et al., 2003; Klepper, 2003).

Such children also have more activity limitation and participation restriction in strenuous activities than do healthy peers (Henderson et al., 1995). A recent study found significant relationship between daily physical activities, parental rating of their children's physical activity, and aerobic capacity (Takken et al., 2005). A wide range of physical activity levels was also found in the study group, and this corresponds well with the clinical impression.

Research results conflict as to the effectiveness of physical activity and exercise for children with JIA. However, some studies have very small samples, and this might bias the outcome. Benefits on range of motion and muscle strength were found after a summer camp, but these improvements were not maintained at follow-up four months later. No reduction of activity limitation was found (Milliet et al., 1996). In a recent randomized controlled study no significant changes in physical fitness, joint score,

activity limitation, or health related-quality of life were found after an aquatic program (Takken et al., 2003). Nor were significant changes in VO_{2peak} found following an aerobic exercise program, although improvements in gait were found (Feldman et al., 2000). There is seemingly more evidence regarding the effects of muscle training. Positive effects have been reported using pool-exercise and land-based resistance exercise for both muscle strength and muscle endurance (Öberg et al., 1994; Fisher et al., 1999; Venkatraman et al., 1999; Klepper, 1999)

While clear evidence on the benefit of physical activity is still lacking, there seems to be enough research to support its safety in children with JIA. No detrimental effects have been found on pain or inflammatory activity (Feldman et al., 2000; Wright et al., 2000; Fischer et al., 1999). This supports international recommendations on physical activity for children with JIA, which include individualized resistance exercise three times weekly to improve muscle strength and endurance and 30 minutes of moderately intensive physical activity at least three times weekly to decrease pain, joint swelling and tenderness and to improve aerobic capacity (Work group recommendations 2002).

Children with JIA wish to be equal to their peers (Sällfors et al., 2001) and it is thus important to enhance their possibilities for frequent physical activity in their social contexts. This may be promoted by e.g. education programs and foot orthoses.

1.3.5.3 Patient education

In adult rheumatology patient education began with joint protection and energy conservation as early as in the 1960s (Corderoy, 1965). Subsequent education interventions targeted self-management involving not only the transmission of knowledge but also the use of multiple strategies to foster healthy beliefs, behavior, and skills (Lorish and Boutaugh, 1997; Brady et al., 2003). Considerable scientific evidence indicates that patient education in the rheumatic diseases can result in positive changes that affect health status and health-related quality of life (Lorig et al., 1987; Tucker and Kirwan; 1991; Hirano et al., 1994; Lindroth et al., 1995).

In pediatric health care the families' resources and coping skills are critical to adaptation to chronic illness (Harper, 1991; Frank et al., 1998). In children with asthma and diabetes, education programs are beneficial as regards knowledge, behavior, stress coping, fewer physician visits and a reduced effect of the disease on daily life (Hackett et al., 1989; Evans et al., 1991; Mesters et al., 1995; Matthews et al., 1998).

Lack of information about the disease and disability, misunderstandings due to insufficient or inappropriate information, and lack of perceived control may predict poor adherence to treatment regimens in pediatric rheumatology (Rapoff, 1989, 1997). It is for this reason that education programs for children with JIA and their parents aim at influencing knowledge, skills, self-efficacy, and coping strategies (Rapoff et al., 1985, 1988; Harris et al., 1991). A multidisciplinary team approach is necessary, to meet the needs of the children and their families (Varni et al., 1988; Konkol et al., 1989; Emery and Bowyer, 1991; Harris et al., 1991; Ansell BM 1994). A need to include parents in interventions to improve children's abilities to cope with e.g. pain in JIA has been suggested (Schanberg et al., 2001). Factors, which may optimize

children's mobility and autonomy of activities, are recommended as important targets for such education (Bartholomew et al., 1994). Further, managing the school environment to facilitate optimal participation and promote the skills to handle pain and stiffness have been identified as important for improving health and health-related quality of life (Schanberg et al., 2003).

Perceived needs for family-based education for children with JIA have been expressed (Konkol et al., 1989; Bartholomew et al., 1994; Hagglund et al., 1996), few of if any studies evaluate the effectiveness of multi-disciplinary education programs for these people.

1.3.5.4 Foot orthoses

The effects of foot orthoses have been studied in adult rheumatology with differing results. Some uncontrolled studies have found clinical benefits (Marks and McKendry, 1996; Fransen and Edmonds, 1997; MacSween et al., 1999), while a randomized, controlled, long-term study showed a reduction in foot disability, including pain, when using custom-designed foot-orthoses (Woodburn et al., 2002). However, another randomized, placebo-controlled trial identified no clinical benefits (Conrad et al., 1996).

Custom-made foot orthoses are frequently recommended and prescribed to children with JIA (Truckenbrodt et al., 1994; Ferrari, 1998; Fairburn et al., 2002). The goal is to reduce pain and to facilitate the foot position in order to improve the possibility for physical activity. However, their effects have not yet been investigated in JIA.

1.3.6 Assessment

1.3.6.1 In JIA

Outcome in clinical trials in pediatric rheumatic diseases previously focused mainly on the measurement of inflammatory activity, which does not necessarily reflect the children's disability and may furthermore not be sensitive enough to detect important changes (Duffy and Duffy, 1997). Formerly, physical therapy assessment focused solely on impairments such as muscle function, joint range-of motion, and aerobic capacity. Assessment of activity limitation was not generally suggested until 1997 when it was added to the recommended "core set" (Giannini et al., 1997). Participation restriction is still not much considered in the evaluation of children with JIA, but might include school attendance, independence, and leisure time activities such as sports and socializing with peers.

Impairment measurement methods include pain rating scales, joint scores, goniometers for range-of-motion, myometer or manual tests of muscle function, or bicycle ergometer tests of aerobic capacity. Activity limitation measurement methods might include standardized tests of capacity such as walking, running, stair-climbing, balance tasks or questionnaires on performance. The most common and diagnose-specific questionnaire is the Child Health Assessment Questionnaire (CHAQ). This instrument is based on the Health Assessment Questionnaire developed in adult rheumatology (Fries et al., 1980) and was modified for children (Sing et al., 1994; Ruperto et al.,

2001). The CHAQ has been translated into many languages, including Swedish (Andersson et al., 1993), but its sensitivity to change in rehabilitation, partly because of possible ceiling effects, has been discussed (Ruperto et al., 1999; Dempster et al., 2001; Lam et al., 2004; Brunner et al., 2005).

Knowledge, skill and self-efficacy are important areas for children and their parents in their management of JIA, but no applicable measurement methods for this existed. Questionnaires for assessing foot functioning and disability have been developed for adults with rheumatic diseases (Budiman-Mak and Roach, 1991; Sundbom and Stenström, 2003), but no questionnaires were available for children with arthritis.

1.3.6.2 Measurement properties

Measurement properties include reliability, validity and the ability of an assessment method to detect changes. A reliable measure must provide consistent values with small error of measurement, but also be able to differentiate among subjects. Validity is the umbrella term employed for the degree to which a measure assesses what it is intended to measure. The terminology has developed and the definitions of Finch et al (2002) are used in the present work.

