

*From the Department of Woman and Child Health  
Karolinska Institute, Stockholm, Sweden*

**Juvenile idiopathic arthritis**  
**Disease consequences and treatment effects**  
**on muscle strength, gait and pain**

**Eva Broström**



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Cover: Catharina Günther-Rådström

Correspondence: Eva Broström  
MotorikLab Q2:07 ALB  
Karolinska Hospital  
SE-171 76 Stockholm, Sweden

Tel: + 46 8 517 77 636  
Fax: + 46 8 517 77 349

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To all of the children with JIA, parents and all of my family members



## **ABSTRACT**

Juvenile idiopathic arthritis (JIA) may have a profound effect on the life of a child. Rheumatic diseases in children manifest mainly as synovitis, pain, stiffness, deformity, growth disturbances and fatigue. It is not uncommon to have reduced activity, lower aerobic endurance, and decreased muscle strength. The inflammatory process of the disease most often affects extremities and restriction of joint motion often occurs. Forty percent of the children with JIA report difficulty in daily activities. The aims of this thesis were to compare lower extremity strength and gait and to evaluate pain in children with JIA and children without musculoskeletal dysfunction.

Both a Hand Held Dynamometer (HHD) and an isokinetic dynamometer were used to test maximal isometric muscle strength. The maximal isometric plantar- and dorsiflexor strength evaluated with the two dynamometers were lower in the children with JIA than in the controls. In dynamic muscle actions, measured with the isokinetic dynamometer the controls were significantly stronger than the JIA group in shortening plantarflexor and in shortening and lengthening dorsiflexor actions. Comparisons between action types showed larger lengthening torques than isometric or shortening, and comparisons between muscle groups showed all plantarflexor torques to be four to five times greater than dorsiflexor torque. Children with JIA walked with a slower velocity than healthy controls. After intraarticular corticosteroid injections (ICI) the children with JIA increased their speed. Difference in the vertical ground reaction forces at loading response and at terminal stance was observed between children with JIA and controls wherein the peaks observed in the normal ground reaction force are attenuated in the children with JIA. The results after treatment with ICI indicated increased knee and ankle flexion extension angles during walking. Hip extension moment at loading response and knee- and plantarflexion moment at preswing increased after treatment with ICI. Positive effects of treatment with ICI were observed during gait even in joints not injected. The Pain-O-Meter word descriptors are a possible tool for assessment of sensory and affective dimension of pain in children age 6-16 yrs. The number of words, however known in small children was fewer and no difference in pain experience was observed between children with acute or chronic pain. Novelty data in this thesis are dynamic plantarflexor torque measured on children with JIA and assessment of ICI on gait. The clinical implications are numerous - weakness in almost all action types in plantar- and dorsiflexors indicates the importance of strength-training these muscle groups and the need to evaluate the muscle strength with a safe and objective method continuously. Three dimensional gait analyses provide information about gait such as joint angles, joint moments and power during walking which are impossible to quantify during a standard clinical medical examination. The findings of lower walking velocity, muscle strength and joint moments in children of JIA indicate the importance of encouraging the children to participate in physical activities as much as possible to prevent a vicious circle with deteriorating fitness.



## **PUBLICATIONS**

This thesis is based on the following publications which are referred to by their roman numerals:

- I     **E Hedengren**, Loretta M Knutsson, Y Haglund-Åkerlind, S Hagelberg  
Lower extremity isometric joint torque in children with juvenile chronic arthritis. *Scand J Rheumatol* 2001;30:69-76
  
- II    **E Broström**, M M. Nordlund , A G. Cresswell  
Isometric, lengthening and shortening muscle action in the plantar- and dorsiflexors in prepubertal girls with juvenile idiopathic arthritis. Accepted for publication in *Arch Phys Med Rehabil*
  
- III   **E Broström**, Y Haglund-Åkerlind, S Hagelberg, A G. Cresswell. Gait in children with juvenile chronic arthritis – timing and force parameters. *Scand J Rheumatol* 2002; 31:317-23
  
- IV    **E Broström**, S Hagelberg, Y Haglund-Åkerlind  
Effect of joint injections in children with juvenile idiopathic arthritis: evaluation by 3D-gait analysis. *Acta Pædiatr* in Press
  
- V     L Jylli, **E Broström**, S Hagelberg, C Stenström, G Olsson, A Langius-Eklöf.  
Sensory and emotional components of pain as recorded with the instrument Pain-O-Meter (POM) among children and adolescents. Manuscript



## **LIST OF ABBREVIATIONS**

|            |  |
|------------|--|
| ACR        | American College of Rheumatology                     |
| ANOVA      | Analysis of variance                                 |
| ASCT       | Autologous stem cell transplantation                 |
| ANA        | Antinuclear antibodies                               |
| CHAQ       | Child health assessment questionnaire                |
| Concentric | shortening   |
| COX        | Cyclo-oxygenase                                      |
| CV         | coefficient of variation                             |
| Eccentric  | lengthening  |
| ESR        | erythrocyte sedimentation                            |
| EULAR      | European League Against Rheumatism                   |
| GRF        | Ground Reaction Force                                |
| HHD        | hand held dynamometer                                |
| HLA-B27    | Human Leucocyte Antigen B27                          |
| ICI        | intraarticular corticosteroid injection              |
| ICC        | intraclass correlation coefficients                  |
| IgG        | immunoglobulin G                                     |
| ILAR       | International League of Association for Rheumatology |
| JAS        | juvenile ankylosing spondylitis                      |
| JCA        | juvenile chronic arthritis                           |
| JIA        | juvenile idiopathic arthritis                        |
| JRA        | juvenile rheumatoid arthritis                        |
| MMT        | manuel muscle testing                                |
| MTX        | methotrexate   |
| MVC        | maximal voluntary contraction                        |
| NSAID      | nonsteroidal-anti-inflammatory drugs                 |
| POM        | Pain-O-Meter   |
| RA         | Rheumatoid arthritis                                 |
| RF         | rheumatoid factor                                    |
| ROM        | range of motion                                      |
| SD         | standard deviation                                   |
| TNF        | tumor necrosis factor                                |
| VAS        | visual analogue scale                                |



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## Studies I-V

## **INTRODUCTION**

Juvenile idiopathic arthritis (JIA) affects around 1,500 children in Sweden and influences many aspects of daily life (Andersson et al., 1987; Berntson et al., 2001). A population-based epidemiological study in the Nordic countries of juvenile idiopathic arthritis (JIA) found an incidence of 15/100,000 children/year (Berntson et al., 2003). The estimated prevalence was in 1987 56/100,000 and the subgroup distribution showed a predominance of mono- and oligoarticular forms (Andersson et al., 1987). There was a peak in disease onset found in girls between 0 and 3 years of age. Forty-two percent of the girls and 34% of the boys with JIA were ANA-positive. JIA in girls predominated over JIA in boys with a ratio of 3:2 (Cassidy and Petty, 2002). Children with JIA report greater limitations in daily activities (Henderson et al., 1995; Singsen, 1995). Some of the most important problems are pain, fatigue, and stiffness and reduced physical ability (Giannini and Protas, 1992; Klepper et al., 1992; Cassidy and Petty, 2002; Schneider and Passo, 2002). Medical treatment together with physical and occupational therapy are important components of the care of children with chronic arthritis. It is important to measure pain, cardio-respiratory fitness, muscle strength and gait with accurate methods, in order to expand our knowledge of children and their arthritis.

### **Juvenile Idiopathic Arthritis**

It is impossible to define criteria which will cover the whole disease population, as some patients will not meet the criteria completely. Different classifications and sets of criteria have been used in different parts of the world. In 1972, the term juvenile rheumatoid arthritis (JRA) was established in United States (Brewer, Jr., 1973) and Canada by the American College of Rheumatology (ACR). Three onset types were described; the systemic form, the oligoarticular (or pauciarticular) form (1-4 joints), and the polyarticular form ( $\geq 5$  joints). According to the criteria, the disease should develop before 16 years of age (Brewer, Jr. et al., 1977) and the

duration must at least be 6 weeks. In Europe the term juvenile chronic arthritis (JCA) was used according to the criteria set by the European League Against Rheumatism (EULAR) (Wood, 1978). The differences from the ACR classification were disease duration of three months, and inclusion of additional subgroups such as juvenile ankylosing spondylitis, arthropathy associated with inflammatory bowel disease, and juvenile psoriatic arthropathy (Table I).

**Table I.** EULAR classification criteria for juvenile chronic arthritis (Wood et al., 1975)

|  | <b>Inclusions</b>                                   |
|--|---|
| Disease duration for diagnosis                         | 3 months  |
| Systemic arthritis                                     | Fever and rashes during at least 2 weeks, arthritis |
| Oligoarthritis   | less than 5 active joints                           |
| Polyarthritis  | 5 or more active joints                             |
| Juvenile Ankylosing Spondylitis                        | HLA-B27 positive, Enthesitisrelated arthritis       |
| Arthropathy associated with inflammatory bowel disease | Ulcerous colit, Mb Chron seronegativ arthritis      |
| Juvenile psoriatic arthropathy                         | seronegative arthritis and psoriasis                |

In 1995, the Paediatric Standing Committee of International League of Associations for Rheumatology (ILAR) proposed a new set of criteria, using the term “JIA”, and in 1997 the Durban criteria were set (Petty et al., 1998). The ILAR criteria introduced a new classification aspect by list exclusionary criteria. The purpose was to find groups which were biologically homogenous (Table II). In this thesis the term JIA will be used.

**Table II.** ILAR classification criteria for juvenile idiopathic arthritis (Petty et al., 1998)

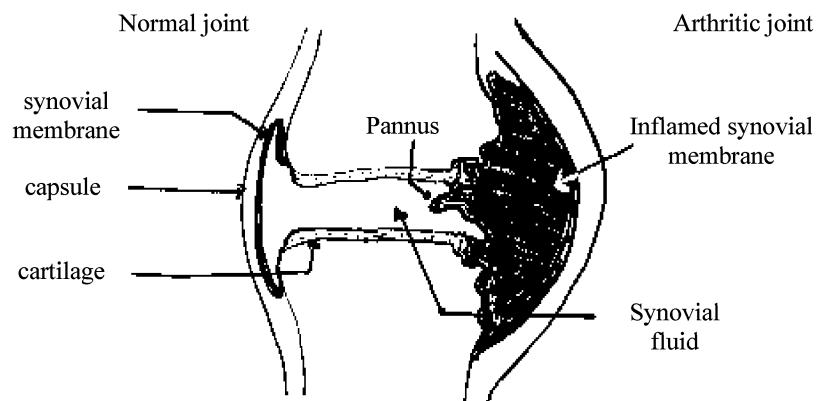
|                                | <b>Inclusions</b>  | <b>Exclusions</b>   |
|--------------------------------|--|---|
| Disease duration for diagnosis | 6 weeks  |   |
| Systemic arthritis             | daily fever with a duration of at least 2 weeks, sometimes with arthritis  | Not listed  |
| Oligoarthritis                 | arthritis affecting 1 to 4 joints  | Family history of psoriasis confirmed or consistent with medically confirmed HLA-B27, positive RF. Presence of systemic arthritis |
| Extended oligoarthritis        | affects a cumulative total of 5 joints or more after 6 months of disease.  |   |
| Polyarthrititis (RF negative)  | affects 5 or more joints with negative RF  | presence of RF. Presence of systemic arthritis  |
| Polyarthrititis (RF positive)  | affects 5 or more joints with positive RF  | Absence of positive test for RF on 2 occasions at 3 month apart. Presence of systemic arthritis                                   |
| Psoriatic arthritis            | arthritis and psoriasis and at least two of the following: dactylitis, nail pitting, family history of psoriasis | Presence of RF and of systemic arthritis.   |
| Enthesitis-related arthritis   | sacroiliac joint tenderness, HLA-B27, onset of arthritis in boys after age of 8 years                            | Psoriasis confirmed by a dermatologist in at last one first or second degree relative. Presence of systemic arthritis.            |
| Other arthritis                | unknown origin   | Patients who meet criteria for other categories.  |

## Disease Characteristics

The long-term prognosis for children with JIA ranges, depending on the disease onset type. It is estimated that 50 % of all children diagnosed with JIA will recover completely (Andersson, 1999). Better prognosis has been reported for oligoarticular onset at early age (Gare and Fasth, 1992). Worse prognosis has been

reported for polyarticular and systemic onset (Andersson et al., 1987; Andersson, 1999). It is usually not possible at the onset of the disease to predict which child will recover and which will have a long-standing and destructive disease.

