Giving Weight to Cardiovascular Health

The feasibility of a progressive resistance-exercise training program
in adults with intellectual disabilities with cardiovascular risk factors.

Stijn Weterings

COLOFON

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The feasibility of a progressive resistance-exercise training program in adults with intellectual disabilities with cardiovascular risk factors

Gewicht geven aan cardiovasculaire gezondheid

De haalbaarheid van een progressief krachttrainingsprogramma voor volwassenen met een verstandelijke beperking die risicofactoren hebben op het krijgen van hart- en vaatziekten

Proefschrift

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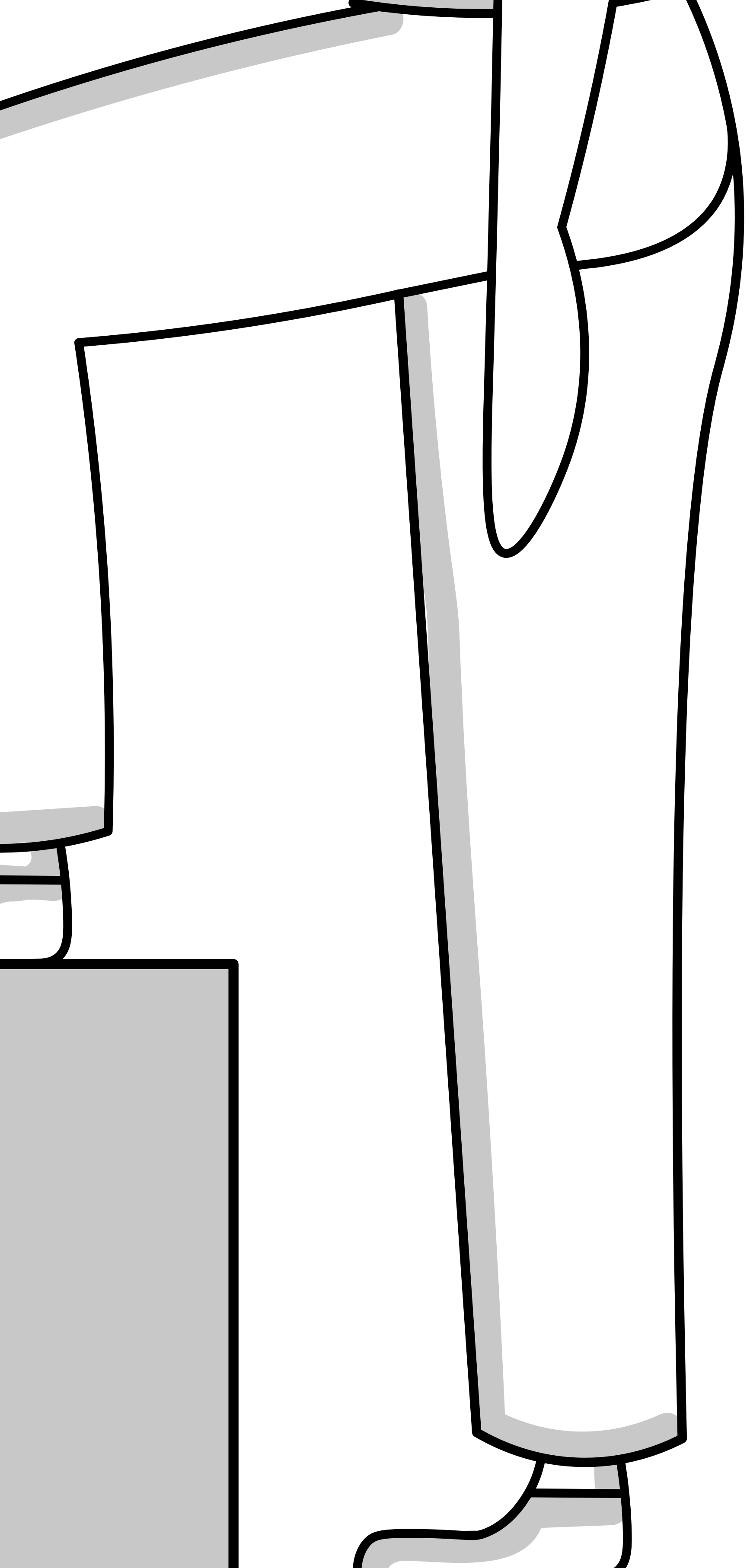