Three types of reliability impact most frequently affect clinical practice; namely “internal consistency”, “test-retest reliability”, and “interrater reliability” (Finch et al., 2002). Validity may be divided into e.g. “face validity”, “content validity”, “criterion validity” and different aspects of “construct validity”. “Content validity” refers to the extent a measure is composed of a comprehensive sample of items that completely assess the domain of interest. In the absence of a gold standard, “construct validity” involves forming theories about the attribute of interest and then assessing how far the measure under investigation provides results that are consistent with the theories. “Convergent” and “divergent” construct validity concerns how far the result of a measure agrees/disagrees with that of another measure that is believed to be assessing the same/another attribute. “Known group” construct validity refers to validation of two or more groups that represent different levels of the attribute of interest (Finch et al., 2002).

1.3.6.3 Agreement between parents and children reports

Many children with JIA are very young, and relevant clinical information must often be obtained from the parents. Maturity and understanding of the construction of a questionnaire are important factors in the choice of measurement methods for children. It has been suggested that a full understanding of measurement is not attained until 9-10 years of age (Champion et al., 1998). The VAS is commonly used as a pain measurement, but also as part of comprehensive questionnaires evaluating other constructs. In the ‘core set of outcome measures’ in JIA the VAS is used to evaluate two of six components (Petty et al., 1998) and also for two variables, pain and global assessment, in CHAQ.

It is thus important to know whether there is agreement between parents and children in different ratings or measurements. Results from previous research are conflicting. Excellent agreement has been found between parents’ and children’s rating of activity

limitation (Duffy et al., 1993; Singh et al., 1994; Lam et al., 2004), while recent studies found discrepancies between the parents' and the children's reports (Palermo et al., 2004). However it seems that agreement in ratings of pain intensity is more difficult to obtain (Brunner et al., 2004), maybe because pain is an individualized and subjective event, and usually parents rate less pain than their children (Doherty E et al., 1993; Sawyer et al., 2004).

1.4 RATIONALE FOR THE THESIS

Physical therapists support physical activity in the daily life of children with JIA. Education programs and foot orthoses are used to enhance this, but their effects are poorly evaluated and there is a lack of specific measurements to evaluate them.

2 AIMS

The overall aim of the work reported in this thesis was to evaluate the outcome of education for children with JIA and their parents, to evaluate the effects of foot orthoses for these children, and to develop valid and reliable measure for these purposes.

Specific aims were:

- I to develop and to test for validity and reliability a questionnaire for use in assessing adolescents' and parents' perceived ability to manage JCA,
- II to evaluate changes in self-reported competencies following an education program among the parents of children with JCA and among adolescents with JCA,
- III to develop a questionnaire for assessing foot related disability among children and adolescents with JIA, and to test it for validity and reliability,
- IV to evaluate the immediate effects of custom-made foot orthoses in children with JIA with different types of malalignments and for different diagnostic subgroups.

3 METHODS

3.1 PARTICIPANTS

Most participants in the present work were recruited from the Department of Pediatric Rheumatology at Astrid Lindgren Children's Hospital, Karolinska University Hospital, Stockholm. In Studies I and II participants from other pediatric departments at nearby hospitals were also included. The children were diagnosed according to the EULAR classification system and belonged to the subgroups oligoarthritis, polyarthritis, and juvenile ankylosing spondylitis (JAS). In Studies I and II the term juvenile chronic arthritis (JCA) was used, whereas in Studies III and IV juvenile idiopathic arthritis (JIA) was used.

Two hundred seven children and adolescents with JIA, 124 represented by their parents, and 29 healthy controls participated in the present work. The children's and adolescent's age varied between two and 19.5 years (Table II) and their disease duration ranged from six months to 16 years. The participants in Studies III and IV had had foot arthritis during the previous six months. Those in Study IV were eight years or older and had been prescribed custom-made foot orthoses.

Table II. Demographic data on all participants included in the present work

	Participants n	Participants with JIA accumulative n	Girls n (%)	Age, years median (range)	Onset type
Study I					
<i>Reliability;</i>	25	25	18 (72)	13 (4-17)	Oligo/poly/syst.
<i>Validity;</i>	107	114	86 (80)	7 (2-19.5)	Oligo/poly
Study II					
<i>Education;</i>					
Children	55		36 (65)	5.5 (2-12)	Oligo/poly
Adolescents	11	114	8 (73)	16 (13-19)	Oligo/poly
<i>Comparison;</i>					
Children	18		13 (73)	4.5 (2-7)	Oligo/poly
Adolescents	5	137	4 (80)	13.5 (13-16)	Oligo/poly
Study III					
<i>Content validity;</i>					
Children	7	141	5 (72)	7 (3-9)	Oligo/poly
Adolescents	7	146	5 (72)	12 (11-18)	Oligo/poly/JAS
<i>Construct Validity and Reliability;</i>					
Children	15	159	13 (87)	7 (2-9)	Oligo/poly/JAS
Adolescents	15	180	14 (93)	13 (10-19)	Oligo/poly/JAS
<i>Healthy Controls;</i>					
Children	14		10 (71)	8 (3-10)	n.a.
Adolescents	15		14 (93)	13 (10-19)	n.a.
Study IV					
Children and adolescents	48	207	34 (70)	13 (8-17)	Oligo/poly/JAS

Oligo=oligoarthritis; poly=polyarthritis, JAS=juvenile ankylosing spondylitis; n.a.=not applicable

3.2 ASSESSMENTS

An overview of the assessment methods used in the present work is given in Table III.

Table III. Outcome measures classified with the components of disability in ICF.

Assessments	Objective	Study	Impairment	Activity limitation Participation restriction
MEPS	Perception	I, II		X
Joint count	Swelling, joint pain, loss of motion	III, IV	X	
CHAQ	Disability	III, IV	X	X
Likert scale	Foot disability	III, IV		X
JAFI	Foot disability	III, IV	X	X
Foot position	Malalignment	IV	X	
VAS	Pain after capacity tests	IV	X	
Timing	Walking/running speed	IV		X
Bruininks-Oseretskys test	Balance capacity	IV		X

MEPS = Medical, Exercise, Pain and Social support; CHAQ = Child Health Assessment Questionnaire; JAFI = Juvenile Arthritis Foot disability Index; VAS= Visual Analog Scale

Demographic data related to age, sex, diagnostic subgroups, and disease duration were collected (Studies I-IV).

Lower-extremity joint impairment in hips, knees, ankles, rear-/midfeet and forefeet were assessed by scoring the presence of (1) capsular swelling or effusion (not for the hips or rear/midfeet), (2) tenderness or pain and (3) loss of motion. A total score

varying from 0 (no impairment) to 26 (maximal impairment) was derived by adding the scores from each joint (Studies III and IV).

The Swedish Childhood Health Assessment Questionnaire (CHAQ) was used to assess activity limitation (Andersson et al., 1993). It includes eight categories of activities: dressing, eating, walking, getting up, reaching, gripping, hygiene, and activity. Four of categories – getting up, walking, reaching, and activities – were used specifically to assess lower-extremity activity limitation (Bekkering et al., 2001). The total score varied from 0 (no limitation) to 3 (extensive limitation). The CHAQ is available in parents’ and children’s versions (Study III and IV).