However, in systemic onset JIA trombocytosis and persistent arthritis after 6 month of disease is associated with poor outcome (Spiegel et al., 2000). Early diagnoses and active therapeutic interventions are essential to minimize residual deformity and disability due to joint contractures and asymmetric bone growth (Sherry et al., 1999; Cassidy and Petty, 2002; Sharma and Sherry, 1999). The classical signs of inflammation are heat sensation, pain, redness and swelling all of which reflect the effects of inflammatory mediators such as cytokines, which are proteins that act as messenger between cells. An inflamed or “active” joint, is usually swollen and warm. In the absence of swelling, both tenderness and decreased range of motion (ROM) is required for the joint to be regarded as “active”. Autoimmune diseases occur when a specific adaptive immune response is mounted against self antigens. The mechanism behind the autoimmune response in rheumatoid arthritis is still incompletely understood. The predominating theory involves an imbalance between two types of T-lymphocytes (Th1 and Th2), in which the Th1 cells activate macrophages, giving rise to an overproduction of pro-inflammatory cytokines like for example tumour necrosis factor (TNF) and interleukin-1 (IL-1). TNF and IL-1 have been shown to play an important role in the arthritic process and is now succesfully being targeted in rheumatic patients, by means of specific TNF-antibodies (infliximab, adalimumab), soluble TNF-receptor (etanercept) or IL-1-receptor-antibodies (anakinra). These cytokines are abundant in the hypertrophic synovia called pannus, which is responsible for the cartilage destruction in the joint (Cassidy and Petty, 2002) (Figure 1).



**Figure 1** Schematic illustration. Normal joint (left) and arthritic joint (right).

### ***Systemic onset***

The systemic form of JIA is characterised by rashes, fever, arthritis, hepatosplenomegaly, generalized lymphadenopathy and serositis. Sometimes arthritis does not develop until several months into the systemic disease which may delay the diagnosis (Cassidy and Petty, 2002).

### ***Oligoarticular onset***

Oligoarticular (pauciarticular) onset has four or less joints involved and comprises sixty percent of children with JIA. Some of these patients have just one joint affected, primarily the knee and secondarily the ankle (Gare and Fasth, 1995; Sharma and Sherry, 1999). It is especially important to observe young girls that are antinuclear antibody (ANA) –positive since they are at a greater risk of developing chronic uveitis (Kotaniemi et al., 2001; Kotaniemi, 2002). The chronic uveitis of JIA is risky and typically runs an asymptomatic course in the initial stage. This may delay diagnosis and treatment which could lead to permanent visual disturbance and even blindness (Saila et al., 2001) (Table III).

**Table III.** Oligoarthritis affects almost 60% of children with JIA.

| Type I       | Type II                  |
|--------------|--------------------------|
| Girls        | Boys                     |
| Early onset  | Later onset              |
| Large joints | Large joints             |
| ANA positive | ANA negative             |
| RF negative  | RF negative              |
| Uveitis      | Spondylitis/sacroiliitis |

### ***Polyarticular onset***

The polyarticular form of the disease (25-35% of patients) involves five or more joints and tends to be symmetrical. It generally involves large joints as the knees, wrists, elbows, ankles, and the small joints of the hand, feet and the cervical spine (Cassidy and Petty, 2002). The frequency of rheumatic factor (RF) in children with chronic arthritis is much lower than in to adults with RA. Teenage girls, with rheumatic factor have a less favourable outcome and may be regarded as having an early onset of adult rheumatoid arthritis (Gare and Fasth, 1992;Cassidy and Petty, 2002).

### ***Juvenile ankylosing spondylitis onset***

The juvenile ankylosing spondylitis (JAS) is a group of HLA-B27-associated disorders, mainly characterized by enthesitis and arthritis, affecting the lower extremities and, in a variable proportion of cases, the sacroiliac and spine joints (Burgos-Vargas, 2002). Radiological changes of sacroiliitis in childhood are not found until a later point. A study has shown that sacroiliac changes do not occur before the age of eight (Brophy et al., 2002). Radiological spinal involvement is usually not apparent until late in the adolescence. Uveitis is not uncommon in JAS but is usually acute and symptomatic, making it easier to detect early (Petty et al., 1973).

## **Clinical symptoms**

Juvenile arthritis may have a profound effect on the life of a child. Symptoms of JIA are stiffness in the joints in the morning and after a period of inactivity. The stiffness is sometimes felt only in the affected joints; sometimes it overwhelms the child as a whole. Inflamed joints are tender and painful; pain can be bothersome particular at night, especially in the polyarticular form of the disease. At onset of the disease a diffuse pain, swelling of joints and an avoidance of loading are often observed.

Apart from articular symptoms, weight loss and fatigue may also occur in all subtypes of JIA (Cassidy and Petty, 2002;Schneider and Passo, 2002). Long duration of an active disease is also associated with a reduction in height, discrepancy in leg length, and shortening of muscles and tendons that may give rise to flexion contractures (Bacon et al., 1990;Fan et al., 1998;Lindehammar and Backman, 1995;Vostrejs and Hollister, 1988). Other findings include a reduced level of physical activity (Giannini and Protas, 1992;Henderson et al., 1995;Klepper et al., 1992;Klepper and Giannini, 1994;Klepper, 1999;Takken, 2003), lower aerobic endurance (Giannini and Protas, 1991;Klepper, 1999;Takken et al., 2002), and decreased muscle strength (Giannini and Protas, 1993;Lindehammar and Backman, 1995;Lindehammar and Sandstedt, 1998). The inflammatory process of the disease most often affects extremities and a restriction of joint motion can occurs.

## **Laboratory indicators**

Laboratory investigations help the clinician to diagnose JIA by excluding other diseases, to subgroup the arthritis, to monitor side effects of pharmacological treatment and evaluate the disease activity. The use of erythrocyte sedimentation (ESR) is one of six outcome variables used to indicate for disease activity of children with juvenile arthritis (Giannini et al., 1997). Nevertheless, Giannini and

Brewer showed (1987) that the ESR is a poor indicator of the amount and status of articular inflammation while one study by Hussein (1987) found that children with an unfavourable functional outcome more often had high ESR. Approximately 60% of the JIA patients are ANA-positive, but do not have the specific auto-antibodies commonly found in collagenosis patients.

## **Imaging**

X-ray, magnetic resonance imaging and computerized tomography scans are important when monitoring and following up disease development. They are also used to exclude differential diagnoses, such as malignancies (Cabral and Tucker, 1999;Cohen et al., 2000;Miller et al., 1995;Trapani et al., 2000). Ultrasound is easy to use and a very helpful tool for assessing effusion and pannus and for evaluating treatment response (Miller, 2002;Poznanski, 1992) – especially in the hip.

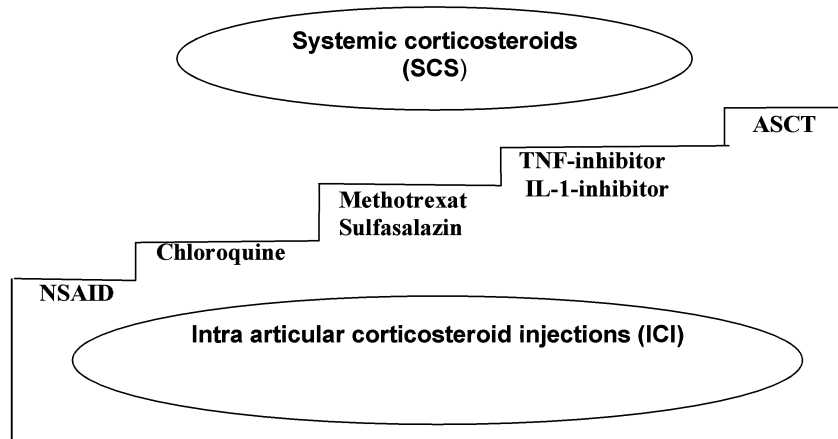
## **Treatment**

### ***Pharmacological***

The outcome for patients with JIA has improved significantly during the past decade (Milojevic and Ilowite, 2002). Better understanding of the biology of inflammation has fuelled the search for more targeted therapeutic agents with fewer side effects. Traditional non- steroidal anti-inflammatory drugs (NSAIDs) continue to be the primary therapy for most children with JIA (Cassidy and Petty, 2002). It is commonly used initially during treatment, in addition to Methotrexate and when disease flares-ups are intermittent and mild to moderate. The new, COX-2 (Cyclo-oxygenase) inhibitors have not been studied in the paediatric population, but have been associated with less gastrointestinal toxicity (Milojevic and Ilowite, 2002;Murray and Lovell, 2002).

Chloroquine is occasionally used for treatment of dermatitis, arthritis and serositis (Milojevic and Ilowite, 2002). Methotrexate (MTX) has become the second

pharmacological step of choice for persistent, active arthritis and the effect has been demonstrated in a placebo-controlled double-blind randomized study (Giannini et al., 1992). Sulfasalazine is used for treatment of inflammatory bowel disease in children and in juvenile-onset spondyloarthritides (Burgos-Vargas, 2002). The recent introduction of antitumour necrosis factor (anti-TNF) agents has been a revolution in the treatment of rheumatic disorders. Lovell et al (2000) reported excellent results after treating children with polyarticular onset of JIA with etanercept subcutaneously twice a week for 7 months and after 2 years of treatment with etanercept (Lovell et al., 2003). The use of TNF-inhibitors in Sweden has not been common due to its lack of availability but is expected to become more widespread due to its fairly recent release. Autologous stem cell transplantation (ASCT) has emerged as a possible treatment for a small group of children with JIA that are resistant to pharmacologic of treatment (Schneider and Passo, 2002;Fasth and Andersson-Gare, 2000). Corticosteroids should be avoided in oral form for prolonged use in the treatment of JIA, due to their long-term side effects, such as growth retardation and osteoporosis (Byron et al., 1983;Bowyer et al., 2003;Murray and Lovell, 2002;Loeb, 1976;Zak et al., 1990). Short-term use is occasionally warranted in cases of very active disease. Intra articular corticosteroid injection (ICI) are very effective and is commonly used in children with JIA (Huppertz et al., 1995;Padeh and Passwell, 1998). It is reported that this treatment is safe and provides continued anti-inflammatory effects on the synovium. In one study 82% of 300 joints that were injected demonstrated full clinical remission of the joint inflammation (Padeh and Passwell, 1998). There is also evidence that leg length discrepancy resulting from unilateral knee synovitis as well as joint deformity and joint damage may be prevented (Sherry et al., 1999). Long-term follow up with magnetic resonance imaging after repeated ICIs in knees of JIA patients showed preserved cartilage (Hagelberg et al., 2000)(Figure2)



**Figure 2.** The pharmacotherapy step-ladder in choice of treatment. ICI is used for residual active joints of any stage of treatment. SCS is used for temporary use in very active disease.

### ***Physical and occupational therapy***

Physiotherapy for children and adolescents with JIA assumes a central position. The treatment includes ROM exercise (passive/active) and pain relief, with for example, heat/cold treatment, acupuncture or transcutaneous electrical nerve stimulation (TENS) and splinting. Other contributions which could be made by physiotherapy are guidance in choosing footwear that absorbs shock during loading response and in exercising to optimize strength training and aerobic or water exercise with their natural limitations. It is important to start with an individualized exercise program soon after diagnosis (Klepper and Giannini, 1994; Klepper et al., 1992).

### **Muscles and strength**

Research into paediatric muscle composition is rare, and the current understanding of muscle metabolism in children is based on a small number of muscle biopsy studies conducted more than 25 years ago on a group of 11- to 15-year-old boys (Eriksson et al., 1971; Eriksson et al., 1973; Eriksson and Saltin, 1974). The invasive

procedures (biopsies) raise ethical concerns, but it is possible to use less invasive methods such as magnetic resonance imaging describing the architecture of the muscles. The muscle architecture describes the organization of the muscle fibres within a muscle relative to the axis of force generation (Lieber and Friden, 2001).

