General introduction

GENERAL INTRODUCTION

The idea for this project emerged in 2012 while I was working for Reinaerde for 2.5 years. I was involved in measuring physical fitness in individuals with intellectual disability (ID), and together with my direct colleagues we were aiming to increase the physical fitness and activity level of the people Reinaerde provided care and support for in the project called 'Reinaerde Fit'. During this project, we experienced that available knowledge, both scientific and clinical, was limited to physical fitness in adults or adults aged 50 and over with an ID. Pediatric physical therapists approached me, with questions like 'Which tests can we use to measure children's physical fitness?' and 'What are "normal" physical fitness levels for these children?'.

In the context of 'Reinaerde Fit', we started a small project at one of Reinaerde's pediatric day program centers in 2011. The project was called 'Skippy Fit'. In this project we evaluated the physical fitness of the children by using tests we had been using in the adult population with ID, since little was known on child- and ID-specific physical fitness tests. These tests were comparable to the tests used in the 'Healthy Aging and ID' study [2]. This project made us realize that tests like the grip strength test and 30 second chair stand were not suitable for the children and adolescents. We concluded that a thorough study was needed, in order to find appropriate tests for this specific population, which would enable us to gather information on the physical fitness of children and adolescents with ID.

Children and adolescents with intellectual disabilities

Intellectual disability (ID) is a disability characterized by significant limitations in both intellectual functioning and in adaptive behavior, which covers every day social and practical skills. The disability originates before the age of 18 [3]. ID can be classified into mild, moderate, severe and profound ID (Table 1). In the Netherlands, 1% of the population has an ID [4], among which are roughly 36,000 children and adolescents [5]. This thesis focuses specifically on children and adolescents with moderate and severe ID, which includes approximately 19,000 children in the Netherlands [6, 7].

Children and adolescents with ID already start off with more health problems than typically developing (TD) peers [8], including epilepsy [9] respiratory problems [10], sensorimotor dysfunction [11]. They also experience more musculoskeletal problems than persons without ID, like cerebral palsy, congenital deviation of feet and hip, hypotonia, and scoliosis [9, 12, 13]. These health problems can affect the quality of life of these young individuals, have a negative effect on their independent daily living skills, and thereby limit their possibilities to fully develop and participate. It is therefore important to monitor the health of this vulnerable population, and to support them to be as healthy as possible.

The Dutch context

In the Netherlands, support for children and adolescents with ID is organized by healthcare organisations, specialized in care and support for individuals with disabilities. The children and adolescents studied in this research project receive care and support from Reinaerde, one of those a healthcare organisation. According to the Universal Declaration of Human Rights [14], all children and adolescents have the right to attend a school that fits their qualities and abilities. Dutch children and adolescents with ID can attend a regular school with extra support, attend a special school when intensive support is needed, or can be exempted from compulsory education. The latter only applies when the child or adolescent has severe psychiatric, intellectual, physical or multiple disabilities. For these children and adolescents, healthcare organisations offer programs matching the possibilities of the child or adolescent at day program centres, to work on their skills to increase their independence and opportunities to participate in daily activities like helping with chores and playing with peers. These forms of care and support are financed by the national government. In these centres, extra support is available from medical, paramedical and psychological staff.

Table 1. Classification of level of intellectual disability

	IQ	Equivalent mental age as adult	Functioning as adult
Mild ID	50-55 to 70	9-12 years	Can live independently with minimum levels of support.
Moderate ID	35-40 to 50-55	6-9 years	Independent living may be achieved with moderate levels of support, such as those available in group homes.
Severe ID	20-25 to 35-40	3-6 years	Requires daily assistance with self-care activities and safety supervision.
Profound ID	≤20-25	< 3 years	Requires 24-hour care.