Self-reported foot disability and self-reported physical activity were assessed on verbal rating scales with five answer options from “never” to “always” (Study III and IV).

Foot positions were classified as normal, cavovarus, or valgoplanus (Study III and IV).

Five capacity tests: standing 30 s with feet together, jumping 30 times (1/s) with both feet together, running 27.4 m including one turn, climbing 22 stair steps and walking 200 m at self-selected speed were performed in standardised order. Pain was rated on a visual analog scale (VAS, 0-100) after each test and time was recorded for walking and running (Study IV).

Eight single balance tasks were assessed and scored (example in Figure 3). The total score may vary between 0 and 36, the latter indicating excellent balance (Bruininks, 1978). The present results were not age-corrected as each child was compared to him/herself (Study IV).



Figure 3. Example of balance test in Bruininks-Oseretsky’s test.

3.2.1 Development of the Medical, Exercise, Pain, and Social support questionnaire (MEPS)

A questionnaire focusing on the perceived ability of parents to manage the disease and the consequences thereof among children with JCA, who are up to 12 years of age, and for adolescents with JCA from the age 13 years upward was developed (Study I). The questions were selected by a three-member panel consisting of a pediatric rheumatologist, a physical therapist experienced in pediatric rheumatology, and a parent of a child with JCA. Some of the questions were selected after reading scientific literature, others on the basis of clinical experience or the experience of everyday life with a child with JCA. The numbers of questions reflected a balance between comprehensiveness and feasibility. The questionnaire consisted of 24 (parents' version) and 23 (adolescents' version) questions within five areas: knowledge, skills, behavior, social support, and self-efficacy. The questions about self-efficacy were adapted from the Arthritis Self-efficacy Scale (Lorig et al., 1989). Each question was answered using visual analog scales (0-100), higher scores indicating greater perceived knowledge, greater self-efficacy etc. Anchor words for perception were 'none at all – enough', 'never - always', 'very little - very great', and 'not at all-often', depending on the questions. A convenience sample commented on the clarity of the questions and a 3-item questionnaire was answered to examine the content.

3.2.2 Development of the Juvenile Arthritis Foot disability Index (JAFI)

Two physical therapists, one experienced in pediatric rheumatology and the other in adult rheumatology, developed the new questionnaire (Study III). The content was based on the therapists' clinical experience combined with the content of two questionnaires on foot-related disability developed for adults with rheumatoid arthritis (Budiman-Mak et al., 1991; Sundbom and Stenström, 2003). This process generated 27 statements for a preliminary version, and these were divided into three dimensions according to the components of the ICF. Five-step Likert scales were attached to each statement (never – always). Some statements were positively formulated, others negatively, to make careful consideration of each statement necessary.

3.3 INTERVENTIONS

3.3.1 Patient education program

The education program in Study II consisted of themes presented during sessions totaling approximately eight hours (APPENDIX I). To suit the participating families the format varied between four times two study hours during one month and full-day seminars (eight hours). The latter was the most desirable. The themes covered were always the same and the professionals – pediatric rheumatologist, physical therapist, occupational therapist, rheumatology nurses, social worker, and dietician - and a representative from the patient association always lectured and led the discussions in their area of expertise, offering both theoretical and practical knowledge. Adolescents attended unaccompanied, primary and secondary school children with their parents but in separate groups with adapted programs. For the pre-school children, the education was aimed only at their parents. A comparison group received 'ordinary care' but no education program.

3.3.2 Custom-made foot orthoses

Soft custom-made foot orthoses (Figure 4) were evaluated (Study IV). The prescription was given by the pediatric rheumatologist, often initiated by the physical therapist, and with the aim of reducing pain and improving the foot position. The orthoses were fitted at the orthopedic workshop.

Two sets of the same capacity tests were performed in random order, one with the subject wearing her/his orthoses and one without orthoses. All tests were observed and scored by the same physical therapist, who was unaware of whether each participant had placed their orthoses in their shoes for the first set of tests or the second.



Figure 4. Example of a custom-made foot orthoses.

3.4 STATISTICS

An overview of the statistical methods used in the present work is given in Table IV. Non-parametric statistical analyses were used mainly in the present work, as the data were generally ordinal. Descriptive data are presented as median values with total ranges or inter-quartile ranges in brackets.

A series of factor analyses was performed in Study I to distinguish how questions grouped on the basis of underlying connections. The test-retest reliability in Study I was calculated with intra-class correlation coefficients (ICC 1.1) and the minimal acceptable ICC is suggested by Chinn (1991) to be 0.60, while Fleiss (1986) recommended that values between 0.40 – 0.75 represent fair to good reliability and values above 0.75 represent excellent reliability. The standard error of measurements (SEM) was also determined in Study I.

Friedman's ANOVA was used, where there were three measurement occasions, and the Wilcoxon signed ranks test, where there were two, to analyze changes within one group over time (Study II). Differences between the groups in Study II were analyzed with the Mann-Whitney test.

Sensitivity, specificity, and positive and negative predictive values were analyzed in Study III based on differences in JAFI responses from participants with JIA and healthy controls. Test-retest reliability was used in Study III with weighted kappa coefficients (K_w) to analyze systematic and random variations. The kappa values were considered as poor (<0.20), fair (0.21 - 0.40), moderate (0.41 - 0.60), good (0.61 - 0.80), or very good (0.80 – 1.00) (Altman, 1991). Spearman's correlation coefficients were used in Study III to determine internal redundancy ($r_s > 0.90$) and internal consistency ($r_s > 0.50$).

The results of Study IV were analyzed to determine differences in capacity tests with the Sign test in the present work and with the Wilcoxon signed ranks test in the submitted manuscript.

Table IV. Statistical methods used in the present work

Analyses	Study I	Study II	Study III	Study IV
Descriptive statistics				
Median, range	X	X	X	X
Median, interquartile range	X	X		
Mean, standard deviation (SD)	X			
Standard error of measurement (SEM)	X			
Analytical statistics				
Factor analysis	X			
Intraclass correlation (ICC)	X			
Friedman's analysis of variance (ANOVA)		X		
Wilcoxon's signed ranks test		X		X
Mann-Whitney's test		X		
Sensitivity			X	
Specificity			X	
Floor/Ceiling effects			X	
Spearman's correlation coefficient			X	
Weighted kappa coefficient			X	
Sign test				X

3.5 ETHICS

All participants (children and parents) were fully informed verbally and in writing, and consent was obtained from both parents and children. The local ethics committee at the Karolinska Hospital approved all the studies included in the present work (Dnr 96-271, Dnr 00-012).

4 RESULTS

4.1 STUDY I - MEPS

4.1.1 Content validity

After input from samples from the target population, the questionnaire was judged to be easily understood, relevant and exhaustive. All participants but one considered all the questions easy to understand. Additional questions were suggested by single participants, but were not included.

A series of explorative factor analyses was performed, of which the one with four factors was chosen as the most theoretically and clinically relevant. The four underlying dimensions were labeled; medical issues, exercise, pain and social support. The suggested questionnaire comprised these dimensions and was named the MEPS questionnaire (APPENDIX II).

