Chapter 1

Aim and introduction to the thesis
Introduction

Juvenile idiopathic arthritis: a chronic disease

Juvenile idiopathic arthritis (JIA) is a heterogeneous group of auto immune diseases that includes all forms of arthritis beginning before the age of 16 years, persisting for more than 6 weeks and of unknown origin\(^1\)\(^-\)\(^4\). The main characteristic of JIA is arthritis due to inflammatory synovitis causing morning joint stiffness, swelling, decreased motion, pain, decreased muscle strength and muscle atrophy. The aim of current therapy is to induce and sustain remission of disease through the use of medication so that normal childhood activities, growth and development can be sustained\(^5\),\(^6\). Through medication the synovial inflammation caused by the derailed immune system is modified and eventually controlled leading to remission of the disease. Unfortunately no medication exists that achieve rapidly, consistently and long lasting remission of disease and therefore JIA patients can have intermittent periods of active and inactive disease\(^6\),\(^7\). Therefore JIA is a chronic disease affecting the child for extended periods of time and often for life. Long-term follow up studies over the last twelve years show that between 37 and 54% of adult JIA patients have active disease and between 2.5 and 37% have severe functional impairments\(^1\),\(^8\)-\(^16\). In the Netherlands it is estimated that between 2000 to 3000 children under the age of 16 years suffer from JIA.

Chronic disease in perspective

Children with a chronic disease are confronted with illness specific ailments. Furthermore, their chronic disease will induce changes in organ or body elements not primarily affected by the disease. Finally, a chronic disease will affect, through the physical domain, the psychological and social domain. Therefore, children with a chronic disease and their families have to deal with the impact of disease in many important life domains. Health is than not the mere absence or control of disease but a human condition with physical, social and psychological dimensions\(^17\),\(^18\). This view on health has created a higher awareness of the importance of function irrespective of the presence or absence of active disease. Assessment of function has become essential to reduce functional limitations and improve well-being\(^19\). For this reason the World Health Organization has provided a unifying framework for classifying the consequences of (chronic) disease by the “International Classification of Functioning, Disability and Health”, abbreviated as ICF\(^20\). The aim of the ICF is to provide a unified and standard framework and language for the description of health status and it is
based on a model of interactions among dimensions of human functioning at body, personal and societal levels whereby the role of the environment is clearly acknowledged\textsuperscript{19, 20}. The ICF conceptual framework is depicted in figure 1.

**Figure 1. “International Classification of Functioning, Disability and Health” conceptual framework**

![Diagram](image)

In this framework function can be qualified in terms of capacity and performance\textsuperscript{19, 20}. Capacity is the child’s ability to execute a task or action within a standardized environment while performance is defined as what an individual child does in his or her current environment\textsuperscript{19, 20}.

**Physical activity, physical fitness and health in chronic disease**

A chronic disease limits the child, direct and indirect in its (physical) activities leading to hypoactivity\textsuperscript{21}. Overprotection and ignorance of parents, teachers and even health professionals, fear towards activity, medication and social isolation are all possible contributing factors. Through a cascade of hypoactivity, deconditioning, detraining and increased and prolonged inactivity a vicious circle is unleashed causing disability (figure 2).
Over the last decade physical activity (PA) and physical fitness (PF) are assessed in a number of chronic conditions and they are summarised in table 1. In these studies PF is expressed as aerobic (exercise) capacity, an important element of health related PF. These studies confirm the assumption of Bar-Or of reduced PA and PF in chronic disease.

PA, PF and health are interrelated although their relationship is highly complex\(^\text{17, 22-24}\). To become and remain physically fit it is necessary to become physically active and to adopt a physically active lifestyle. PA can be described as all leisure and non-leisure body movements resulting in an increased energy output from the resting condition\(^\text{25}\). PF is a physiologic state of well-being that allows one to meet the demands of daily living and that provides the basis for sport performance\(^\text{26}\). Evidence for the health benefits of PA and PF is accumulating although conclusions are mostly based on prospective observational studies in adults\(^\text{27}\). PA and PF are effective in the primary and secondary prevention of several chronic conditions\(^\text{26, 28, 29}\). PA and PF are also associated with a reduction in all-cause mortality\(^\text{26, 29, 30}\). There is evidence that in youth aged 6 to 18 years, PA controls body weight, reduces blood pressure in hypertensive youth, improves aerobic capacity, muscular strength, endurance and skeletal health, reduces anxiety and depression and improves self-concept\(^\text{31}\). PA has also a positive
effect on academic performance, concentration, memory and classroom behaviour\textsuperscript{31}. For children with JIA and other chronic diseases it is recognised that they could take advantage of the same health benefits\textsuperscript{32, 33}.