The architecture of the muscle is important for the development of force and for flexibility in that the muscle force is proportional to the physiologic cross-sectional area of the muscle fibres, whereas the contraction velocity of the muscle is proportional to the muscle length (Lieber and Friden, 2000;Lieber and Friden, 2001). Contractility properties in childhood are proportionally to increased muscle fibres diameter and increased number of sarcomeres (Asai and Aoki, 1996).

There are two main types of muscle fibres, referred to as Types I and II. Type I fibres are better suited for sustained periods of activity at low tension levels such as walking, long distance running and most functional activities of daily life. Type II fibres fatigue easily but are better suited for rapid, high power explosive contractions (Enoka, 1994). The distribution of fibres types varies between individual muscles. For example the soleus muscle is dominated by Type I fibres, and gastrocnemius by Type II fibres.

Muscle strength and muscle functions are important factors when following the progress of the disease. It is therefore vital to be aware of developmental changes in strength due to neurological maturation, muscle tissue changes, body proportion changes, sex, age, and day-to-day variations (Beunen et al., 1992;Beunen et al., 1997). Muscle atrophy, with an associated reduction of strength, is characteristic in children with JIA (Fan et al., 1998;Giannini and Protas, 1993;Lindehammar and Backman, 1995). There is also evidence that children with early onset of unilateral knee arthritis are at greater risk for muscle atrophy than children whit onset after 3 years of age (Vostrejs and Hollister, 1988). An earlier study of children with this

disease reported a reduced cross-sectional area of the quadriceps and a reduction in quadriceps strength regardless of whether knee inflammation was active (Lindehammar and Sandstedt, 1998).

Muscle weakness and reduction in strength have been shown to have a profound effect on the level of physical activity of children with JIA (Henderson et al., 1995; Klepper et al., 1992; Klepper and Giannini, 1994; Singsen, 1995; Takken et al., 2001; Takken, 2003). There is, however, no correlation between physical fitness and the severity of the articular disease (Giannini and Protas, 1993; Klepper et al., 1992; Klepper, 1999).

Children and adolescents with chronic polyarticular onset as well as adults with rheumatoid arthritis can improve their aerobic endurance without increased disease activity, increased pain or radiological progression of joint disease, through participation in physical conditioning programs (Giannini and Protas, 1991; Takken et al., 2002; Augustsson et al., 1998; Stenstrom and Minor, 2003; Stenstrom et al., 1999; Stenstrom, 1994; Stenstrom et al., 1991). Nordemar et al found that exercise increases the size of Type II muscle fibres of adults with arthritis after aerobic exercise on a bicycle and self-selected exercise such as swimming, jogging or cycling (Nordemar and Ekblom, 1981; Nordemar et al., 1976b; Nordemar et al., 1976a; Nordemar, 1979). Stenström et al have in a review article reported that exercise can be performed in adults with RA (Stenstrom and Minor, 2003). Alexanderson (2003) showed that patients with poly- and dermatomyositis were able to change some Type I fibers into Type II fibers, approaching normal proportions, after 12 weeks exercise.

Three types of skeletal muscle contractions can be observed: lengthening contractions (eccentric contraction), where sarcomeres are stretched while trying to contract and generate the greatest amount of tension, isometric where no change in

muscle length occurs, and shortening (concentric contraction) wherein the force generated by the muscle is always less than that in lengthening or isometric contractions. These different types of muscle contractions allow skeletal muscles to function as springs, pulleys, shock absorbers and stabilizers (Enoka, 1994).

## **Strength testing**

Physical therapists have used manual muscle testing (MMT) to evaluate muscle strength in children (Kendell and McCreary, 1983;Hinder and Hinderer, 1993;Kendell and McCreary, 1983;Kroksmark et al., 2001). MMT often results in large measurement variability, and its suitability for accurate strength assessment has been questioned (Hinder and Hinderer, 1993).

Using a hand-held dynamometer (Backman et al., 1989;Bohannon, 1999;Boiteau et al., 1995;Lindehammar and Backman, 1995;Lindehammar and Sandstedt, 1998;Wessel et al., 1999) provides more objective estimates of isometric strength than MMT. It has been used to measure lower extremity isometric strength in healthy children (Backman et al., 1989;Lindehammar and Backman, 1995) and adults with arthritis to evaluate training (McGibbon et al., 2003).

A more objective measurement device for measuring strength is the isokinetic (constant-velocity) dynamometers. The use of isokinetic dynamometers is established in the measurement of muscle strength assessment in both clinical and sports settings and shows highly reliable measures of strength in both healthy and diseased populations (Pinniger et al., 2000;Westing and Seger, 1989;Ayalon et al., 2000;Bohannon, 1999;Boiteau et al., 1995;Holmback et al., 1999;Baltzopoulos and Brodie, 1989).

## **Gait analysis**

Gait analysis is a method for documenting disease progression quantitatively (Gage, 1991). It may offer important information about children's gait patterns, thus helping physicians to make better treatment decisions (Rose et al., 1991; Perry, 1994; Sutherland et al., 1980). An early use of gait analysis can be instrumental in discovering developments of potentially destructive gait deviations (Fairburn et al., 2002; Frigo et al., 1996). Some studies (Beck et al., 1981; Cupp et al., 1999; Ounpuu et al., 1991; Sutherland et al., 1980) have found that gait in children seven years of age is similar to that of an adult.

A fundamental law of static physics is that a force is balanced by reaction force. In standing the amount of vertical ground reaction force under both feet is equal to the weight of the subject. During walking an additional component of force is due to the acceleration of the body. Ground reaction force (GRF) has a characteristic "butterfly shape" during stance phase with two peaks force (Winter, 1990). The amplitude of these forces peak has been shown to correlate with the walking speed and stride length during normal walking (Nilsson and Thorstensson, 1989). The force plate is one of the most important measurement devices in biomechanics, quantifying external forces during human locomotion (Winter and Wells, 1981; Winter, 1990) and has high accuracy and precision in measuring force.

Today, it is possible to analyze gait by using a biomechanical model based on the measured position of markers placed on the subjects' skin to infer the positions of body segments (Sutherland et al., 1980; Rose et al., 1991; Stolze et al., 1997; Perry, 2002; Davis and Ounpuu, 1991). The most common model includes the lower limbs as seven rigid segments (one pelvis, two each of thigh, shank and foot) which are defined by 13 markers (Kadaba et al., 1990; Ounpuu et al., 1991; Ounpuu, 1994). Three dimensional gait analyses, including kinematics and kinetics, provide objective information about gait changes, such as joint angles and moments, which

are impossible to examine clinically. The kinematics shows the description of joints movement, including joint segment angles, velocities and accelerations. Kinetics describes the mechanisms that cause movement (e.g., ground reaction forces, joint moments, and joint powers). By combining segment motion and forceplate information, moments and powers can be calculated using a process called inverse dynamics. A moment of force is force acting at a distance from the joint axis of rotation, causing the body to rotate. It is expressed as  $M = F \times l$  ( $M$ =moment of force,  $F$ =force and  $l$ =lever arm) and is presented in units of Newton-meters (Nm). By examining kinetics, we can determine why a particular gait deviation occurs. To evaluate the effect power can be calculated. Joint powers combine both kinematic information (angular velocity) and kinetic information (moments) and appears to be a good indicator of a person's ability to drive and control the lower limbs (Vardaxis et al., 1998). It is important to understand lower-limb muscle power relationships contributing to control and propulsion during gait and could be useful in distinguishing between asymmetries caused by a disability and those resulting from compensations. Power are defined as rate at which work is done or energy is expended (Winter, 1990). The power does not, however, give information about why the muscles contract. To calculate more specific muscle timing, electromyography must be used. Time and distance parameters, such as gait cycle, support and swing phase durations, vary with the walking velocity (Beck et al., 1981;Andriacchi et al., 1977). An important factor is the walking velocity's effect on moments and power (Andriacchi et al., 1977;Oberg et al., 1993;Ounpuu et al., 1991). Since walking velocity is known to vary with age, and more importantly with factors such as height and leg length, it is important to normalise walking velocity before comparing subjects and evaluating treatment effects (Frigo et al., 1996;Oberg et al., 1993;van der Linden et al., 2002;Wheelwright et al., 1993a;Wheelwright et al., 1993b).

To describe gait pattern adequately a certain terminology must be adopted. The gait cycle begins when one foot comes in contact with the ground and ends when the same foot contacts the ground again. The stance phase of gait refers to the period in which the foot is in contact with the ground and the swing phase is when that foot is being advanced. Double support describes the phase when both feet are in contact with the ground. The stance phase is approximately 60% of the gait cycle, and includes two periods of double support.

Stance can be divided into five phases. The first is initial contact as the heel reaches the ground followed by loading response. The third stance phase is mid-stance and the fourth is terminal stance, when the heel rises from the ground. The fifth and last stance phase is pre-swing when the foot leave the ground (Ounpuu et al., 1991;Perry, 2002;Sutherland et al., 1980;Winter and Wells, 1981). The swing has three phases; the first phase is initial swing followed by the mid swing phase. The last phase, terminal swing, ends the entire gait cycle.

In this thesis focus is placed on the foot and ankle. Ankle sagittal plane movements are divided into three phases, called “rockers” (Perry, 2002). The first rocker occurs when the fore foot is lowered towards the floor after initial contact. The second rocker occurs when the tibia advances over the fixed foot in midstance. The third rocker occurs when the heel rises and loading is on the forefoot.

### **The gait pattern in JIA**

The gait of children with JIA can differ from that of healthy children and a common clinical observation is that children with JIA have a reduced walking speed. Several studies have specifically addressed this issue and it has been shown that children with JIA walk more slowly than healthy children of the same age (Lechner et al., 1987;Witemeyer et al., 1981). Interestingly, however, is that after normalisation to height no significant difference in walking velocity between JIA

and healthy children has been found (van der Linden et al., 2002). It has been reported that children with JIA had a reduced vertical ground reaction force during push-off, and both an increase in hip flexion angle and a decrease in knee flexion angle during the late phase of stance as compared to controls of the same age (Beck et al., 1981; Fairburn et al., 2002; Frigo et al., 1996).

### **Pain in children**

A large number of studies have demonstrated that pain has both sensory and affective dimensions. The International Association for the Study of Pain Subcommittee on Taxonomy describes pain as “an unpleasant sensory and emotional experience associated with actual or potential tissue damage” (Merskey and Bogduk, 1994).

Chronic pain differs from acute pain in that it is based on the duration of pain, more or less than three months (Merskey and Bogduk, 1994). A common adverse nociceptive stimulus is pain after surgical procedures (Romsing and Walther-Larsen 1996). Children with JIA suffer from daily symptoms of pain, stiffness, and fatigue, which leads to reduced participation in school and social activities (Schanberg et al., 2003). While mild to moderate pain has been reported in numerous studies (Sherry et al., 1990; Gragg et al., 1996; Lovell and Walco, 1989; Gragg et al., 1996; Varni et al., 1988), Schanberg et al. have shown that 25% of children with JIA experience high intensities of pain (Schanberg et al., 1997). According to a study by Sherry et al. (1990), 86% of children with juvenile rheumatoid arthritis (JRA) reported pain during a routine control at the clinic, and Lovell and Walco found that 60% of children with JRA reported joint pain at onset, with 50% still reporting pain 1 year later, and 40% reporting pain at 5-year follow-ups (Lovell and Walco, 1989). Consequently, pain is a factor that cannot be ignored when treating children with arthritic diseases.



## **SPECIFIC AIMS**

The aim of this thesis was to evaluate muscle strength and pain in children with JIA, and to analyze gait patterns before and after intraarticular corticosteroid injections. The specific aims were:

1. To evaluate muscle strength in lower extremities with a hand held dynamometer in JIA, which is a simple and clinically feasible test
2. To evaluate muscle strength with a computerized dynamometer in both controls and children with JIA to gain improved accuracy of strength estimates.
3. To evaluate ground reaction forces and time parameters in JIA and compare them to healthy children of the same age.
4. To evaluate gait patterns before and after intraarticular corticosteroid injection on gait in children with JIA.
5. To identify pain descriptors in children and adolescents with acute and chronic pain.