4.1.2 Reliability

Test-retest reliability varied between ICC=0.68 and 0.96 (median 0.87) among the single questions. Twenty of the twenty-four questions had ICC above 0.75. The question about self-efficacy for regulation of activities indicated the lowest ICC (0.68). The standard error of measurement varied between eight and 17 (median 11.5).

4.2 STUDY II – OUTCOME OF THE EDUCATION PROGRAM

4.2.1 Parents

Answers to the MEPS questionnaire from 55 parents of children below age 13 years indicated consistent and significant improvements in the medical area and in the ability to manage pain directly after the education program. Significant improvement was also found in two of four exercise and social support questions. The significant changes remained four months after the end of the education program. Questionnaire answers from 18 parents in the comparison group indicated a significant improvement in one of 28 questions over four months.

Differences between the education group and the comparison group were significantly in favor of the education group in all four areas of the MEPS, most pronouncedly in the medical area and least in the social support area.

4.2.2 Adolescents

Questionnaire answers from 18 adolescents indicated improvements after the education program in two separate questions, one in the medical area and one on pain. Significant differences between the education group and the comparison group, in favor of the former, were also found in two questions in the medical area.

4.3 STUDY III - JAFI

4.3.1 Content validity

After the addition of a suggested statement on participation restriction, the JAFI was found to be easily understood, relevant, and exhaustive. Thus, the measurement properties of a 28-statement JAFI were further investigated.

4.3.2 Construct validity

A generally consistent pattern of increasing JAFI scores in its three dimensions corresponded largely with similar patterns of increasing scores in other assessments of impairment, activity limitation, and participation restriction respectively. The children with JIA scored higher on all three JAFI dimensions than healthy controls did.

4.3.3 Sensitivity and specificity

The sensitivity for the three JAFI dimensions was; impairment=0.83, activity limitation=0.86, and participation restriction=0.77 and the specificity was 0.93, 0.93, and 1.0 respectively.

4.3.4 Ceiling and floor effects

There were no ceiling or floor effects in 27 of the 28 statements. One statement on participation restriction had a floor effect and proved to be irrelevant to 11 of 30 participants. It was thus excluded from the suggested 27-item JAFI (APPENDIX III).

4.3.5 Reliability

No systematic differences were found in single statements or in total scores between two sets of JAFI answers separated by one week. Weighted kappa coefficients for the total scores on the three dimensions were: impairment=0.90, activity limitation=0.85 and participation restriction=0.88.

4.4 STUDY IV – OUTCOME OF THE USE OF FOOT ORTHOSES

4.4.1 General outcome

Pain after standing, running, stair-climbing and walking was significantly lower with foot orthoses than without. Significant differences in walking speed and balance capacities were also found in favor of foot orthoses. Pain after jumping or running speed were not significantly related to the wearing of orthoses (Table V). The subgroup, including only those (n 33-45) with pain when not using orthoses had significantly less pain, higher walking speed, and better balance capacities when using orthoses. No significant difference was found in running speed with versus without orthoses in this subgroup (Table V).

Table V. Results from assessment without and with custom-made orthoses for 48 children with JIA. Results are also shown separately with those not indicating pain in single tests without foot orthoses (n=3-15) excluded.

	All participants n = 48			Participants with pain n = 33-45		
	Without md (range)	With md (range)	Sig.level	Without md (range)	With md (range)	Sig.level
Standing 0-100	9 (0-86)	3 (0-72)	<0.01	15 ¹ (1-86)	11 (0-72)	<0.001
Jumping, 0-100	18 (0-75)	15 (0-80)	n.s.	21 ² (1-75)	20 (0-80)	<0.05
Running, 0-100	27 (0-83)	15 (0-86)	<0.001	27 ⁴ (1-93)	21 (0-86)	<0.001
Running, s	9.2 (6-22)	9.0 (6-26)	n.s.	9.2 ⁴ (6-22)	9.1 (6-26)	n.s.
Stairs, 0-100	16 (0-92)	14 (0-75)	<0.01	25 ³ (2-92)	20 (0-75)	<0.01
Walking, 0-100	18 (0-86)	11 (0-75)	<0.05	28 ² (1-86)	15 (0-75)	<0.05
Walking, s	107 (80-167)	106 (84-177)	<0.05	108 ² (86-167)	106 (85-177)	<0.05
Balance, 0-36	22 (11-31)	24 (7-32)	<0.001	22 ¹ (11-31)	26 (11-32)	<0.001

¹ n=33, ² n=39, ³ n=38, ⁴ n=45
s=seconds, n.s.=non significant

4.4.2 Outcome related to foot position

Pain after standing, jumping and stair-climbing was significantly lower ($p < 0.05$) with orthoses in the normal foot-position subgroup, and balance improved ($p < 0.05$) in favor of the orthoses. No significant difference was found in the subgroup with valgoplanus. However, in the cavovarus subgroup significant differences were found in pain after standing ($p < 0.01$), walking ($p < 0.05$) and running ($p < 0.01$), in walking speed ($p < 0.05$), and in balance capacities ($p < 0.01$), again in favor of orthoses.

4.4.3 Outcome related to onset type

The three diagnostic subgroups had differing results. The children with oligoarthritis indicated significantly less pain after running ($p < 0.05$), stair-climbing ($p < 0.05$), and walking ($p < 0.01$), and higher walking speed ($p < 0.05$), when using orthoses than when not using them. Those with polyarthritis also indicated significantly less pain after standing and running ($p < 0.01$), in walking speed ($p < 0.05$), and in balance capacities ($p < 0.01$) when wearing their orthoses. However, no significant effects of foot orthoses were found among the children with JAS.

5 DISCUSSION

5.1 MAIN FINDINGS

The main focus of the present work was to study the outcomes of an education program for parents of children with JIA and for adolescents with JIA, and of the use of custom-made foot orthoses. The work also introduced two outcome measures specifically developed to assess perceived ability to manage JIA and foot-related disability. The results indicate significant benefits from the education program among the parents and from foot orthoses in improved balance capacities, walking time and reduced pain. The new questionnaires possessed satisfactory measurement properties and seemed well suited for assessing the actual interventions.

5.1.1 MEPS and JAFI

Self-reported measurements yield information that may not be attainable from clinician-based scales (Duffy and Duffy, 1997). To encompass all aspects of the construct under study it is useful to derive information from the literature as well as from experts when developing new measures. Theoretical models and empirical data exist in the literature and experts may be recruited among clinicians as well as among patients and their relatives.

The MEPS (Study I) was originally based on clinical experience and scientific literature (Lorig et al., 1989; Lorig and Holman, 1993), to cover five areas; knowledge, skills, behavior, social support and self-efficacy. However, four areas were identified during content validation; medical, exercise, pain and social support. All these have been mentioned as important issues in JIA (Rapoff et al., 1988, 1989; Harris et al., 1991; Sällfors et al., 2004). The four-factor model was chosen partly based on clinical relevance and partly on the results of the explorative factor analyses. The grouping of items was considered more important than the loadings of separate items. The MEPS was subsequently found to possess satisfactory reliability and sensitivity to change.