CHAPTER 1

General Introduction

GENERAL INTRODUCTION

BACKGROUND

The World Health Organization (WHO) estimates that between 1-3% of the world's population has an intellectual disability (ID) (McKenzie et al. 2016). The definition states that ID originates before the age of 18 and is characterized by significant limitations in both intellectual functioning and adaptive behaviour, which covers many everyday social and practical skills (AAIDD 2010). An IQ under 70 is often used as a cut-off for limitations in intellectual functioning, and based on IQ scores the severity of ID is further divided into mild to profound (Woittiez et al. 2014). Approximately 0.85% of the Dutch population has an ID (Woittiez et al. 2014). This is a relatively small percentage, however in 2017 the healthcare costs for this group were 9.4% (8.3 billion euros) of the total Dutch healthcare budget (RIVM 2019). There is a general consensus that the demand for care and support for people with ID will continue to rise in many countries (Coppus 2013, Woittiez et al. 2018). This is because people with ID nowadays live longer due to improved healthcare (Patja et al. 2000, Coppus 2013). Also more people with borderline ID will be in need of professional support due to our modern society demanding more and more intellectual, practical and social skills (Woittiez et al. 2014).

CARDIOVASCULAR DISEASE RISK IN PEOPLE WITH ID

People with ID generally have more health problems than the general population (Draheim et al. 2002, Draheim 2006, Bastiaanse et al. 2012, de Winter et al. 2012, de Winter et al. 2012, Evenhuis et al. 2012, Jansen et al. 2013). Some are linked to the specific cause of the ID, but many are at least partially lifestyle related. Multiple studies show that the prevalence of cardiovascular disease (CVD) risk factors is high in (older) adults with ID (de Winter et al. 2012, Hilgenkamp et al. 2012, Franke et al. 2018). Diabetes, hypertension, obesity (in men), and metabolic syndrome (a cluster of the interconnected CVD risk factors dyslipidaemia, high blood pressure, type-2 diabetes mellitus and abdominal obesity) were as prevalent in older adults with ID as in the general population (de Winter et al. 2012, Axmon et al. 2017). Peripheral arterial disease, and obesity (in women) were even more prevalent in older adults with ID than in the general population (de Winter et al. 2012, de Winter et al. 2013, Axmon et al. 2017). In the general population increased physical activity (PA) and exercise are found to improve CVD risk factors. Increasing PA levels and exercising can reduce or even eliminate medication use for CVD risk factors (American College of Sports Medicine 2022). However, the effect of PA or exercise on CVD risk factors has not been widely studied in adults with ID.

The standard therapy for people with ID when diagnosed with CVD risk factors such as type-2 diabetes mellitus, hypertension and dyslipidaemia, is still providing medication (Axmon et al. 2017, Taggart et al. 2021). However, because of the effects of PA on CVD risk factors seen in the general population, increasing PA levels might be a good alternative. This might especially be a good alternative because only 9% of adults with ID meet the PA levels compliant with public health recommendations (Dairo et al. 2016). Also, physical activity and physical fitness levels of adults with ID are very low (Hilgenkamp et al. 2012, Hilgenkamp et al. 2012). These low PA levels and low physical fitness increase the risk of many chronic diseases, such as type-2 diabetes mellitus, cardiovascular diseases (CVD) and osteoporosis in the future (Savage et al. 2011, Beltran Valls et al. 2014, Piepoli et al. 2016). This thesis will search for ways to improve the health of adults with ID with CVD risk factors through exercise.

PREVENTION

The recent Dutch guideline on PA, similar to most international guidelines, prescribes at least 150 minutes of moderate PA a week spread out over multiple days to stay healthy and reduce health risks. This PA should include bone and muscle strengthening exercises at least twice a week, and the amount of sitting should be reduced (Gezondheidsraad 2017, Van Den Berg 2020). Recently it has been added to the guidelines that doing more than you do now is already better and being physically active longer, more intensive or more frequent is also better for your health.