## MATERIALS AND METHODS

### Subjects

In Studies I and III the children with JIA were diagnosed according to the EULAR classification system. In Studies II, IV and V the children with JIA were diagnosed according to the criteria set by the ILAR. All of the patients were treated regularly at the Department of Pediatric Rheumatology, Astrid Lindgren Children's Hospital, Karolinska Hospital in Stockholm. They were all independent walkers and had lower extremity involvement. In Study II only girls with polyarticular involvement participated (Table IV). The Research Ethical Committee of the Karolinska Hospital had approved all five studies. Verbal and written information was given to the children and their parents.

**Table IV** Distribution of the subjects in the different studies.

| Study | Subject classification and gender            | Age        | Overlapping subjects                | Disease Subgroup <sup>1</sup>           |
|-------|--|------------|-------------------------------------|---|
| I     | JCA <sup>2</sup> = 4m, 7f<br>Controls=6f, 8m | 5-15 years |                                     | Poly=7,<br>syst=1,<br>oligo=1,<br>JAS=2 |
| II    | JIA <sup>2</sup> =10f<br>Controls=10f        | 8-12 years | From Study I=2                      | Poly=10                                 |
| III   | JCA=11f, 4m<br>Controls=8f, 6m               | 6-14 years | From Study I=3<br>From Study II=3   | Poly=11,<br>oligo=4                     |
| IV    | JIA=15f, 3m                                  | 5-16 years | From Study II=5<br>From Study III=1 | Poly=13,<br>oligo=3                     |
| V     | JIA= 29f, 7m<br>acute pain=8f, 17m           | 6-16 years |                                     |   |

<sup>1</sup> Poly=polyarticular arthritis, syst=systemic arthritis, oligo=oligoarticular arthritis, JAS=juvenile ankylosing spondylitis

<sup>2</sup> JCA=juvenile chronic arthritis, JIA=juvenile idiopathic arthritis

## **Equipment**

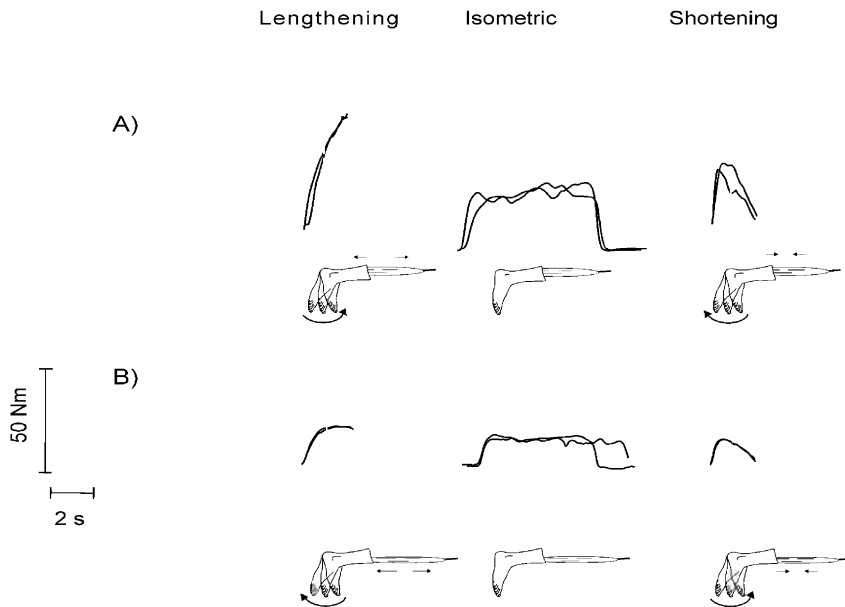
### ***Hand held dynamometer (HHD)***

In Study I the CompuFet HHD (Hoggan Health Industries, Draper, USA) was used to measure torque. The dynamometer was applied at an angle approximately perpendicular to the line from the joint axis to the site of HHD placement. Force values were provided in units of pounds and were later converted to Newtons during data analysis. When testing knee joint torque (knee extensors and knee flexors) the subjects lay on the left side with the right knee flexed to 60 degrees as measured with a goniometer. For testing the ankle joint torque (plantar- and dorsiflexors), the subject was positioned supine with hip, knee and ankle of the tested extremity at neutral. Five repetitions of isometric contractions, each held for five seconds, were completed. The lever arm was calculated with a standard plastic coated tape.

### ***Isokinetic dynamometer***

Torque was measured again in Study II, but this time with a computerized dynamometer. The left foot was secured to a steel plate, which was mounted to the torque motor with screws.

The position of the foot was adjusted to align the axis of the ankle joint with the axis of the torque motor. Plantar- and dorsiflexion ankle joint torques were measured using a torque transducer located in the axle of the torque motor. The signal was amplified and 10 Hz low-pass filtered before undergoing A/D conversion at a sample rate of 100 and signal data collection software. A 30° ROM was used for strength testing if the subjects were able to comfortably move through 15° of plantar flexion and 15° of dorsiflexion from the neutral position. Torque was measured for three muscle action types; lengthening, isometric and shortening (Figure 3).



**Figure 3** Torque about the ankle during lengthening isometric and shortening plantarflexor action and dors flexor action. Two consecutive trials from a representative subject are overlaid to demonstrate the reproducibility of the data

### ***Ground reaction forces***

Force plates (9281CA, Kistler, Switzerland) were used in Studies III and IV to measure ground reaction forces during gait. A walking trial was considered completed if the subject's right or left foot made a clean contact on the force plate. Force plate data was sampled at 240 Hz using SC/ZOOM, software (Umeå University, Sweden) in Study III. One force plate was used in Study III, while two were used in Study IV and sampled at 1000 Hz in Vicon workstation (Vicon, Oxford Metrics, England).

### ***Temporal parameters***

In Study III laboratory built foot-switches (round plastic coated switches, each with a thickness of a diameter of 35mm) and were used to measure temporal parameters. Foot switch data were sampled at 100 Hz. The data collection system was triggered

by a light-beam at the beginning of the walkway. A second light-beam placed 5 meters further down the walkway was used to determine the end of the walking session.

### ***Movement analysis***

In Study IV the movement recordings were done with a 6-camera 3D motion analysis system (Vicon, Motion System, Oxford, England) (Davis and Ounpuu, 1991). Thirty-four reflective markers (25mm) were attached bilaterally on the subject's skin at the head, shoulders, arms, pelvis, legs and feet according to the biomechanical gait model (Plugin Gait, Vicon Motion Systems). Combining kinematics of motion and force plates data joint moments can be calculated. A total of three completed walking trials were recorded. A second gait analysis was performed 8-17 days after treatment with ICI.

### **Pain and function assessment**

#### ***Visual analogue scale (VAS)***

In Studies III-V the VAS was used to measure the intensity of pain. The VAS is a 10 cm long horizontal line with a movable bar with the words "no pain" at the left and "worst possible pain" at the right. The children were asked to evaluate their pain by adjusting the moveable bar, and numerical equivalent on the reverse side was recorded.

#### ***Pain-O-Meter (POM)***

In Study V the POM was used. The POM is a hand-held pain assessment tool that is made of hard plastic (Gaston-Johansson, 1996). The POM consists of a 10 cm vertical VAS with the words "no pain" at the bottom and "worst possible pain" at the top. Furthermore, it includes the POM-WDS, which is a word descriptor scale that consists of 12 sensory words and 11 affective words. Each word has an assigned intensity value with 1 representing the lowest intensity and 5 representing 24

the highest. Children were first asked, for each word whether or not they understood the words, and if so, whether the word described their pain. Scores were calculated based on the number of words chosen and their respective intensity values.

### ***Childhood Health Assessment Questionnaire (CHAQ)***

The CHAQ was used in Study IV to assess the functional and health status in eight areas: dressing and grooming, arising, eating, walking, hygiene, reach, grip, and activities. The questions range from 0-3, (0= able to do with no difficulty, 1= able to do with some difficulty, 2= able to do with much difficulty, 3= unable to do). The mean score of the eight domains provide the CHAQ disability scale. The children answered the questionnaire only before ICI treatments, not after.

### ***Goniometer***

Prior to the gait sessions in Study III and IV, a goniometer was used to measure the Range of Motion (ROM) in the lower extremities.

### ***Intra articular corticosteroids injections (ICI) (Study IV)***

Methylprednisolon acetate (Depo-Medol™) with lidocain was used for ICI (80 mg in hip; 40 mg in knee; 20 mg in ankle). Triamcinolone hexacetonid (Lederspan™, Aristospan™) is considered as the most effective and long acting steroid for intra-articular use but at the time for study IV it was not available. After ICI the patients were not allowed to walk for 24 hours. Thereafter normal physical activity was permitted, but strenuous exercise the first week was advised against.

## **Data analysis**

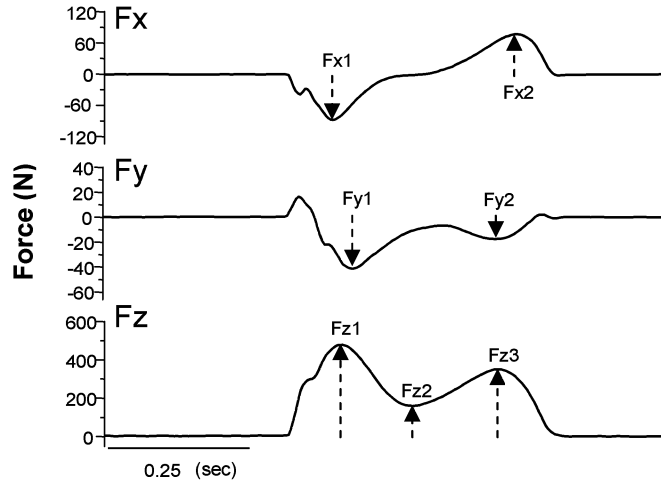
### ***Studies I-II***

In Study I force data in pounds were converted to Newtons, the preferred SI-unit of measurement, through multiplication by the constant 4.4482. Distance data were entered as meters and multiplied by the force in Newtons to obtain torque data in Newton-meters. Three maximum voluntary contractions (MVC) were performed.

In Study II plantarflexor torque were defined as positive and dorsiflexor torque as negative. For isometric maximal voluntary actions, the mean throughout the highest 1-s period of torque was used. Only the trial providing the highest torque value was used for further analysis.

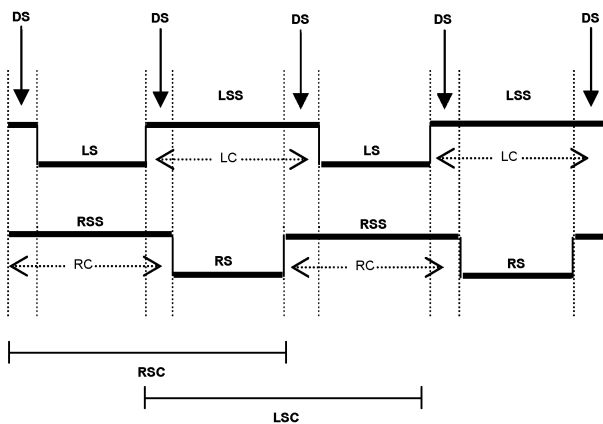
### ***Studies III-IV***

In Studies III and IV ground reaction force data was analysed in the anterior-posterior, medio-lateral and vertical directions (Fx, Fy and Fz, respectively). Each force measurement was normalized to the subject's body weight (BW) and analyzed to determine the magnitude and timing of defined points in the ground reaction force data (Figure 4). In the anterior-posterior direction, the first negative peak, indicating a backward directed force, was defined as Fx1. The forward directed push off force was seen as a late positive peak and was defined as Fx2. For the medio-lateral force, Fy1 and Fy2 were defined as the first and second medially directed force peaks. In the vertical direction, two peaks (Fz1 and Fz3) separated by a valley (Fz2) were clearly definable. Fz1 corresponded to the initial maximal loading of the force plate, Fz2 to mid-stance and Fz3 to terminal stance (the peak force just prior to heel lift off).



**Figure 4** Representative force plate traces from one child in the control group showing anterior posterior (Fx), mediolateral (Fy) and GRF (Fz).