The ICF was explicitly used as a theoretical framework in the development of the JAFI. Clinical experience in pediatric rheumatology was used and items from existing questionnaires in adult rheumatology were selected and grouped in accordance with the ICF components. Input from adolescents and parents of children with JIA was also requested. For construct validity, the present hypotheses about relations between the three JAFI dimensions and other measures related to corresponding ICF components were generally confirmed. However, the JAFI dimension participation restriction also related well to the CHAQ, which may be ascribed to the latter including some items in this ICF component. Further, the JAFI dimension impairment, although well related to the joint impairment score, also seemed to relate well to self-rated participation restriction. The main reason for this is probably that a one-dimensional question was used for validation. This was probably not ideal, but without valid multidimensional participation restriction measurements, seemed a reasonable choice. Sensitivity and specificity values indicate that the JAFI is sensitive enough to record foot-related disability among children with JIA, but still specific enough to exclude individuals

without this condition. The JAFI was used in the present work (Study IV) for descriptive purposes only and its sensitivity to change following different kinds of physical therapy and other treatments such as long-term effects of foot orthoses, intra-articular corticosteroid injections or surgery is still unknown.

Two questionnaires were developed in the present work in processes that partly differed. Thus, different numbers of participants and different statistical procedures were used. The contents of the MEPS were based on clinical experience and literature, and strongly connected to the expected outcome of the education program. The ICF was used as a theoretical framework for the development of the JAFI, which is intended for more general application. The content validity of the MEPS was carefully tested, while other measurement properties of the JAFI were also tested. Yet the present results indicate that both questionnaires seem to be suitable tools that cover areas where outcome measures have been lacking in the population of JIA.

5.1.2 Outcome of education program

Results from studies investigating parental distress in families with a child with JIA reveal conflicting results. Some suggest increased risks (Lustig et al., 1996; Manuel 2001), while other find no differences in health-related quality of life or psychological status (Press et al., 2002). Patient education and family support have been suggested to reduce emotional distress among caregivers, and to increase knowledge and parenting skills, thus reducing disability among children with JIA (Harris et al., 1991; Daltroy et al., 1992; Lineker et al., 1996). A family retreat has previously been found to improve emotional functioning of children with JIA and reduce the strain on caregivers' leisure activities, but not to give reduction in reported pain (Hagglund et al., 1996). To the present author's knowledge the present work is the first in the field of JIA to investigate the outcome of a patient education program aiming at improving self-reported competence to manage JIA. As to the participating parents, who improved, the result here tallies well with effects of education programs for adults with arthritis and for parents of children with other chronic diseases (Finney and Bower 1992, Lorig et al., 1987; Matthews et al., 1998). Previous research investigating patient education of adolescents with chronic disease is scarce. Thus, the present findings of only minor improvement are hard to compare with other studies and should be interpreted with caution, in particular because of the small sample (Study II).

The positive outcome in the parent group was most pronounced in the medical area, where significant improvement was indicated in all questions. It is known that the acquisition of knowledge and the learning of specific skills are the factors that are easiest to influence as an outcome of an education program (Bloom, 1956; Lorig et al., 1989). However, it is worth noting that, although the disease had lasted up to 10 years, the participants obviously had not acquired this knowledge within 'ordinary care'.

The children in the present work seemed to be participating very actively in physical education at school/pre-school already at baseline, which left very little room for improvement following the education program. It might however be that 'participation' is not always active or includes participation in all parts of the physical education program. A better question to capture this might have been 'Do you attend most

activities (all parts) during physical education at school?' The participation in leisure-time physical activity was reportedly not as frequent before the education program. The tendency toward an increase did not reach statistical significance, which was possibly due to a wide range of results. On the other hand, significant improvement in self-efficacy – the ability to continue and regulate daily activities - might predict future increases in physical activity.

Pain and its consequences are important issues in JIA and were highlighted from different professional perspectives in the education program. After the program, the parents, in five of six questions, increased their self-efficacy to manage pain. This confirms previous suggestions that the teaching of pain-coping strategies may increase the perception of pain control (Schanberg et al., 1997). Another recent study reported a significant relationship between pain-coping strategies and health-related quality of life (Sawyer et al., 2004). This might indicate that the program improved health-related quality of life among the parents although this aspect was not studied in the present work.

Social support is important for the parents of children with chronic diseases and the value of both informal and more formalized social contacts was rated very highly in this work. Considering the highly rated value of social contacts, it may seem surprising that so few during the follow-up period took advantage of opportunities for such contacts. However, the large catchment area for the participants in the education program and the high demands on two-job families with the added burden of JIA might explain this. Children with JIA have also expressed their wishes to equal their peers (Sällfors et al., 2001), and maybe give priority to friends in their own social context.

The MEPS were completed in all three measurements by 55 parents and the significant changes directly connected with the education program were mainly maintained at the four-month follow-up. This indicated a change presumably not attributable to possible placebo effects. Six adolescents dropped out from the study, because they did not complete the MEPS at the four-month follow-up. The limited number of adolescents and the high frequency of dropout among them (35%) preclude specific conclusions about effects of the education program for this subgroup. Poor adherence is common in studies with adolescents. One's impression is, however, that dropouts were caused rather by their reluctance to complete questionnaires than by experienced poor outcome of the education program. However, another format of education program might have suited them better.

It has been found that parents of children with oligoarticular onset, which might be considered as less severe than other onset types, report lower health-related quality of life and more frequent depression than do parents of children with other onset types. Suggested explanations for this were that these patients had shorter disease duration or were less frequently seen in the clinic (Press et al., 2002). Effects related to onset type were beyond the aim of the present work. However, the education program appeared particularly appreciated by parents of children with less severe disease.

5.1.3 Outcome of the use of foot orthoses

The aim of physical therapy for children with JIA is to maintain or increase physical activity (Klepper 2003), with walking, stair-climbing, running, and balance as important elements. Foot impairment has been identified as the most important predictor of activity limitation in e.g. walking, running and playing, in children with JIA (Bekkering et al., 2001). Thus, the use of foot orthoses may enhance physical activity. As far as is known the present work is the first to study the effects of the common intervention with foot orthoses in children with JIA. The results (Study IV) indicated positive effects as to pain relief and improved capacities in activities. While several participants in each of the single tests indicated no pain, only one individual was pain-free in all tests. Further, despite a large proportion of subjects who were pain-free when standing, the set of tests used did capture the participants' problems. The positive effects were more pronounced when pain-free individuals were excluded from the analyses.

No effect of foot orthoses on running speed was found. This might be due to the instruction: 'run as fast as you can'. While these children run fast even without orthoses, they may have to 'pay' with pain. This hypothesis is confirmed by the higher pain found after running without orthoses. The opposite seems to be true for the walking test, where the participants were instructed to 'walk at your own speed'. Consequently, a significantly higher speed was found with orthoses and these results tally with the clinical experience that children with JIA can force themselves to do activities, but 'pay' in pain unless they can move more slowly. Little is known about balance capacities among children with JIA, but clinical experience and textbooks indicate that they might be reduced. Balance difficulties and consequent feelings of insecurity may decrease physical activity, which might be particularly relevant for younger children with JIA who are still developing their balance skills. As balance capacity was also found in the present work to be better when orthoses were used, these may be extra important for young children.