### Table 1: Physical activity and aerobic capacity in children with chronic disorders compared to healthy controls\textsuperscript{*}

<table>
<thead>
<tr>
<th>Study</th>
<th>Chronic disease or disorder</th>
<th>Number of patients</th>
<th>Aerobic capacity</th>
<th>Physical activity</th>
</tr>
</thead>
<tbody>
<tr>
<td>Smith 2008\textsuperscript{34}</td>
<td>Pre-term born children (&lt; 32 weken, &lt; 1000 gr)</td>
<td>126</td>
<td>↓</td>
<td>n.a.</td>
</tr>
<tr>
<td>Painter 2007\textsuperscript{35}</td>
<td>Dialysis and kidney transplant patients</td>
<td>40</td>
<td>↓</td>
<td>↓</td>
</tr>
<tr>
<td>Shaibi 2008\textsuperscript{36}</td>
<td>DM Type 2</td>
<td>13</td>
<td>↓</td>
<td>↓</td>
</tr>
<tr>
<td>Valerio 2007\textsuperscript{37}</td>
<td>DM Type 1</td>
<td>138</td>
<td>n.a.</td>
<td>↓</td>
</tr>
<tr>
<td>Komatsu 2005\textsuperscript{38}</td>
<td>DM Type 1</td>
<td>72</td>
<td>↓</td>
<td>n.a.</td>
</tr>
<tr>
<td>Williams 2008\textsuperscript{39}</td>
<td>Asthma</td>
<td>n.r.</td>
<td>n.a.</td>
<td>↓</td>
</tr>
<tr>
<td>Henderson 1995\textsuperscript{40}</td>
<td>JIA</td>
<td>23</td>
<td>n.a.</td>
<td>↓</td>
</tr>
<tr>
<td>Takken 2002\textsuperscript{41}</td>
<td>JIA</td>
<td>144</td>
<td>↓</td>
<td>n.a.</td>
</tr>
<tr>
<td>Takken 2003\textsuperscript{42}</td>
<td>JDM</td>
<td>15</td>
<td>↓</td>
<td>n.a.</td>
</tr>
<tr>
<td>Nixon 2001\textsuperscript{43}</td>
<td>CF with normal lung function</td>
<td>30</td>
<td>↓</td>
<td>↓</td>
</tr>
<tr>
<td>Van Brussel 2005\textsuperscript{44}</td>
<td>ALL follow-up</td>
<td>102</td>
<td>↓</td>
<td>n.a.</td>
</tr>
<tr>
<td>Van Brussel 2006\textsuperscript{45}</td>
<td>ALL follow-up</td>
<td>13</td>
<td>↓</td>
<td>n.a.</td>
</tr>
<tr>
<td>Engelbert 2005\textsuperscript{46}</td>
<td>Hypomobility syndrome</td>
<td>19</td>
<td>↓</td>
<td>↓</td>
</tr>
<tr>
<td>Long 2008\textsuperscript{47}</td>
<td>Chronic pain</td>
<td>20</td>
<td>n.a.</td>
<td>↓</td>
</tr>
<tr>
<td>Takken 2004\textsuperscript{48}</td>
<td>OI Type 1</td>
<td>17</td>
<td>↓</td>
<td>n.a.</td>
</tr>
</tbody>
</table>

* Abbreviations: DM = diabetes mellitus; JIA = juvenile idiopathic arthritis; IDM = juvenile dermatomyositis; CF = cystic fibrosis; ALL = acute lymphatic leukemia; OI = osteogenesis imperfecta; ↓ = decreased compared to healthy controls; n.a. = not assessed; n.r. = narrative review

### Aims and outlines of the thesis

The aim of this thesis is to measure PA and PF in children and adolescents with JIA and to determine the effects of a program to improve PA in children with JIA. The studies described in this thesis are focussed on the following topics. First a measurement tool is evaluated to
measure aerobic capacity. Secondly aerobic and anaerobic capacity, PA and illness perceptions are measured in children and adolescents with JIA. Finally an intervention is designed and tested within a randomised controlled trial to improve PA and exercise capacity in patients with JIA.

The thesis consists of the following chapters: In the first chapter the perspective of JIA as a chronic disease is described. Reduced PA and aerobic capacity is common in children with a chronic disease. In the second chapter we examine the validity of the 6-minute walking test as a simple and inexpensive measuring tool for aerobic capacity in children with JIA. In chapter 3 we describe the aerobic and anaerobic exercise capacity in children with JIA in comparison with healthy peers while in chapter 4 we do the same for adolescents with JIA. Chapter 5 describes PA in adolescents with JIA and these are compared with healthy age mates. In chapter 6 we describe the illness representations of adolescents with JIA and associations with functional status are explored. In chapter 7 we introduce a new interactive internet-based program for children with JIA and we examine the effectiveness of the intervention on PA and PF using a randomised controlled trial. In chapter 8 the general discussion can be found.

References

Introduction