The Dutch government and Dutch health insurance companies acknowledge the need for prevention programs in the general population to promote a healthy lifestyle by improving PA and fitness levels, a healthy diet, and quit smoking and alcohol abuse. For the first time national health insurance providers are covering the costs of participating in approved general prevention programs, underlining the need for preventative action. Furthermore, the ‘European guidelines on CVD prevention in clinical practice’ states that prevention programs to prevent CVD risk factors should not just target older adults who are already at risk but should also target adults of all ages at risk for developing CVD (Piepoli et al. 2016).

The importance of preventative action was also highlighted recently during the COVID-19 pandemic when it was found that an unhealthy lifestyle and sedentary behaviour proved to be correlated with more severe symptoms after a COVID-19 infection and a higher risk for hospitalisation (de Frel et al. 2020, Holly et al. 2020, Gao 2021).

Adults with ID are less likely to receive health promotion interventions than the general population (Hanlon 2018). This could be caused by the fact that adults with ID in general have more behavioural and motivational challenges and physical limitations (Bossink et al. 2017, American College of Sports Medicine 2022). There are many studies that report the need to resolve this health inequality for adults with ID through the development of more ID-specific health promotion programs (Bhaumik et al. 2008, Ng et al. 2015, Franke et al. 2018, O’Leary et al. 2018). There have been some studies into training programs to promote PA and/or physical fitness for adults with ID, but so far these evidence-based programs are mainly focused on improving PA and physical fitness (Shields et al. 2008, Calders et al. 2011, van Schijndel-Speet et al. 2016, Dijkhuizen et al. 2018), and not so much on reducing CVD risk factors directly. Therefore, this thesis we address the lack of exercise programs developed for adults with ID with CVD risk factors.

BENEFITS OF EXERCISE ON CARDIOVASCULAIR DISEASE RISK

There is an irrefutable relationship between the level of PA and many chronic diseases, such as type-2 diabetes mellitus, cardiovascular diseases and osteoporosis, where higher levels of PA are linked to a lower risk of these chronic diseases (Fiuza-Luces et al. 2018). Exercising (more) is a good way to achieve a higher level of PA (American College of Sports Medicine 2022). A review by Fiuza-Luces (2018) explained the potential underlying mechanism of the exercise benefits on CVD (Fiuza-Luces et al. 2018). Skeletal muscles contract during PA, and during these contractions they produce, express and release special hormones, called myokines. These myokines work in a hormone-like fashion and exert specific endocrine effects on distant organs such as adipose tissue, liver, pancreas, bones and the brain. Through these endocrine effects they, for example, contribute to the reduction of insulin resistance (preventing diabetes), hypertension, and dyslipidaemia, thereby reducing the risk for developing CVD and improving health. Increasing PA creates a higher myokines-response. This relationship is dose dependent, meaning that more and more vigorous exercise results in more and more myokines being released into circulation, which lowers CVD risk even more. Because myokines are produced and released by muscle contractions, a loss of muscle mass is in itself already a risk factor for developing CVD. The myokines also counteract the adipokines released by adipose tissue in the body. Adipokines are mostly pro-inflammatory and an accumulation can result in chronic inflammation, which is often the case in people who are overweight/obese. With the low PA levels seen in adults with ID, increasing PA and exercising could therefore be a promising intervention to help reduce CVD risk in adults with ID, and improve their overall health. Therefore this thesis will focus on finding a feasible exercise program to improve health of adults with ID with CVD risk factors.

WHAT TYPE OF EXERCISE IS NEEDED TO REDUCE CVD RISK

Traditionally, aerobic training (AT) has been recommended to improve health and physical fitness and lower CVD risk. Only, AT may not be the most effective and successful intervention for adults with ID. In daily practice, physiotherapists and movement specialists experience that most adults with ID quit AT exercises at higher intensities, when they start to feel that their heart is pounding, their breathing is getting heavier and they begin to sweat. This is a problem because it is necessary to continue AT for a sustained period before a physical training response is seen that results in beneficial effects on CVD risk factors. Therefore, AT seems less feasible for reducing CVD risk and increasing physical fitness and health in adults with ID.