The present results are similar to those in studies of foot orthoses in adult rheumatology (Woodburn et al., 2002), particularly as regards pain reduction. Another recent study (Woodburn et al., 2003) found significantly reduced malalignment in gait when foot orthoses were used. Most interestingly a significant reduction of malalignment while walking barefoot was observed after treatment with foot orthoses for 12 months. The authors hypothesized that the orthoses might have improved the orientation and alignment of soft-tissue structures in and around the ankle and thus improved proprioception and neuromuscular control while maintaining joint flexibility. Children's foot alignment is much more flexible than that of adults with RA and children may thus be expected to derive even more pronounced long-term benefit from foot orthoses.

It is important in the clinic, to develop guidelines for prescribing foot orthoses for children with JIA. A secondary aim was thus to identify characteristics of individuals, among the heterogeneous JIA group, who might benefit from foot orthoses. Therefore the results of orthosis use were also studied in relation to different types of malalignment and different diagnostic subgroups. However, the number of participants

in some of the groups was sometimes very small and confounding factors might have influenced the results. Thus, these results should be interpreted with caution.

5.2 METHODOLOGICAL CONSIDERATIONS

It may be argued that some samples used in the process of developing the MEPS and the JAFI were too small (Studies I and III). However, they were strategically chosen to represent a wide variety of background factors, which might be more important in this respect than the actual number of participants.

Unfortunately, floor and ceiling effects of the MEPS were not analyzed. Thus, some questions, in particular that on exercise participation at school, later proved to have an obvious ceiling effect, which might have contributed to a false negative outcome of the education program (Study I). The answers to single questions of the MEPS were generally satisfactorily stable over one week. However, one question on self-efficacy to manage pain seemed somewhat instable. This might have reflected a true change rather than indicating low reliability of the question, particularly in the case for parents, who needed to consider their own self-efficacy as well as to interpret their children's situations when answering the question (Study I).

Visual analog scales were used in the MEPS and Likert scales in the JAFI. Assessing children is always a challenge and both scales have their pros and cons. A recent study (Lam et al., 2004) of a revised version of the CHAQ investigated the applicability of different response scales. It was found that the VAS appears to offer the best differentiation between subjects and controls and is almost normally distributed. However, a five-categorized response alternative was easiest to complete and showed the best agreement between children and parents. Thus, there seems to be no ideal response scale to use in the assessment of children and their parents.

The measurement properties of new assessment methods need to be investigated in a population similar to the for which they are to be used. As regards children, their age and developmental stage are important to consider. The MEPS was tested for adolescents (age 13 years and above), while the JAFI was tested for children from 10 years and up. The reason for this is that the education program, for which the MEPS was developed was evaluated by the adolescents independently of their parents, while the younger children were represented in the evaluation by their parents. The JAFI, however, was developed for all children with satisfactory reading and writing skills. For the present work this was at age 10, which is more or less in accordance with other self-reported questionnaires (Andersson et al., 1993). It may be tempting to include younger children, who might very well possess the required skills, but this could threaten the validity of the answers.

Both questionnaires developed in the present work produce ordinal data. The type of statistical procedure suggested for their analysis is constantly under debate. Thus, parametric analyses were used for the analysis of the MEPS, while non-parametric procedures were applied to the JAFI. The result of the factor analysis of the MEPS was, however, interpreted with respect to the grouping of the items rather than to the numbers produced.

One limitation of the evaluation of the education program (Study II) was the lack of a randomized control group. This was partly solved by a subsequent inclusion of a matched comparison group. Although there is no reason to suspect any systematic differences between the groups, which were both recruited from a rather homogenous population, unobserved differences might possibly have been present and influenced the outcome. Lack of statistical power might have been one reason for the meager results as to outcome among the adolescents participating in the education program (Study II). The high dropout rate among them was not expected, but should be noted for future studies. Children between the age of seven and twelve participated in the adapted education program, but not in the evaluation of its outcome. It would have been of interest to also assess their perceptions and compare them with their parents'.

The design of the study on foot orthoses (Study IV) with two consistent sets of tests on the same day may be questioned. Some participants may have been less stiff in the second set of tests and others may have had more pain after already performing one set. However, given the random order of the tests, this should not have biased the results. In fact there was no difference in outcome that could be attributed to whether orthoses were used during the first set or the second set of tests (data not shown).

Gender differences have not been investigated in the present work, neither between girls and boys nor between fathers and mothers. The main reason for this was that the samples included were generally too small, particularly in consideration of possible interaction effects between parents and children of the same or different gender. While both parents often participated in the education program, only one of them, determined by them, completed the MEPS at all occasions. This might have been a drawback as a recent study has found discrepancies in self-reports related to gender (Sällfors et al., 2003).

5.3 CLINICAL IMPLICATIONS

It is recommended that the two questionnaires developed in the present work be included in routine clinical work to assess competence to manage JIA and foot-related disability. They both fill a void in the arsenal of assessment methods presently available to physical therapy for children with JIA.

The positive outcome of the education program for the parents suggests that this should become a self-evident part of the treatment of JIA. The program might decrease parents' distress and reduce the children's disability in the long run. The physical therapist's contribution, focusing on physical activity, pain-management skills, fatigue and stiffness, in the education program, might be particularly important in view of the fact that a recent study identified increased daily symptoms of pain, stiffness and fatigue as significant predictors of reduced participation in school and in social activities (Schanberg et al., 2003). Other contents and formats of education programs are needed to better suit this group of adolescents.

The present results regarding the effects of the foot orthoses suggest that these should be prescribed to children with JIA. The improved balance, the pain reduction and the

higher walking speed are important for appropriate levels of physical activity in daily life.

The promotion of physical activity is an important element of physical therapy. Previous studies, mainly of planned structured exercise, have not evaluated outcome in terms of changed physical activity behavior, but rather of various elements of impairment (Takken et al., 2003). Thus, possible effects on everyday physical activity have not been evaluated. Another problem in these studies might have been poor adherence. Significant benefits of the present interventions have been found. Whether they actually influence physical activity behavior still remains unclear.

5.4 FURTHER STUDIES

The two questionnaires MEPS and JAFI were developed and validated in a Swedish context and thus translation and cross-cultural adaptation (Guillemin, 1995) will be required before they can be used in other countries. Further, the MEPS might need slight revision to increase sensitivity; and to be tested for younger children. The JAFI should be further tested as to sensitivity to change for various interventions and also for children aged 8-9 years.

The education program needs further development for, and testing among, adolescents. It might also be of interest to study the effects of education on affective factors, pain, activity limitation and participation restriction.

The immediate effects of foot orthoses have been studied in the present work, but this type of long-term treatment needs to be evaluated in long-term studies with the JAFI as one outcome method. This would be similar to the use of the Foot Function Index in adult rheumatology (Budiman-Mak et al.1991; Woodburn et al., 2002).

Studies of the effects of education and foot orthoses on daily physical activity levels are also suggested. Such studies could be performed with parents' ratings of their children's physical activity levels or with tests of aerobic fitness. Both seem to be valid in terms of correlation with Caltrac motion counts (index for the daily physical activity) (Takken et al., 2005).