Footswitch data were recorded bilaterally in Study III and subdivided into the gait phases of right and left contact (RC and LC), right and left single support (RSS and LSS), right and left swing (RS and LS), and double support (DS) (Figure 5). A stride cycle (SC) was defined as the time between subsequent RC or LC events. In study III all analyses were performed using the Matlab software package (The MathWorks, Inc, USA).



**Figure 5.** A schematic diagram describing events throughout the gait cycle. DS=double support, LSS=left single support, RSS=right single support, LS=left swing, RS=right swing, LC=left contact, RC=right contact, LSC=left stride cycle and RSC=right stride

In Study IV kinematics, kinetics, and time-distance parameters were obtained from the gait analysis data. The time-distance parameters were correlated to CHAQ. Ground reaction forces, moments and powers were normalized to body weight. Stride parameters were normalized to height. An ensembled, or point-to-point average, gait cycle was generated, from which gait patterns of the hip, knee and foot joint angles and moments were derived both before and after treatment with ICI. Joint power was defined as the scalar product of the moment and the joint angular velocity. Peak values from the subjects' ensembled gait cycle were analyzed of hip flexion and extension angles, hip flexion, extension and abduction moments, knee flexion and extension angles, knee flexion, extension and varus moments, ankle plantarflexion and dorsiflexion angles; plantarflexion and dorsiflexion moments, and power generation and absorption at the ankle.

## **Statistics**

In Study I the torque data was imported to SPSS (Statistical Package for the Social Sciences), and in Studies II-V the Statistica software package (StatSoft Inc, USA) was used for statistical analysis. The level of significance for main effects and interactions within the analysis of variance (ANOVA) was set to  $p \leq 0.05$ . A tendency toward a significant difference was considered if the level of significance was  $0.05 \leq p \leq 0.09$ .

In Study I intraclass correlation coefficients (ICCs) were calculated to assess the intratester reliability in order to evaluate differences between five or three trials. ICCs are included in the ANOVA procedure in which differences across trials is addressed. A paired t-test was used to compare the mean isometric torque of the patients and their age-matched control subgroups. In Study II a three-way ANOVA (subject group, action type, and muscle) with repeated measures of two factors

(action type and muscle group) was used. The level of significance was Bonferroni adjusted to correct for multiple comparisons.

In Study III an analysis of variance (ANOVA) was used to assess differences in ground reaction forces, contact times and stride frequencies between children with JCA and healthy controls. The Mann-Whitney U Test was used to compare time and stride variables between the two groups. Bonferroni corrections were made for pair-wise post-hoc comparisons, and Spearman's rank correlation coefficient was used to identify any relationship between pain (VAS) and walking velocity. The coefficient of variation ( $CV = SD / \text{mean} \times 100$ ) was used to describe variability.

In Study IV two-way analysis of variance (ANOVA) and repeated measures design were used to assess the kinematics and kinetics data from the 3D-gait analysis. The factors were side (left and right) and treatment (before and after ICI). The non-parametric Wilcoxon matched pair test was used to compare velocity and distance parameters before and after treatment with ICI, while Friedman ANOVA by rank test was used to identify pain (VAS). In Study V nominal variables differences between groups were evaluated using Chi-squared Test, and for two independent samples Student t-Test. The Wilcoxon Signed-Rank Test was used to determine difference between two related groups on a nominal level (Table V)

**Table V** Statistical methods used in study I-V

| Methods                                    | Study I | Study II | Study III | Study IV | Study V |
|--|---------|----------|-----------|----------|---------|
| ANOVA repeated measure                     | x       |          |           |          |         |
| Two-way of variance ANOVA                  |         |          | x         | x        |         |
| Three-way ANOVA                            |         | x        |           |          |         |
| Intraclass correlation coefficients (ICCs) | x       |          |           |          |         |
| Paired student t-test                      | x       |          |           |          | x       |
| Mann Whitney U test                        |         |          | x         |          |         |
| Bonferroni post-hoc                        |         | x        | x         |          |         |
| Wilcoxon matched pair test                 |         |          |           | x        | x       |
| Spearman's rank correlation coefficient    |         |          | x         |          |         |
| Coefficient of variation (CV)              | x       |          | x         |          |         |
| Friedmans ANOVA                            |         |          |           | x        |         |
| Chi-squared Test                           |         |          |           |          | x       |

## **RESULTS AND DISCUSSION**

### **Isometric muscle action (Studies I and II)**

Maximal isometric plantar- and dorsiflexor torques were significantly lower in the children with JIA than in the controls in both Studies I and II. The strength measured with the HHD was approximately 50% lower than that found when using the computerized dynamometer for isometric dorsiflexion torque and four times lower for isometric plantarflexor torque. These lower values are probably a consequence of the uncertain stabilization of the patient when using the HHD.

The reduced isometric strength in the dorsiflexor in children with arthritis is documented in one study by Lindehammar and Backman (Lindehammar and Backman, 1995). They pointed out that a reduced muscle thickness could lead to lower muscle strength in those muscles closest to the inflamed joint. Their results from muscle testing were presented in force instead of torque, which makes it impossible to compare with other results.

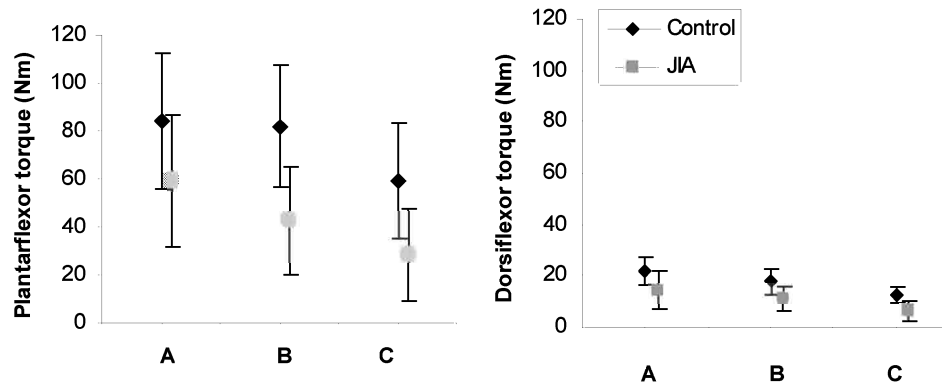
The Quadriceps/Hamstring (Q/H) ratio in Study I was less than 1 in the patient group and 1 in the control group, which implies that the quadriceps muscles are less capable of generating tension than the hamstrings muscles. These ratios point to an altered profile that may warrant greater clinical attention including focus on strengthening the quadriceps.

The Q/H torque ratio provides a means for identifying impairments of strength between major muscles acting at a joint. However, it has not been widely applied in children but in adults (Aagaard et al., 1998; Baltzopoulos and Brodie, 1989; Kannus, 1989). The strength difference between groups of muscles crossing opposite sides of the same joint has been of interest. Both injury prediction and rehabilitation programs have focused on strength ratios between muscle groups, with the Q/H

strength ratio being the most commonly investigated (Aagaard et al., 1998; Burnie and Brodie, 1986; Kannus, 1989; Seger and Thorstensson, 2000). Many studies investigating this relationship utilize the peak torque that each muscle group can produce rather than the torque produced at the same joint angle (Damiano et al., 2001). Such measurements are, however, problematic as peak isometric torque values may occur at different joint angles (different muscle length) for the two opposing muscle groups. The agonist-antagonist strength relationship may be better described by comparing the functional lengthening and shortening muscle strengths for each muscle group (Aagaard et al., 1998).

### **Shortening and lengthening muscle action (Study II)**

In shortening plantarflexors the JIA group created 52% lower torques than the controls. The maximal lengthening plantarflexor torques, however, did not differ between the groups (JIA 59 Nm vs control 84 Nm). Maximal muscle action in the dorsiflexors was higher in the controls than in the JIA group in both shortening and lengthening actions (52% and 37% respectively). Of the muscle action types in children with JIA lengthening torques were highest, followed by isometric, and finally shortening torques. Plantarflexor torques was observed to be four to five times greater than comparable dorsiflexor torques (Figure 6).



**Figure 6** Angle specific maximal voluntary torque (strength) measured at an ankle angle of 90° during lengthening (A), isometric (B) and shortening (C) plantar and dorsiflexion actions in children with JIA (n=10, grey) and healthy age and gender matched controls (n=10, black)

The significant reduction of shortening torque is velocity dependent (Damiano et al., 2001; Fenn, 1938) and the lower shortening torques for both the children with JIA and healthy controls are comparable to the torques reduction seen in healthy adults at the same velocity (15°/s) (Pinniger et al., 2000). Interestingly, there was a much larger reduction in dorsiflexion shortening torque than in isometric torque in both groups (approximately 60-70%).

Maximal voluntary lengthening actions produce significantly greater torque than shortening actions at the same velocity. This is shown in several studies, but it has been debatable whether maximal lengthening actions produce greater torques than maximal isometric actions under voluntary activation. Our results showed that children with JIA could produce significantly higher lengthening torques than isometric torques in both plantar- and dorsiflexion, while the healthy control children could not. This might indicate an elimination of involuntary inhibition during lengthening actions in children with JIA. Caution must be taken, however, when interpreting this result as it may be that the maximal voluntary isometric

torques produced by the children with JIA were influenced by inhibitory factors and were lower than that which the muscle was truly capable of producing. Such inhibition is thought to occur during lengthening actions in healthy adults and children and can only be accurately assessed by involuntarily activating the muscle with electrical stimulation superimposed on a maximal voluntary effort (Belanger and McComas, 1981). Any additional torque produced by the electrical stimulation indicates an incomplete activation of the muscle (Herbert and Gandevia, 1999). Due to ethical reasons, this technique was not utilised in this study and the maximal voluntary torque values attained may be somewhat underestimated despite considerable verbal encouragement given to the children to produce a maximal effort.

While all dorsiflexor strength measures appeared to be somewhat lower for the five children with JIA with a reduced range of dorsiflexion motion compared to those with the full 30° ROM, no significant differences in strength was found between these two groups.

A method to show differences in shortening (concentric-C) and lengthening (eccentric-E) muscle strength has been to utilize the E/C ratio (Damiano et al., 2001). An issue with this method is that peak torque values may occur at different joint angles (different muscle length) for the two opposing muscle groups. Our results showed a significant difference in E/C ratios in both plantar- and dorsiflexors between children with JIA and controls (Table VI). Our findings in the ratios between plantarflexors and dorsiflexors in Study II perhaps indicates that arthritic disease can affect muscle strength likewise on both sides of the joint.

**Table VI** Comparison between children with JIA and controls eccentric (lengthening) to concentric (shortening) (E/C) ratios presented as mean values ( $\pm$ SD) and ANOVA results.

| E/C Ratios          | JIA           | Conrols | p    |
|---------------------|---------------|---------|------|
| Ankle plantarflexor | 2.1 $\pm$ 1.4 | 0.3     | 0.01 |
| Ankle dorsiflexor   | 2.3 $\pm$ 1.7 | 0.2     | 0.04 |

It is important to include developmental changes in strength due to neurological maturation, muscle tissue changes, body proportion changes, sex and age and the day-to-day variation. In Study II the participants were only girls, in order to avoid gender difference. Sex differences have been reported during the childhood years with the mean strength measures for boys being consistently higher than for girls (Sunnegardh et al., 1988). The boys in puberty increase their strength due to increased androgen production (Round et al., 1999). Seger and Thorstensson (1994; 2000) found in comparison between genders no significant differences in absolute torque values between the boys and girls of age 11, either for lengthening or shortening muscle actions in the knee. Follow-up values after 5 years were significantly higher in boys than in girls in both lengthening and shortening knee extension actions (Seger and Thorstensson, 2000). In a recent longitudinal study by De Ste Croix et al (2002) no evidence of sex differences in shortening knee extension and flexion were shown.

### **Velocity and distance parameters (Studies III-IV)**

In Study III the children with JIA had slower walking velocity ( $1.06 \text{ m} \cdot \text{s}^{-1}$ ) than the control group ( $1.28 \text{ m} \cdot \text{s}^{-1}$ ). These findings have been reported in an earlier study by Lechner et al. (Lechner et al., 1987). The difference between the two groups was reduced to being a tendency ( $p=0.09$ ) when normalization to height was performed. Similar changes have been found by Frigo et al. (Frigo et al., 1996) in an earlier study.