More recently, resistance training (RT) has been recommended as an alternative training mode to reduce CVD risk (Colberg et al. 2010, American College of Sports Medicine 2022). RT is a form of PA in which a muscle or a muscle group is working against an external resistance (American College of Sports Medicine 2022). The American College of Sports Medicine (ACSM), the American Heart Association, the American Diabetes association and the European Society of Cardiology provide clear guidelines for RT to improve cardiovascular health. These guidelines state that for an effective workout to reduce CVD risk, all large muscle groups (a full body workout) need to be involved and trained at high intensity (75-80% 1RM) (American College of Sports Medicine 2022).

RT has potentially even more additional benefits in comparison to AT. RT results in an increase in muscle strength and mass, which has beneficial effects on reducing sarcopenia (Savage et al. 2011, Beltran Valls et al. 2014), which is also present at a relatively young age in adults with ID. Increases in muscle strength and mass also improve the ability to perform daily activities (Bastiaanse et al. 2012).

Instead of the long training bouts needed for AT, RT only requires short bouts of a limited number of repetitions followed by a resting period. This could be more feasible for adults with ID to maintain and RT could therefore be a better type of exercise modality than AT to reduce CVD risk in this population. However, it is not known which resistance exercises are feasible for adults with ID to execute, because of the physical and cognitive limitations adults with ID frequently experience (American College of Sports Medicine 2022). Furthermore, it is known that a higher volume and more vigorous RT can cause greater gains in muscle strength and health (Grgic et al. 2018, Grgic et al. 2018, American College of Sports Medicine 2022), and greater reductions in CVD risk factors (American College of Sports Medicine 2022). However, it is unknown whether RT at vigorous intensity is feasible for adults with ID. So, before a study can start to examine the effects of a vigorous RT-program on CVD risk in adults with ID, we first have to study the feasibility of a vigorous RT-program.

Therefore, this thesis will focus on the important questions regarding the feasibility of an RT-program: Which exercises can be used in an RT-program for adults with ID; Can individuals with ID perform RT at vigorous intensity; Is it safe for adults with ID to exercise at vigorous intensity; and do the participants attend the training sessions and what are their experiences with the RT-program?

EXTERNAL SUPPORT IS NEEDED

A recent study by Kuiken et al. showed that most adults with ID do know what a healthy lifestyle is but need much more external support to achieve it (Kuijken et al. 2016). It is difficult for adults with ID to reflect on their own behaviour, to understand the long-term consequences of their behaviour, and to make the necessary lifestyle adaptations in their daily life, such as what groceries to buy and how to become more physically active. For adults with ID, this external support has to come mainly from their support system, such as health care professionals, family and friends. So, in prevention programs for a healthy lifestyle for people with ID the social support system plays a key role.

Health professionals such as general practitioners, physiotherapists, nurse practitioners, and dieticians have the necessary knowledge and skills to activate people and support good dietary habits. However, their focus is now mainly on curative therapy and they are only marginally involved in the prevention of health problems by means of health promotion (Kuijken et al. 2019). There is an increasing awareness in health care professionals and policymakers that prevention is important and it gets more and more attention. This is a new area for health professionals to work in and there is a lack of evidence based health programs specifically for adults with ID to support health care professionals with this. Therefore, this thesis will include health care professionals to provide this external support for adults with ID.

EXERCISE TESTING TO EVALUATE PROGRAMS

Exercise tests are used to measure the various components of physical fitness. They are used to evaluate exercise interventions and therapies and to compare fitness levels over time or between groups. Testing the physical fitness of adults with ID comes with specific challenges. They often have difficulties performing fitness tests, due to motivational, behavioural, cognitive and/or physical challenges (American College of Sports Medicine 2022). They are not always used to exercising, may stop before an exercise test is completed, may not always be able to perform a test at their best capacity, can have difficulties understanding the test instructions, or may not be able to participate in certain tests at all (American College of Sports Medicine 2022). Phrases like ‘as hard as you can’ or ‘as fast as you can’ are abstract and difficult for most adults with ID to comprehend and put into action. It is also often more difficult for them to focus on the task at hand due to attention problems, thereby quitting the test before the test is complete (Bossink et al. 2017, American College of Sports Medicine 2022). These problems limit the number of tests that can be used in adults with ID compared to the general population. Furthermore, the research on exercise testing in adults with ID is still limited, leaving many questions unanswered with regards to the psychometric properties of exercise tests (Hilgenkamp et al. 2012, Oppewal et al. 2019).