6 CONCLUSIONS

- The MEPS questionnaire is reliable and possesses satisfactory content validity in the assessment of adolescents' and parents' perceived ability to manage JIA (Study I). It is also sensitive to change after an education program (Study II).
- An eight-hour education program confers consistent and significant benefits in self-reported competence among parents of children with JIA. Adolescents are less likely to benefit (Study II).
- The JAFI appears to be valid and reliable for assessing foot-related disability among children/adolescents with JIA (Study III).
- Custom-made foot orthoses confer major immediate benefits related to pain relief, improved balance capacities and walking speed among children with JIA. The most pronounced improvements might be expected among those with cavovarus and those with oligoarthritis or polyarthritis (Study IV).

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APPENDIX I

Contents in the eight-hour Education Program

Pediatric Rheumatologist

General disease information, prevalence and incidence, different onset types, prognosis, treatment overview, pharmacology, possible side-effects of medical treatment, update on research.

Pediatric Rheumatology Nurse

Preparation for treatment: information on how to prepare children for treatment such as blood sampling and intra-articular injections.

Physical therapist

Joint anatomy, how to handle stiffness, the benefit and safety of physical activity, how to adjust physical activity and facilitate movement, increase self-efficacy to continue with physical activity.

Occupational therapist

Influence of the disease on wrists and fingers and how to handle it. Information on technical aids and orthoses, the development of activities and skills to maintain independence.

Social Worker

Information and advice about having a child with a chronic disease and the impact on the family. Social security systems and rights.

Dietician

Information on the importance of good nutrition for a growing child and overview of different diets.

Patient association

Information patient association and their activities.

APPENDIX II

Medical Exercise Pain Social support, MEPS

Föräldrar till barn < 13 år.

Markera på skalorna nedan med ett kryss där Ni tycker att det stämmer med er kunskap eller uppfattning idag.

Medicinsk, Ankarord på VAS; Fråga 1-9 Ingen alls – Tillräcklig.

- 1) Hur stor kunskap har du om den medicinska bakgrunden till barnreumatism ? (mekanismerna bakom sjukdomsaktiviteten)
- 2) Hur stor kunskap har du om ledens uppbyggnad ? (vad består leden av)
- 3) Hur stor kunskap har du om vad labsvaren ger för information till läkaren?
- 4) Hur stor kunskap har du om vad som händer vid inflammation inne i leden?
- 5) Hur stor kunskap har du om olika typer av medicinering vid barnreumatism?
- 6) Vet du hur du kan förbereda ditt barn inför provtagning?
- 7) Vet du hur du kan förbereda ditt barn inför cortisoninjektioner?
- 8) Hur stor kunskap har du om vilka de synliga symptomen på inflammation är?
- 9) Hur stor kunskap har du om olika typer av smärta? (belastningssmärta, stelhetssmärta)

Rörelse, Ankarord på VAS; Fråga 1-2 Aldrig - Alltid, Fråga 3-4; Mycket osäker – Mycket säker

- 1) Deltar ditt barn i gymnastik på dagis/lekskola eller skolan?
- 2) Deltar ditt barn i fysisk aktivitet (ex fotboll, dans) utanför dagis eller skola?
- 3) Hur säker känner du dig på att kunna låta ditt barn fortsätta med sina dagliga aktiviteter?
- 4) Hur säker känner du dig på att kunna anpassa barnets aktiviteter så att han/hon kan vara aktiv utan att förvärra sina symptom?

Smärta, Ankarord på VAS; Fråga 1-4, 7; Mycket osäker – Mycket säker, Fråga 5; Aldrig - Alltid, Fråga 6; Inte alls – Tillräckligt

- 1) Hur säker är du på att kunna åstadkomma en liten till måttlig minskning av barnets smärta genom andra metoder än ökad medicinering?
- 2) Hur säker är du på att kunna minska barnets smärta avsevärt?
- 3) Hur säker är du på att kunna hantera barnets symptom så att barnet kan göra saker som han/hon tycker om att göra?
- 4) Hur säker är du på att kunna åstadkomma en avsevärd minskning av barnets smärta genom andra metoder än ökad medicinering?
- 5) Kan du idag se att ditt barn har ont, även om de själva inte påtalar detta?
- 6) Vet du hur du kan hjälpa ditt barn att lindra smärta?
- 7) Hur säker känner du dig på att kunna undvika att barnets smärta stör sömnen?

Social, Ankarord på VAS; Fråga 1-2; Obetydligt – Mycket stort, Fråga 3; Aldrig – Alltid, Fråga 4; Ingen alls - tillräcklig

- 1) Vad anser du om värdet att träffa andra familjer med barn som har reumatism?
- 2) Vad anser du om värdet av en föräldrarförening?
- 3) Träffar och utbyter du erfarenheter med andra familjer med barn som har reumatism?
- 4) Hur stor kunskap har du om samhällets stödåtgärder? (ex vårdbidrag, utlandsvård)

Medical Exercise Pain Social support, MEPS

Ungdomar \geq 13 år.

Markera på skalorna nedan med ett kryss där du tycker att det stämmer med din kunskap eller uppfattning idag.

Medicinsk

Ankarord på VAS; Fråga 1-9 Ingen alls – Tillräcklig.

- 1) Hur stor kunskap har du om den medicinska bakgrunden till barnreumatism? (mekanismerna bakom sjukdomsaktiviteten)
- 2) Hur stor kunskap har du om ledens uppbyggnad? (vad består leden av)
- 3) Hur stor kunskap har du om vad labsvaren ger för information till läkaren?
- 4) Hur stor kunskap har du om vad som händer vid inflammation inne i leden?
- 5) Hur stor kunskap har du om olika typer av medicinering vid barnreumatism?
- 6) Känner du rädsla inför provtagning?
- 7) Känner du rädsla inför cortisoninjektioner?
- 8) Hur stor kunskap har du om vilka de synliga symptomen på inflammation är?
- 9) Hur stor kunskap har du om olika typer av smärta? (belastningssmärta, stelhetssmärta)

Rörelse

Ankarord på VAS; Fråga 1-2 Aldrig - Alltid, Fråga 3-4; Mycket osäker – Mycket säker

- 1) Deltar du i idrottslektionerna i skolan?
- 2) Deltar du i fysiska aktiviteter (ex fotboll, dans) utanför skolan?
- 3) Hur säker känner du dig på att kunna fortsätta med dina dagliga aktiviteter?
- 4) Hur säker känner du dig på att kunna anpassa dina aktiviteter så att du kan vara aktiv utan att förvärra dina symptom?

Smärta

Ankarord på VAS; Fråga 1-4; Mycket osäker – Mycket säker, Fråga 5; Inte alls – Tillräckligt, Fråga 6; Aldrig - Alltid

- 1) Hur säker känner du dig på att kunna åstadkomma en liten till måttlig minskning av din smärta genom andra metoder än ökad medicinering?
- 2) Hur säker är du på att kunna minska din smärta avsevärt?
- 3) Hur säker är du på att kunna hantera dina symptom så att du kan göra saker som du tycker om att göra?
- 4) Hur säker är du på att kunna åstadkomma en avsevärd minskning av din smärta genom andra metoder än ökad medicinering?
- 5) Kan du idag lindra din smärta?
- 6) Händer det att din smärta stör sömnen?