Physical fitness

Physical fitness includes a set of attributes people have or achieve that relates to the ability to perform physical activity [15]. It can be divided into health-related and skill-related fitness. Health-related physical fitness is the focus of the current study and consists of the components body composition, muscular strength, muscular endurance, and cardiorespiratory fitness. For adults, flexibility is another health-related fitness component, however the relationship between flexibility and health has not been confirmed in children [16, 17]. Therefore, this component has not been addressed in this study.

Low physical fitness levels are a risk factor for cardiovascular diseases, diabetes mellitus and poor mental health [18-21]. Several longitudinal studies show that risk factors of cardiovascular diseases, like low physical fitness, transfer from childhood to adulthood [22, 23]. Therefore, good physical fitness is not only important for children's and adolescence's direct health, but also for their health when they become adults. Furthermore, physical fitness is important to perform activities of daily living, and low levels of strength and endurance will limit independence in adulthood [24, 25].

Low levels of physical fitness have been found in children and adolescents with ID. Previous studies indicate higher rates of overweight and obesity than TD peers [26-30], low scores on muscular strength and muscular endurance tests [31-34], and low cardiorespiratory fitness levels [26, 27, 31, 34-36]. The before mentioned studies have mainly been conducted in youth with mild to moderate ID, and many studies were performed in samples of solely participants with Down syndrome (DS).

Correlates of physical fitness

Physical fitness can be increased by various types of exercise and physical activity, including habitual physical activity. The low physical fitness levels in children and adolescents with ID are often explained by the lack of habitual physical activity, as children and adolescents with ID have low levels of physical activity compared to TD peers [37-40]. As we all know, the association between physical fitness and physical activity is well studied in the general population. TD children and adolescents who are physically active have higher levels of physical fitness than their less-active peers [41, 42]. Furthermore, longitudinal studies show that inactive TD children can increase their fitness by increasing their physical activity [43, 44]. In children and adolescents with ID this relationship is hardly studied [40]. The few studies performed on the relationship between physical fitness and physical activity were all in youth with DS and were inconclusive: two studies found associations between physical fitness and activity [32, 45], while a more recent study did not find any association [46].

Another factor that is expected to contribute to the low physical fitness is motor competence or motor development. Children and adolescents with ID have lower age-related motor competence than their TD peers [47], which is associated with the musculoskeletal problems that are highly prevalent in children and adolescents with ID [9, 12, 13]. In TD children and adolescents, motor competence is found to be a prerequisite to become physically fit and active during childhood, adolescence and into adulthood [48-52].

The relationship between physical fitness and motor competence was assessed in two studies in children and adolescents with mild to moderate ID. These studies did find an association between physical fitness and motor competence [53, 54]. However, little is known on this association in children with more severe levels of ID, and on what role physical activity plays in this relationship.

Conceptual framework

The International Classification of Functioning, Disability and Health framework, known more commonly as the ICF-framework [55], provides a framework to describe the relationship between physical fitness, motor development, physical activity, health and participation, both at an individual and a population level (Figure 1).

In this framework, physical fitness and motor development can be positioned under 'body functions & structure', and physical activity under 'activities'. According to the ICF, all compo-

nents have a reciprocal relationship to each other, and contribute to health and participation. All are also influenced by environmental and personal factors.

This framework suggests that in order to support and maximize health and participation in children and adolescents with ID, physical fitness, physical activity and motor development should be optimized. To evaluate the start situation and monitor changes over time, we need feasible, reliable and valid measurement methods.