The large epidemiological Healthy Ageing and Intellectual Disability (HA-ID) study into the health of older adults with ID led to the development of the ID-fitscan, a set of feasible, reliable and valid fitness tests to measure the physical fitness of adults with ID (Oppewal et al. 2019). It is the first evidence-based set of exercise tests to measure physical fitness in adults with ID in clinical practice, but this set also has its limitations. There are currently no tests included in the ID-fitscan to measure the strength of specific muscle groups. Neither is there a test included that measures cardiorespiratory fitness (CRF), because there is yet no feasible, valid and reliable CRF test available. In daily practice and research, there is a strong need to measure the muscle strength of specific muscle groups and CRF. Additionally, as most tests are used to evaluate exercise programs or interventions, responsiveness is an important psychometric property of exercise tests. Unfortunately, the responsiveness of tests is often unknown, as is the case for the tests included in the VB-fitscan. In this thesis we try to fill in some of the gaps in knowledge by looking for a feasible CRF test, a feasible and reliable test to measure the muscle strength of specific muscle groups and by assessing the responsiveness of muscle-strength and muscle-endurance tests in adults with ID.

PSYCHOMETRIC PROPERTIES OF EXERCISE TESTING

One of the ways to validate a potential submaximal CRF test is comparing it with a 'gold standard'. The gold standard for measuring CRF is a maximal CRF test on a treadmill or bicycle ergometer in which participants have to walk, run or bicycle until exhaustion. A valid test result is assured when the participant has reached a plateau in the maximal oxygen uptake (VO_{2max}). Unfortunately, many participants with and without ID don't reach a plateau before stopping the test, because of fatigue. Instead of reaching a plateau they reach a peak in VO_2 (VO_{2peak}). A supramaximal exercise test after the maximal CRF exercise test, in the same session, is a valid way in the general population to verify if the obtained VO_{2peak} score is a representation of the true maximal score (MacDougall 1991, Rossiter et al. 2006). Supramaximal means a workload of 110% above the peak workload attained during the CRF. However, the question is whether supramaximal verification after a maximal exercise test is feasible for adults with ID.

For muscle strength measurement, the gold standard is an isokinetic strength measurement with a Cybex or Biodex. However, this equipment is very expensive, big and not portable and therefore not feasible to use for most professionals in daily practice. Muscle strength measurement with a handheld dynamometer (HHD), like the Microfet2, is more promising to use in daily practice. HHD is portable, relatively cheap, and has been found feasible in the general population (Stark et al. 2011, Douma et al. 2014, Schrama et al. 2014, Mentiplay et al. 2015). The question is whether muscle strength measurement with the HHD is also feasible for adults with ID.

Next to feasibility, the test-retest reliability of the HHD is also unknown. The reliability is defined as the degree to which the measurement is free from measurement error (de Vet et al. 2015). This means that when a measurement is repeated it does not show much change in scores. For the HHD, the question is whether muscle strength measurement with the HHD is reliable for adults with ID.

Finally, responsiveness is an important property of a test to evaluate the change over time. Responsiveness is defined as the manner in which a test changes when there is a true change over time (de Vet et al. 2015). There is very little research into the responsiveness of fitness tests in the general population and even less in people with ID, despite its importance for daily practice. However, at the end of the RT-program, it is important to know whether the participants are indeed stronger, and we need responsive tests to pick up on differences. The question is whether muscle strength tests are responsive in adults with ID.

AIM AND OUTLINE OF THIS THESIS

The aim of this thesis was to design and study the feasibility of vigorous resistance training for adults with ID with CVD risk factors. The secondary aim was to study the above-identified gaps in the psychometric properties (feasibility, and/or reliability and/or responsiveness) of different fitness tests to measure physical fitness in adults with ID.