Walking velocity is known to vary with age and, more importantly, with factors such as height and leg length (Oberg et al., 1993; Ounpuu et al., 1991; Wheelwright et al., 1993b). There is a recognized problem that children with JIA generally have a disturbance in growth (Cassidy and Petty, 2002), and it is therefore important to take this into consideration when comparing the walking velocity of different children. The introduction of new treatments such as ICI and TNF-inhibitor has shown to decrease the growth disturbances (Hagelberg et al., 2001; Sherry et al., 1999). We recommend normalization to the individual's height when comparing walking velocity between groups, both before and after treatment.

The participants in Study IV increased their walking velocity after treatment with ICI, but unexpectedly there was no correlation between pain rating and walking velocity. Such a correlation was, however, observed in Study III.

No differences in cadence, single- and double support were determined between children with JIA and the control group. In Study III, however the children with unilateral involvement showed a shorter duration of single support loading on the affected leg.

Pirpiris et al (2003) found that walking speed was significantly higher while walking in a 10-m gait laboratory with markers and electrodes than during walking for 10 minutes freely. In an unpublished study prior to this thesis work, children with arthritis reported impaired physical ability because of the trouble caused by the inflamed joints. They felt, for example, that they were walking more slowly than their peers. This appeared, for example, in statements such as "I'm not as quick as the others are", and "I'm less strong and I tire more easily than my friends". Some also mentioned problems with their physical education teachers who did not fully understand the complaints since they compared the children with JIA with the others in the class. More than half took part in school sports while

some did not - to varying degrees - in skating, long walks running, football and certain types of apparatus gymnastics and athletics. About half of children took part in some form of physical activity outside school hours, e.g. bandy, football, dancing, workout, riding a horse and swimming. Or, as one of the youths expressed it: "I keep fit by living very much like the healthy people of my age".

## **Ground reaction force**

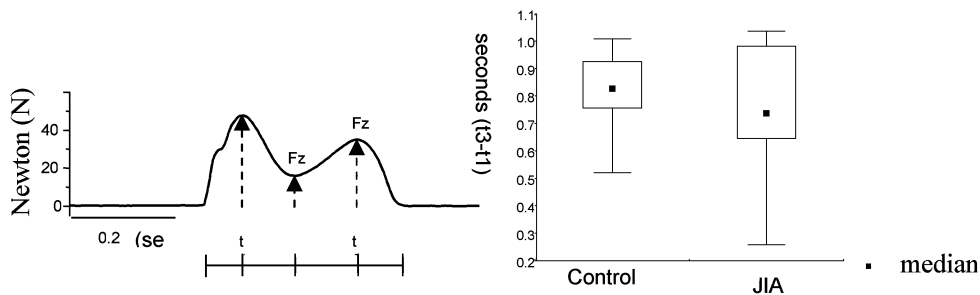
### ***JIA vs controls (Study III)***

A significant decrease in the peak vertical GRF during heel contact (Fz1, and push-off (Fz3), were observed in the children with JIA in Study III compared with controls. At heel contact in normal gait action of the ankle dorsiflexors is associated with a few degrees plantarflexion in the "first ankle rocker" (Perry, 2002). The children with JIA showed around 40% lower lengthening torque in the dorsiflexors in Study II than the control group. This muscle weakness affects the capacity to resist a rapid plantarflexion which results in flat foot landing (Perry, 2002). During toe-off the ankle plantarflexors act while shortening, which requires adequate ankle plantarflexion ROM (Perry, 2002). This action, "third rocker", is the most likely to be affected by impairments typically associated with arthritis (Dhanendran et al., 1980;Platto et al., 1991;Rodgers, 1988). The results from Study II support that the shortening torque action of the plantarflexors are weaker than in the control group. No significant differences in vertical GRF in midstance (Fz2) were found. A lengthening action in the ankle plantarflexors (triceps surae) is performed during the "second rocker", a phase during midstance of tibial advancement over the foot (Perry, 2002). During midstance the foot position is neutral, motion of the stance leg is minimal, and the stance leg position is stable, with very little demands on muscle strength (Perry, 2002;Winter, 1990;Ounpuu et al., 1991).Our results from Study II indicated that there was no significant difference in lengthening plantarflexor muscle strength between children with JIA

and controls. No other study to date investigates the agreement between muscle strength measurement and ground reaction forces.

It has been shown in several investigations that the velocity influences both the shape and magnitude of the ground reaction forces (Wheelwright et al., 1993b; Ounpuu et al., 1991; Frigo et al., 1996; Stansfield et al., 2001; Cupp et al., 1999). Larger vertical and anterior-posterior ground reaction forces demand greater lower limb muscle strength (Perry, 2002). It is therefore possible that, due to pain, the slower walking velocities of the children with JIA resulted in the reduced vertical ground reaction forces at heel strike and push off (Fz1 and Fz3, respectively). This supports the subjective clinical observation of children with JIA having a less pronounced heel-strike and toe-off than healthy children. Children with JIA are also reported to have increased pronation and have frequent deviations of the foot that include pes planusvalgus, pes cavus and hallux valgus (Truckenbrodt et al., 1994). These deviations were not assessed in our studies, so it is uncertain to which degree, if any, these factors would affect the ground reaction forces. We were, however, unable to measure any significant differences in ground reaction force shape or amplitude between children with JIA and controls in these two directions.

In the time from peak loading (t1) to toe-off (t3), no significant differences were seen between the children with JIA compared to the controls. However, a tendency even if not significant showed that the children with arthritis had a shorter time, and varied more than the controls (Figure 7). Shorter support time would confirm our clinical experience that the children try to avoid weight bearing in their affected limb.



**Figure 7** Representative force plate trace (A) from one child in the control group, times from heel strike (t1) to toe off (t3). Figure B shows the loading time for the controls and children with JIA.

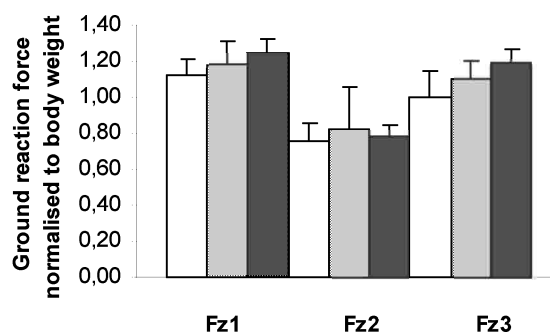
### Comparison between the Child Health Assessment Questionnaire (CHAQ) and stride parameters (Study IV)

The children's mean score on the CHAQ was 1.0 (range 0 to 2.3) before ICI treatment. There was a low negative correlation ( $r=-0.38$ ) between the mean score on CHAQ and the walking velocity. Negative correlation between step- and stride length and CHAQ was found ( $r=-0.70$  vs  $-0.80$ ), indicating that the children with the highest mean score on the CHAQ had the shortest step and stride length. The CHAQ is the most widely used measure of function in childhood arthritis (Bowyer et al., 1998; Dempster et al., 2001; Huber et al., 2001; Arguedas et al., 1997; Andersson et al., 1993). This finding indicates that the children with arthritis disease have a negative influence on functions such as velocity and stride parameters.

## Effects of ICI on gait (Study IV)

### Ground reaction force

In Study IV no significant differences in vertical GRF after treatment with ICI were found though values of Fz1 and Fz3 approached those of Study III's control group (Figure 8).



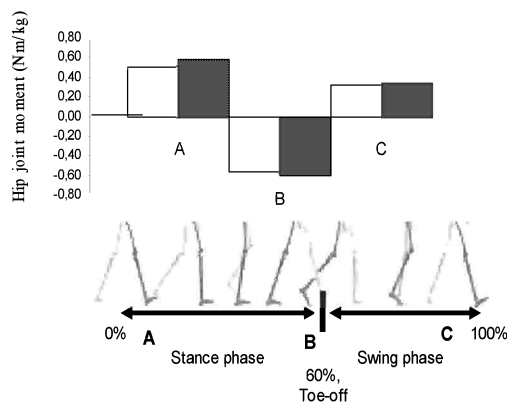
**Figure 8** Mean vertical ground reaction forces ( $\pm$ SD), normalised to body weight, for times corresponded to the initial maximal loading of the force plate (Fz1), mid-stance (Fz2) and terminal stance (Fz3) for the children with JIA pre treatment with ICI (open bars), after treatment with ICI (grey bars) and controls (filled bars).

### Gait kinematics and kinetics

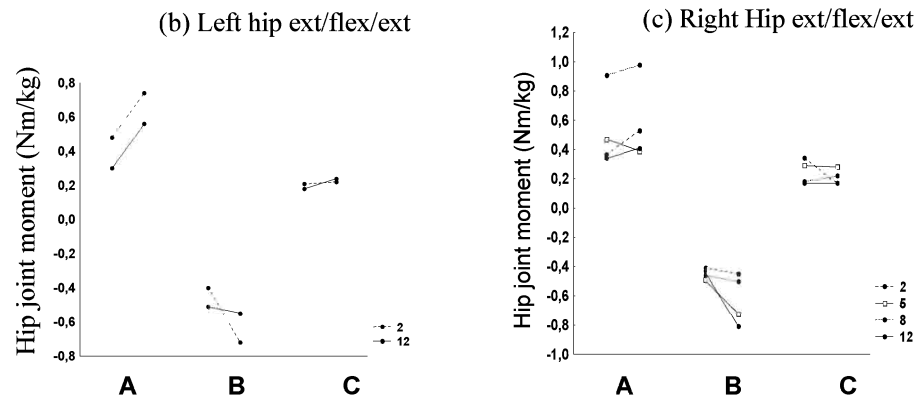
From gait analysis, increased range between maximum flexion and extension angles during walking were observed in the knee and ankle after ICI treatment. These findings indicate that the stiffness in the joints is not due to contraction but instead to active inflammation and/or pain. Care, however, should be taken when interpretation is done from such a small group. It is routine clinical practice to measure passive ROM to follow and evaluate the disease. During walking, dynamic ROM can be assessed, which is impossible without an electronic goniometer (Nicol, 1989) or gait analysis. Measurement of dynamic ROM is useful, particularly before surgical treatment, to determine, for example, whether the foot can be placed plantigrade and whether the contralateral leg can support the

operative leg during postoperative rehabilitation. Hopefully with new treatment options, surgical interventions such as athrodesis will remain uncommon in children though it is performed routinely in adult RA (Maenpaa et al., 2001).

The hip extension moment in the first 5-10% of the gait cycle increased significantly (Figure 9) after treatment with ICI, even if the subjects were not treated in the hip joint. Two children with JIA were treated bilaterally in the hip joint and two were treated only in the right hip (Figure 10). In the frontal/coronal plane there were no significant differences in abduction moment at mid-stance (~20% of the gait cycle) or in abduction moment at terminal swing (~50% of the gait cycle) after treatment with ICI.

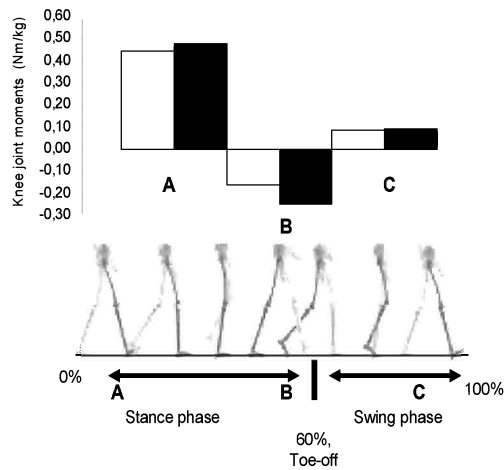


**Figure 9** Hip extension and flexion moments (Nm/kg) in sagittal plane before (open bars) and after (filled bars) treatment with ICI in children with JIA (n=18). A=extension moment at initial contact (loading response ~10% of gait cycle), B=flexion moment at preswing (~ 50% of gait cycle) and C=extension moment at terminal swing (~85% of the gait cycle).