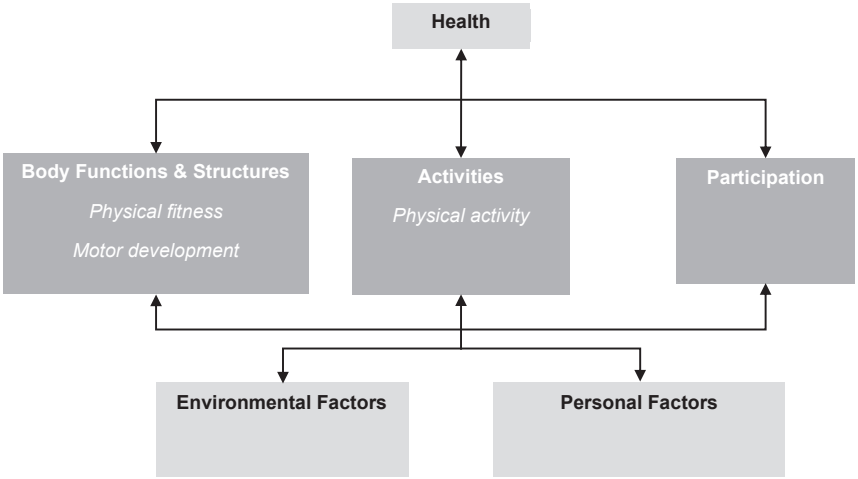

Figure 1. The relationship between the main topics of this thesis and the ICF-framework.

Measuring physical fitness

As I experienced during my work at Reinaerde, measuring physical fitness in children with ID comes with challenges. The limitations in intellectual functioning (e.g. learning, problem solving and adaptive behavior) [3] will influence children's understanding of standardized task instructions and their ability to perform a test according to these instructions. They frequently need adapted communication, such as sign language and icons, and they often need someone to demonstrate the task. Also, these children often lack the understanding or motivation to perform the test with the required complete exertion or maximum performance [56]. In addition to these issues, children with ID often have additional health problems as described before, that can interfere with test outcomes [47, 53, 57-60]. Test results can also be influenced by autism spectrum disorder or challenging behavior, which both are more common in children with ID [8]. Besides the above-mentioned difficulties, visiting unfamiliar settings or being subject to new equipment for test assessments can be stressful and thus hamper test performance. Therefore, the psychometric properties of tests developed for the general population need to be studied in children and adolescents with ID.

Since these testing challenges are likely to increase when the level of ID is more severe, it not surprising that physical fitness testing has hardly been studied in children with more

severe levels of ID, resulting in almost a complete lack of information on physical fitness levels in children with moderate to severe ID. The lack of physical fitness information in this group of children is striking, since more severe levels of ID are related to a higher risk of chronic health conditions [9, 61], making good physical fitness even more important.

Additionally, improving or maintaining physical fitness is mostly the focus of physical therapists in the day program centres, and their therapy is often paid for by medical insurance policies. The therapists, and the insurance companies, want to evaluate the effectivity of applied therapies and interventions, and therefore suitable test are needed.

Aim and outline of this thesis

The aim of this thesis was to study the physical fitness of children with moderate to severe ID, and its relationship with physical activity and motor development. In order to be able to measure the physical fitness in this challenging population, the secondary aim was to find suitable field-based tests.

The main data for this thesis was collected within Reinaerde, a care organization in the Netherlands. Ambulatory children and adolescents with moderate to severe ID attending Reinaerde's day program centers formed the study population.

This thesis starts off in chapter 2 with a systematic review of the literature with regards to instruments measuring health-related physical fitness in children and adolescents with ID. All studies that assessed the psychometric properties of the tests in this population were included.

Based on the literature review, a test selection was made, which we studied for feasibility and reliability in a subsample of the study group. The results of this pilot study are described in chapter 3.

After confirmation of feasibility and reliability in the pilot study, the suitable tests were used to measure physical fitness in a large sample of children with moderate to severe ID. In chapter 4, we describe this cross-sectional study on physical fitness. The results on the fitness tests were compared to reference values of TD children, and the relationship with physical activity and motor development was studied.

In chapter, 5 we further look into the physical activity levels of children and adolescents with moderate to severe ID.

Chapter 6 focuses on maximal heart rates of children and adolescents with ID, which are needed to determine the intensity of physical activities, in order to increase the physical fitness. This was a retrospective study, based on a large dataset of children and adolescents in the United States of America and Spain.

Chapter 7 provides a summary of the key findings, and a discussion on the work presented in this thesis. Implications of the findings for parents, professionals, organizations and government are discussed and directions for future research are suggested.

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