Chapter two of this thesis starts with the design of a feasible resistance-exercise set for adults with ID (RESID) to be used as a total body workout.

This RESID is then incorporated into the vigorous RT-program, of which the feasibility is studied in adults with ID with CVD risk factors in **chapter three**.

Then the focus shifts to the psychometric properties of fitness tests, with a study into the feasibility of supramaximal exercise test to verify if the peak oxygen uptake scores obtained during a graded maximal treadmill test in adults with ID represent the maximal oxygen uptake **(chapter four)**.

Chapter five continues with the feasibility and reliability of muscle strength measurements of the arms and legs with a HHD in adults with ID with CVD risk factors.

The responsiveness of five muscle strength and strength endurance tests is studied in **chapter six**, in which we correlate the test results with the progression made in the RT-program.

The general discussion, **chapter seven**, starts with a summary of the key findings after which the implications of this study for professionals and researchers and directions for future research will be discussed.

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CHAPTER 2

A Resistance Exercise Set for a total body workout for adults with an Intellectual Disability, a pilot study

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ABSTRACT:

Background: Resistance training has beneficial effects on fitness levels, cardiovascular disease risk, risk of sarcopenia and on performing activities of daily living. The focus of this study is to design a total body Resistance Exercise Set for adults with an Intellectual Disability (RESID) with minimal equipment required, and to test its feasibility.

Method: The RESID was selected in an expert meeting, and its feasibility was determined in a cross-sectional pilot study. The feasibility was determined with completion rate, correct execution of exercises, and the participant's experience.

Results: The expert group (n=7) selected seven exercises for the RESID. The participants (N=11) performed the RESID twice during regular sports classes. Completion rate and correctness were excellent for all exercises. The participants did not experience any major problems with the RESID.

Conclusions: The RESID is feasible for use in different training settings. A physiotherapist or fitness-instructor is required to supervise the training sessions.

INTRODUCTION

Physical activity (PA) is important to maintain physical and psychological health (World Health Organization 2003). However, only a small proportion of adults with ID are physically active at a level that is compliant with public health recommendations (Peterson et al. 2008, Hilgenkamp et al. 2012). Additionally, their fitness levels are very low (Hilgenkamp et al. 2012, Oppewal et al. 2013).

To increase health and physical fitness, aerobic training (AT) is traditionally recommended, but resistance training (RT) is nowadays also recommended (Colberg et al. 2010, ACSM 2013). RT has potential additional benefits in comparison to AT. RT focuses on increasing muscle strength, which has positive effects on performing activities of daily living (ADL) and for reducing the age-related decline in muscle mass, called sarcopenia (Savage et al. 2011, Beltran Valls et al. 2014). Furthermore, in sedentary older adults compliance rates seem to be higher in RT-programs than in AT programs (Dunstan et al. 2002, Hong et al. 2008).

The ACSM guidelines give clear recommendations for health-improving RT: all large muscle groups need to be trained at vigorous intensity (ACSM 2013). However, it is often difficult for adults with ID to perform RT-exercises adequately, because they often have motor control issues; can have motivational problems; require more encouragement; often use PA influencing medication; and generally have a shorter attention span (ACSM 2013). Therefore, the ACSM recommends that simple instructions and close supervision have to be provided (ACSM 2013). Also, because of the motor control issues, the use of weight machines during training is recommended (ACSM 2013). However, in daily practice many adults with ID are less likely to have the opportunity to access these kinds of training facilities.

There are only few studies done with an RT-program for people with ID. These studies used fitness equipment (Suomi 1998, Podgorski et al. 2004, Carmeli et al. 2005, Machek et al. 2008, Shields et al. 2008, Calders et al. 2011, Mendonca et al. 2011), and/or did not describe the exercises used (Podgorski et al. 2004, Carmeli et al. 2005). A resistance exercise set for a total workout, without weight machines, that is feasible for adults with ID, would make resistance training more accessible for a larger number of adults with ID and potentially align future research.

Therefore, the focus of this study was to design a Resistance Exercise Set for a total body workout for adults with mild or moderate ID (RESID), without the use of weight machines, and to determine its feasibility.