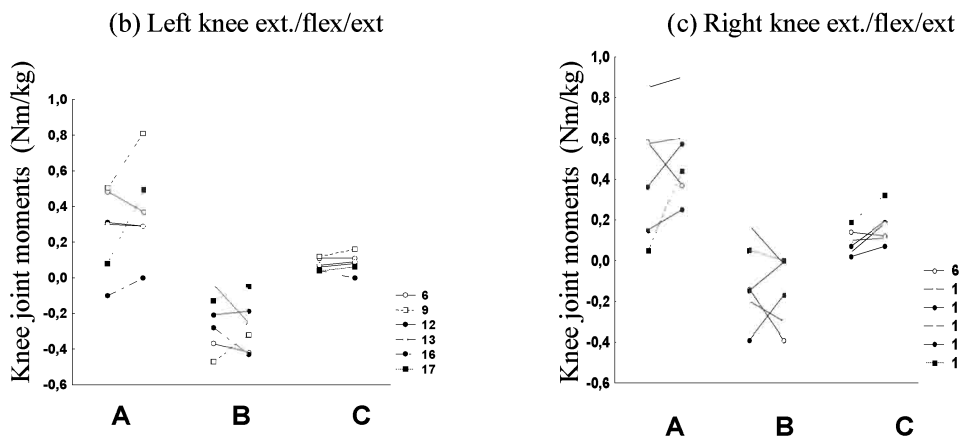


**Figure 10** Two children with JIA profiles before and after treatment with ICI in left hip (b), four children were treated in the right hip, A, B and C are the same moments as in Figure 9.

Seven subjects were treated with ICI in the knee joint, five bilaterally and two unilaterally. Knee sagittal range of motion (extension/flexion) increased with 4° after treatment with ICI. Knee flexion moment at preswing (~50% of the gait cycle) decreased after treatment, even if ICIs were not made in the knee joint (Figure 11). Three participants who were treated with ICI in the left knee joint showed increased knee flexion moment (negative value) and four of the participants who were treated with ICI in the right knee joint (negative) showed increased flexion moments at midstance (Figure 12). The knee extension moments at terminal swing (~85% of the gait cycle) showed increased values after treatment for all the children that were treated with ICI in the knee joint (Figure 12). The knee varus moment at initial contact and at preswing did not show any significant differences after treatment with ICI in the knee joint.

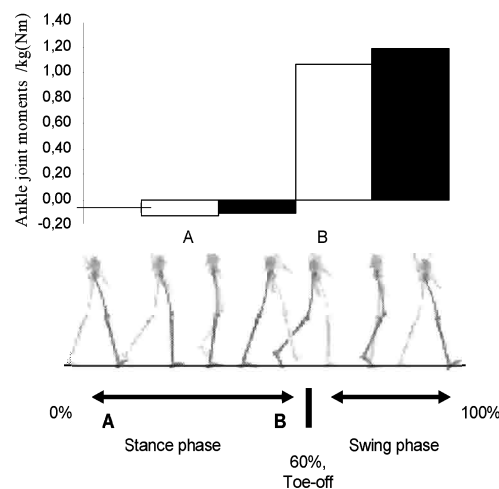


**Figure 11** Knee extension and flexion moments in sagittal plane before (open bars) and after (filled bars) treatment with ICI on children with JIA (n=18). Not all children with JIA were treated with ICI in the knee joint. A=knee extension moment at initial contact (loading response ~10% of gait cycle), B=knee flexion moment at preswing (~ 50% of gait cycle) and C=knee extension moment at terminal swing (~85% of the gait cycle).

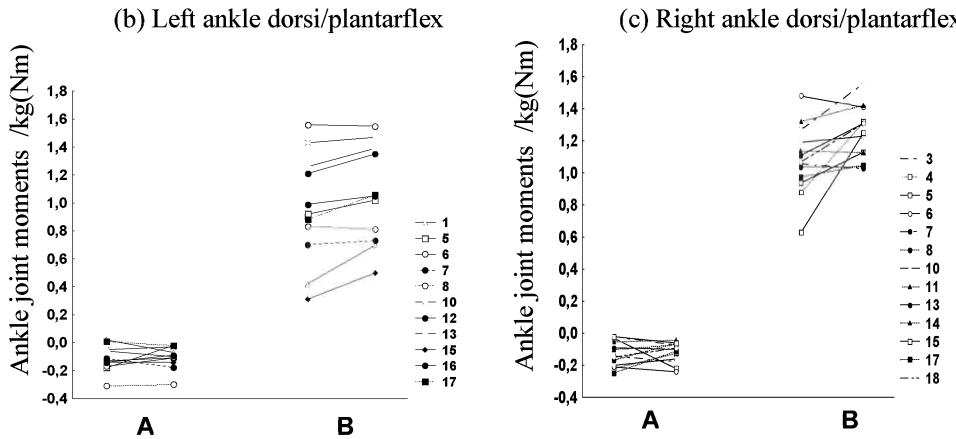


**Figure 12** Five children with JIA were treated bilaterally in the knee joint and two were treated unilaterally. Their profiles before and after treatment with ICI in the left (b) and right (c) knee are shown. A, B and C are the same moments as in Figure 11.

Thirteen children were treated in the right foot and 11 were treated in the left foot. The ankle joint sagittal range of motion (maximum dorsiflexion to maximum plantarflexion) increased significantly with 3° and the peak plantarflexion moment at preswing increased after treatment with ICI (Figure 13). Eight children were treated unilaterally and eight bilaterally in the foot joints. All children that had ICI in their foot showed increased plantarflexion moment at preswing (Figure 14).



**Figure 13** Ankle dorsi- and plantarflexion moments in sagittal plane before (open bars) and after (filled bars) treatment with ICI in children with JIA (n=18). Not all of the children with JIA were treated with ICI in the ankle joint. A=dorsiflexor moment at loading response (~10% of gait cycle) B=plantar flexor moment at preswing (~ 50% of gait cycle)



**Figure 14** Eleven children with JIA profiles before and after treatment with ICI in the left (b) and right (c) ankle. A and B are the same moments as in Figure 13.

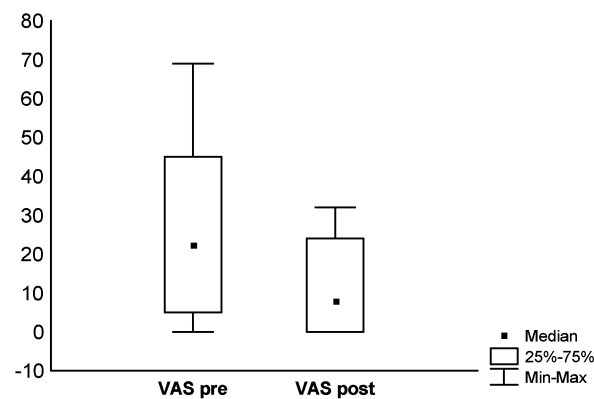
The main muscle group that is active at the ankle throughout stance is the plantarflexor group (triceps surae) (Perry, 2002). It was shown in Study IV that the plantarflexor moment increased after treatment with ICI, which is important in order for generation of power during toe-off. Improvement of plantarflexion moment is particularly important in children with JIA who were shown in Study I and II to be weaker in plantarflexion than healthy controls.

The ankle powers increased significantly at foot-off (~50% of the gait cycle) after treatment with ICI. This is not unexpected findings when fourteen of the children were injected in the ankle and subtalar joints.

The static joint angles in the hip, knee and ankle did not show any significant changes when measured with a goniometer after treatment with ICI, but during gait, increased dynamic ranges of motion were found.

### **Pain evaluation**

The participants in Study IV rated their pain significantly lower (26 mm before vs. 11 mm after) on the VAS after treatment with ICI (Figure 15).



**Figure 15** Pain rating after treatment with ICI

This thesis presents positive effects of treatment with ICI in the lower extremities - especially on pain, walking velocity and joint moments. Furthermore, and importantly, the data indicates that ICI improves the dynamic ROM and function even in joints not being treated.

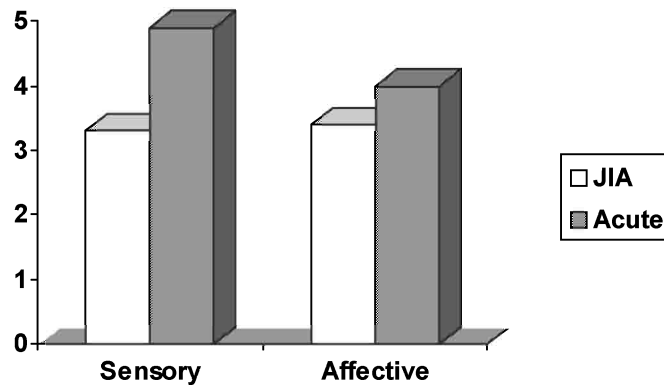
### **Pain (Studies II-V)**

There were no significant differences in Study II in the rating of pain on the VAS in children with JIA before (4mm) and after (14mm) the strength testing session. In Study III a significant negative correlation was found between the level of perceived pain (as measured by VAS) and walking velocity of children with JIA indicating that children with pain walked more slowly.

In Study V the Pain-O-Meter was evaluated to determine whether children and adolescents knew the words and if so, whether the words described their pain. Seventeen of 23 words were known by at least 70% of the children (n=61). An age-

related trend was observed in the number of words not known; children (age 6-11 y, group I) knew fewer words on the POM than adolescents (12-16 y, group II). In the adolescent group (group II) six of the sensory and affective words were known by all of the children. This was not the case in the small children (group I) in which no word was known by all. The affective words *suffocating* and *killing* were known more frequently by the JIA group, who also used *tiring* somewhat more frequently, though not significant. The children with acute pain somewhat more frequently, though not significantly, described their quality of pain as *aching* and *terrible*.

The mean number of sensory and affective words selected was somewhat higher in the acute group than in the JIA group, though not statistically significant (Figure 16). Factors influencing the number of words chosen could be pain intensity (Wilkie et al., 1990), developmental factor or children's vocabulary skills (Watkins and DeThorne, 2000). Some earlier studies show that the number of words increased with increased pain (Savendra et al., 1993; Wilkie et al., 1990). Results from this study show that pain intensity was equal in both groups (Md VAS scores 5.4 cm). The developmental level and vocabulary skills were not evaluated in this study. One explanation for the difference between groups could be that those children with JIA were more acquainted to having pain whereas it was an unfamiliar experience for children experiencing acute pain.



**Figure 16** The mean number of sensory and affective words selected in respective diagnosis group.

While advances have been made in developing pain assessment tools for children, there is currently no Swedish version of a multidimensional tool. One attempt was done to translate the European APPT tool (Abu-Saad et al., 1990). Care was taken when translating the English words to Swedish since direct translations often led to inappropriate and uncommon Swedish words. We therefore decided to evaluate the utility of the affective and sensory word descriptors on the Swedish version of Pain-O-Meter (POM).

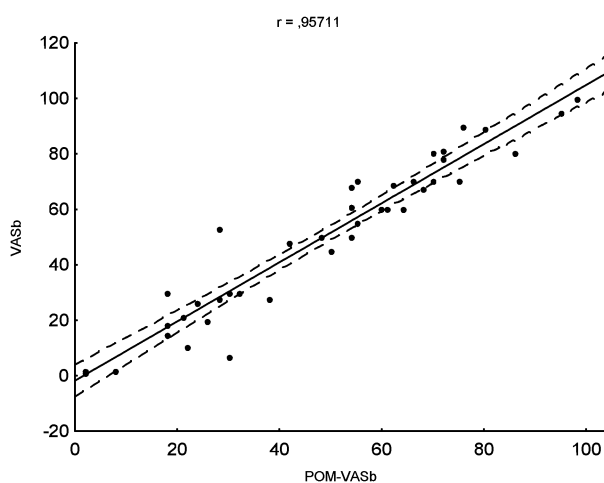
The use of pain descriptors is a possible approach to develop our ability to assess pain as well as our understanding of pain in children. The children's cognitive-developmental stage and conceptualisation of pain and other pain-related words are valuable to consider when assessing pain. Wilkie et al (1990) developed a list of sensory, affective and evaluative words for measuring pain quality in children. The criterion for a word to be included was that it was known by 50% of the subjects. It is important to discuss which words should be included in a pain list when assessing pain. The word *grinding* (Swedish *molande*) was surprisingly unknown for most of the participants. In clinical situations, *grinding* is a word commonly used in the adult population when communicating about pain. Questions can arise,

such as whether the adults fully know the meaning of the word grinding and whether this word is too difficult to define.