Social

Ankarord på VAS; Fråga 1-2; Obetydligt – Mycket stort, Fråga 3; Aldrig – Alltid, Fråga 4; Ingen alls - tillräcklig

- 1) Vad anser du om värdet att träffa andra ungdomar som har reumatism?
- 2) Vad anser du om värdet av en patientförening? (ex Unga reumatiker)
- 3) Träffar och utbyter du erfarenheter med andra ungdomar som har reumatism?
- 4) Hur stor kunskap har du om samhällets stödåtgärder? (ex vårdbidrag, utlandsvård)

JUVENILE ARTHRITIS FOOT DISABILITY INDEX – JAFI

Ungdomar ≥ 10 år.

Följande påståenden gäller hur besvären varit den senaste veckan i den sämsta foten.

De närmast följande 9 påståendena har fem svarsalternativ, som anger hur ofta de omnämnda besvären förekommer: Aldrig, Enstaka tillfällen (mindre än en gång i veckan), Ibland (en gång i veckan), Ofta (2-3 ggr/vecka) och Alltid.

1. Jag är morgonstel i foten/fötterna
() Aldrig () Enstaka tillfällen () Ibland () Ofta () Alltid
2. När jag går och står mycket svullnar min fot / mina fötter
3. Jag har värk/ont i foten/fötterna innan jag stiger upp och belastar mina fötter
4. Jag har ont i framfoten/-fötterna vid gång
5. Jag har ont i bakre delen av foten vid gång
6. Jag har ont/värk i foten/fötterna efter att ha varit igång hela dagen
7. Jag får ont i foten/fötterna när jag sätter på mig skor/stövlar
8. Jag kan belasta hela min fot när jag går
9. Jag blir fort trött i fötterna vid rörelse och lek på grund av mina fotproblem

Nästa 14 påståenden har fem svarsalternativ. Fortfarande handlar det om hur besvären varit den senaste veckan.

10. Jag är ostadig/har dålig balans när jag går barfota
11. Jag är ostadig/har dålig balans när jag går med skor
12. Jag avbryter rörelse och lek på grund av mina fotproblem
13. Jag undviker rörelse och lek på grund av mina fotproblem
14. Jag kan utan svårighet gå fort trots mina fotproblem
15. Jag kan utan svårighet gå barfota trots mina fotproblem
16. Jag kan utan svårighet gå så långt jag vill trots mina fotproblem
17. Jag kan utan svårighet gå i ojämn terräng, det vill säga i skogen eller backar, trots mina fotproblem
18. Jag kan utan svårighet gå i trappa trots mina fotproblem
19. Jag kan utan svårighet springa en kort sträcka (ca 20 m) trots mina fotproblem
20. Jag kan utan svårighet springa en längre sträcka (ca 100 m) trots mina fotproblem
21. Jag kan utan svårighet hitta bekväma skor
22. Jag känner mig ledsen över mina fotproblem
23. Jag känner mig orolig för att inte kunna delta i rörelse och aktivitet som jag vill på grund av mina fotproblem

Nästa 4 påståenden har fem svarsalternativ. Fortfarande handlar det om hur besvären varit den senaste veckan.

24. Mina fotproblem hindrar mig från deltagande i fysiska aktiviteter, till exempel skolidrotten, dans, sport, fritidsaktiviteter
25. Mina fotproblem hindrar mig från deltagande i sociala aktiviteter till exempel utflykter med skolan, umgänge med kamrater efter skoltid, biobesök
26. Jag behöver skjuts till skolan (bil/buss) istället för att gå på grund av mina fotproblem
27. Det är svårt för omgivningen att förstå hur mina fotproblem påverkar mig

JUVENILE ARTHRITIS FOOT DISABILITY INDEX – JAFI

Föräldrar till barn < 10 år

Följande påståenden gäller hur besvären varit den senaste veckan i den sämsta foten.

De närmast följande 9 påståendena har fem svarsalternativ, som anger hur ofta de omnämnda besvären förekommer: Aldrig, Enstaka tillfällen (mindre än en gång i veckan), Ibland (en gång i veckan), Ofta (2-3 ggr/vecka) och Alltid.

1. Mitt barn verkar morgonstel i foten/fötterna
() Aldrig () Enstaka tillfällen () Ibland () Ofta () Alltid
2. Mitt barns fot/fötter svullnar när han/hon står och går mycket
3. Mitt barn verkar ha värk/ont i foten/fötterna på morgonen innan han/hon belastar sina fötter
4. Mitt barn verkar ha ont i framfoten/-fötterna vid gång
5. Mitt barn verkar ha ont i bakre delen av foten vid gång
6. Mitt barn verkar ha ont/värk i foten/fötterna efter att ha varit igång hela dagen
7. Mitt barn verkar ha ont i foten/fötterna när jag/han/hon sätter på skor/stövlar
8. Mitt barn kan belasta hela sin fot när han/hon går
9. Mitt barn blir fort trött i fötterna vid rörelse och lek på grund av sina fotproblem

Nästa 14 påståenden har fem svarsalternativ. Fortfarande handlar det om hur besvären varit den senaste veckan.

10. Mitt barn verkar ostadig/ha dålig balans när han/hon går barfota
11. Mitt barn verkar ostadig/ha dålig balans när han/hon går med skor
12. Mitt barn avbryter rörelse och lek på grund av sina fotproblem
13. Mitt barn undviker rörelse och lek på grund av sina fotproblem
14. Mitt barn kan utan svårighet gå fort trots sina fotproblem
15. Mitt barn kan utan svårighet gå barfota trots sina fotproblem
16. Mitt barn kan utan svårighet gå så långt hon/han vill trots sina fotproblem
17. Mitt barn kan utan svårighet gå i ojämn terräng, det vill säga i skogen eller backar, trots sina fotproblem
18. Mitt barn kan utan svårighet gå i trappa trots sina fotproblem
19. Mitt barn kan utan svårighet springa en kort sträcka (ca 20 m) trots sina fotproblem
20. Mitt barn kan utan svårighet springa en längre sträcka (ca 100 m) trots sina fotproblem
21. Jag kan utan svårighet hitta bekväma skor till mitt barn
22. Mitt barn är ledsen över sina fotproblem
23. Mitt barn känner oro för att inte kunna delta i rörelse och aktivitet som han/hon vill på grund av sina fotproblem

Nästa 4 påståenden har fem svarsalternativ. Fortfarande handlar det om hur besvären varit den senaste veckan.

24. Mitt barns fotproblem hindrar honom/henne från deltagande i fysiska aktiviteter, till exempel skolidrotten, dans, sport, fritidsaktiviteter
25. Mitt barns fotproblem hindrar honom/henne från deltagande i sociala aktiviteter till exempel utflykter med skolan, umgänge med kamrater efter skoltid, biobesök
26. Mitt barn behöver skjuts till skolan (bil/buss) istället för att gå på grund av sina fotproblem
27. Det är svårt för omgivningen att förstå hur mitt barns fotproblem påverkar honom/henne