The words selected and the number of words selected may have been affected by several factors in this study. Vocabulary knowledge is highly experience-dependent (Watkins and DeThorne, 2000). In a study by Toole et al. (2000) children with a high level of literal understanding chose fewer words to describe pain than did children with a lower level of verbal comprehension, which used more words when they experienced more pain.

#### ***Horizontal visual analogue scale (VAS) compared to vertical visual analogue scale on the POM (POM-VAS)***

In 40 cases POM-VAS was compared to VAS (Figure 17). There were no statistical differences in pain intensity score when children were assigning their pain using the POM-VAS (median 5.4 cm) or VAS (median 5.4 cm). Our results have corresponded to earlier studies between horizontal and vertical VAS (McGrath et al., 1996)



**Figure 17** Comparison between pain assessing on POM-VAS and VAS (n=40)

## **General discussion**

### ***Muscle strength measurement and gait analysis (Studies I, II, III & IV)***

It is important to measure plantarflexor strength because of their involvement in almost all movements of the lower extremities. The dorsiflexors major role in gait occurs just after initial contact when the foot is lowered to the floor and during the swing phase. This is controlled by eccentric contraction of the dorsiflexors and the range of motion during this phase of gait is very small (Perry, 2002).

The plantar flexors act eccentrically under the single limb support period (Perry, 2002). During push-off, the plantar flexors shorten to actively plantarflex the foot and to generate an explosive push-off force (Perry, 2002; Winter, 1990). It follows that heel-contact and push-off is most likely to be affected by the impairments typically associated with arthritis in childhood and in adults (Truckenbrodt et al., 1994; Spraul and Koenning, 1994; Mann and Horton, 1996; O'Connell et al., 1998)

Lower limb strength is vital for proper locomotion. It has been shown that the kinematics of the hip, knee and ankle joints are strongly related to walking velocity (Nilsson et al., 1985) and that both the shape and magnitude of the external ground reaction forces is also velocity dependent (Nilsson and Thorstensson, 1989). In addition to joint pain, this reduction in velocity may partially be due to the functional weakness of the plantar- and dorsiflexors.

The potential clinical implications of the results in this thesis are important. The weakness in almost all action types in plantar- and dorsiflexors in children with JIA indicates the importance of exercising the plantar flexors and the need to evaluate muscle strength continuously. The weakened muscle strength also affects walking patterns as there is a risk for a reduced velocity. In an unpublished study children with arthritis were found to be more tired and easily irritated, to experience pain

and stiffness and to suffer from the fact that they could not be as active physically as their peers. It is vital to encourage children with JIA to participate in physical activities as much as possible, since there otherwise is a risk that they end up in a vicious circle with deteriorating fitness. Children and adolescents with rheumatic diseases can improve their aerobic endurance through participation in weight-bearing physical conditioning programs without increased disease activity or increased pain (Klepper, 1999; Klepper et al., 1992). Two studies (Takken et al., 2001; Takken et al., 2002) have shown that children with JIA have a moderate to large impairment in their cardio-respiratory fitness compared to healthy children. This knowledge, together with the results from Studies I-IV, show that it is important to create exercise opportunities for children with JIA so they can participate in physical activities and sports. The results from this and other studies shows that further studies are needed to investigate the differential effects of specific exercises on specific muscle activations.

## ***Aspect of methods***

### ***Dynamometry***

There are restrictions when using the HHD because it can be difficult to stabilize the patient's joint position. Furthermore, it is important to have control of the examiner's strength in order to record accurate data. One way to avoid the above problems is to use a computerized dynamometer as in Study II, which also enables control over joint angles and muscle lengths. The HHD can only measure isometric muscle strength while the computerized dynamometer can measure different action types at different angular velocities.

To compare muscle strength between subjects and evaluate treatment the distance between the dynamometer and the joint axis of rotation must be measured. Strength comparison must be made in torque (newtonmeter) and not in force (Newton). Subjects in study I were tested with the HHD while lying on a mat in order to

neutralize gravity. In other studies subjects were tested sitting (Lindehammar and Backman, 1995; Lindehammar and Sandstedt, 1998). Reproducibility of strength measurements is important and therefore we tried to avoid movement of the pelvic and thigh when using the isokinetic dynamometer. When testing children, it is important that the isokinetic dynamometer is safe and stops when the child finishes the movement. Most of the children reported discomfort at the end of the test session; it was uncomfortable to lie on a bench in supine position. Some of the children with JIA also reported a feeling of stiffness in the neck and spine at the end of the test session. This is also reported in a earlier study by Giannini et al (1993). Further discussions and investigations about the best test position for dynamometry studies are necessary in order to avoid the measurement of incorrect values.

### ***Gait analysis***

Several methodological problems arise in movement and gait analysis. Gait varies slightly between walks and subjects. Several trials are therefore needed to get consistent results, which can be a problem when dealing with patients who have poor physical conditions (Gage, 1993; Sutherland, 1990; Winter and Wells, 1981). The subjects also have to strike the force plates with only one foot, which can be difficult for children, elderly, or subjects with pathology, in whom stride length may not be ideal for the force plate configuration (Gage, 1993; Kadaba et al., 1989; Oberg et al., 1993; Sutherland, 1990; Winter and Wells, 1981; Winter and Eng, 1995)

When dealing with kinematic data there are always concerns about marker misplacement (Davis, 1997; Ounpuu et al., 1991; Alexander and Andriacchi, 2001). Marker misplacement leads to changes in the kinematics data. To minimize displacement in Study IV the same (EB) trained physical therapist placed the markers on the subject.

### ***Pain evaluation***

Measuring pain is a challenge since pain is a subjective experience and may be complicated due to the age of the subjects. There is no evidence that children are more or less truthful or accurate in expressing their pain than adults. Two important demands on a pain instrument are 1) does it consistently measures the same results, particularly when applied to the same subjects at different times (reliable) and 2) Does it actually measure the underlying aspect or not (validity). Validation of pain measures is difficult and there are no available self-reports of pain that fulfill all the criteria for an ideal validity (for example construct validity, content validity, convergent validity, divergent validity and predictive validity). The two pain instruments used in this thesis were VAS and the POM. Both instruments have shown good validity and reliability (Gaston-Johansson, 1996; McGrath et al., 1996; Abu-Saad, 1984). The measure must be free from response bias, meaning that children give their answers regardless of how they wish to please the administrator, physician or parents. Many children need help to differentiate between pain and stiffness, and between muscle and joint pain.



## **CONCLUDING REMARKS**

The main findings of this thesis establish that juvenile idiopathic arthritis influences muscle function at the ankle joint, by reducing strength by, 40-50 % lower muscle strength. These muscles are involved in almost all movements in the lower extremities and are therefore important to strength train.

Clinically, we can see that children with juvenile idiopathic arthritis walk more slowly decrease their heel strike and push off, and sometimes have flexed hips and knees. The treatment today is more aggressive with intra-articular corticosteroid injections in the joints and treatment with TNF-inhibitors, and as a result less joint destruction is seen in children with arthritis.

Gait analysis is a proper and clinical relevant tool to analyze gait, especially joint moments that are impossible to study otherwise. Some of the joint moments showed increased values after treatment with intra-articular corticosteroid injections.

Future research may focus on assessing the effects of strength training, aerobic and water exercise, and treatment with TNF-inhibitors. The established and tested methods in this thesis (isokinetic dynamometers and 3D-gait analysis) may be used to evaluate the outcomes. Furthermore, pain assessment using sensory and affective word descriptors and functional outcomes can be an important tool in evaluating the participants' physical and psychological condition

## SVENSK SAMMANFATTNING

Barnreumatiker (juvenil idiopatisk artrit JIA) får ofta ledstelhet (kontrakturer) och skelettdeformiteter samt kan utveckla muskelsvaghet såväl som muskelförtvining (atrofi) om inflammationen blir långvarig. Barnen blir stela och får ont i lederna, vilket leder till rörelsehinder och minskad fysisk aktivitet. Det orsakar i sin tur muskelsvaghet och ökad stelhet – en ond cirkel bildas.

Det övergripande syftet med denna avhandling är att med hjälp av olika mätmetoder utvärdera behandling med ledinjektioner med kortison (intraartikulära kortikosteroidinjektioner - ICI) hos barn med JIA. Mer specifikt har avsikten varit att:

A. Kartlägga isometrisk excentrisk och koncentrisk muskelstyrka hos både friska barn och barn med JIA.

B. Objektivt bedöma rörelseförmågan före och efter ICI med hjälp av tredimensionell gånganalys.

C. Utveckla en metod för att bedöma hur den långvariga smärtan påverkas.

För att mäta muskelstyrka användes en handhållen dynamometer (HHD) samt en datoriserad dynamometer, medan golvreaktionskrafter (kinetik) mättes med hjälp av två kraftplattor (Kistler, Switzerland). Ett tredimensionellt rörelseanalyssystem, (Vicon, Motion system, Oxford, England) användes för att kvantifiera rörelser, dvs mäta ledvinklar, ledkraft, effekt, hastighet samt acceleration under gång. Försökspersonerna har beskrivit sin smärta med ord med hjälp av smärtskattningsinstrumentet Pain-O-Meter (POM).

Vid mätning av muskelstyrka (maximal viljemässig kontraktion – MVC) med den handhållna dynamometern påvisades minskad styrka hos en grupp barn med JIA

jämfört med en kontrollgrupp. Det visade sig att det var god upprepbarhet i användandet av HHD, men det är samtidigt viktigt att tillägga att stabilisering av testpersonen är svår. I den studie där den datoriserade dynamometern använts för att registrera muskelstyrkan i sträck- och böjmusklerna (plantar- och dorsalflexorer) i fotleden visade resultatet att barn med JIA är svagare isometriskt (muskeln arbetar statiskt) i såväl plantar- som dorsalflexorer jämfört med jämnåriga barn i kontrollgruppen. Då den excentriska MVC (muskeln arbetar under förlängning) av dorsalflexorerna mättes påvisades att barn med JIA är ca 40-50 % svagare jämfört med kontrollgruppen. Detta gäller även vid koncentrisk MVC (muskeln arbetar under förkortning) av både plantarflexorer och dorsalflexorer. Inga skillnader kunde konstateras i excentrisk MVC av plantarflexorerna.

Vi har kunnat påvisa att barn med JIA har en lägre gånghastighet samt en tendens till något längre dubbel stödfas under gång. Kraftdata registrerades med hjälp av en kraftplatta som visade att barn med JIA hade en försiktigare hälisättning och ett svagare frånskjut än den jämnåriga kontrollgruppen.

Vid utvärdering av behandling med ICI med hjälp av tredimensionell gånganalys observerades ett ökat rörelseomfång i knä och fotled under gång. Genom gånganalysen kunde man påvisa ett ökat vridmoment i höftleden då hälen sätts i. Vid frånskjut konstaterades en ökning av vridmoment i knä- och fotled efter behandling med ICI. Barnen skattade även sin smärta lägre samt ökade sin gånghastighet efter behandlingen.

I Sverige får barn vanligtvis skatta sin smärta med hjälp av en visuell analog skala (VAS). Mer kvalitativ information ger smärtskattningsinstrument som använder sensoriska och emotionella ord. Ett sådant är Pain-O-Metern. Ordförståelsen av de sensoriska och emotionella orden på POM testades på barn mellan 6 och 16 år med akut och kronisk smärta. Resultatet visar en ökad förståelse med stigande ålder. De

ord som barnen använde mest frekvent för att beskriva sin smärta var värkande, ömmande, besvärlig och tröttande. Inga skillnader kunde påvisas vad gäller smärtans intensitet mellan barn med akut smärta eller med kronisk smärta.

Det finns ingen tidigare dokumentation av behandlingseffekter på gång före och efter behandling med ICI. Denna avhandling har visat på goda effekter av ICI för såväl behandlade som icke behandlade leder. I utvärderingen av denna och andra behandlingar är objektiva mätmetoder av största vikt. Tredimensionell gånganalys, isokinetisk dynamometer samt Pain-O-Metern har samtliga visat sig vara användbara i detta sammanhang.

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