METHOD

Study design

This study consisted of an expert meeting to select appropriate exercises for the RESID, and a pilot study to test the selected exercises for feasibility (Bowen et al. 2009, Thabane et al. 2010). This study was part of the 'Healthy Aging and Intellectual Disabilities' (HA-ID) consortium; a consort of three care providers for people with ID in the Netherlands; Abrona (Huis ter Heide), Ipse de Bruggen (Zoetermeer) and Amarant (Tilburg) in collaboration with the Chair for Intellectual Disability Medicine of the Erasmus MC, University Medical Centre Rotterdam (Hilgenkamp et al. 2011).

The expert meeting:

Representatives of six physiotherapy teams and four teams of physical-activity-instructors of the three care providers were invited to the expert meeting. A semi-structured interview format was used, which allowed the group to discuss main topics and to explore unanticipated topics as they arose from the group's discussion (Berg et al. 2011). The experts discussed: 1) Which conditions determine the feasibility of RT exercises (i.e. safety, support, duration, setting)?, 2) How to motivate participants (motivation techniques, do's and don'ts)?, 3) Which exercises for the RESID would be most feasible for a total body workout for adults with ID in accordance with existing guidelines (6-8 large muscle groups; concentric, eccentric and isometric; multi-joint and single-joint exercises) (Haskell et al. 2007, Colberg et al. 2010, ACSM 2013). The aim was to reach a broad consensus on all topics.

Pilot study

The pilot study regarding the feasibility of the RESID was held during regular sports classes of the participating care providers Amarant and Ipse de Bruggen (convenience sample). To participate in the pilot study participants had to be 18 years or older, diagnosed with mild (IQ = 50-69) or moderate (IQ = 35-49) ID, and participate in these sport classes. These sports classes are given once a week for adults with mild to moderate ID. Both groups normally performed aerobic exercises (cycling or treadmill walking) and/or fitness exercises with weight machines. Within these sport classes, 13 potential participants with mild (n = 11) and moderate (n = 2) ID were asked to participate, of which eleven participants (six male, ten mild ID) provided informed consent. Participants were excluded when significant physical problems inhibited exercise participation when there was no medical clearance provided by their physician.

The pilot study consisted of two training sessions. A physical-activity-instructor or physical therapist explained and demonstrated each exercise to the group. The participants performed two sets of ten repetitions per exercise. Exercises were performed without weights for safety reasons. During exercising, the instructor positively motivated and supported the participants as much as needed. The warming-up and cooling down (both 5-10 minutes) consisted of easy cycling or treadmill walking.

Participant characteristics

The participant characteristics (age, sex and level of ID) were recorded at the start of training one.

Workout characteristics

Attendance was recorded at the start of training. Tailoring of the exercises was recorded when exercises were adapted to meet the physical and mental capabilities of the participants.

Feasibility

The feasibility of the RESID was scored on the parameters 'completion rate', 'correctness of exercise execution', and 'participants' experience'.

Completion rate

Completion rate of each exercise of the RESID was defined as the percentage of participants completing two sets of ten repetitions characterised as low (<25%), moderate (≥ 25% - <50%), good (≥50% - <75%), and excellent (≥75%) (Hilgenkamp et al. 2013).

Correctness of exercise execution

Correctness of the execution of each exercise was defined as the percentage of participants capable of performing the exercise safe and with good posture and technique characterised as low (<25%), moderate (≥25% - <50%), good (≥50% - <75%), excellent (≥75%) (Hilgenkamp et al. 2013). The researchers, who were present during the training, evaluated this.

Participants' experience

A custom-made questionnaire was used to evaluate the experience of the participants with each exercise. The questionnaire gave insight into the experience, the difficulty, and the acceptance of the participants regarding each exercise. Participants could mostly respond on a 5-point Likert scale. Some questions were open questions, so participants could give feedback in their own words.

Feasibility

The overall feasibility of the RESID based on these three parameters was interpreted as (Thabane et al. 2010):

1. Feasible without adaptations necessary, when the completion rate and correctness were good to excellent, and no major problems arose from the participants' experience for all exercises of the RESID.

1. Feasible with adaptations necessary, when the completion rate and correctness were less than good in both training sessions, and/or a major problem arose from the participants' experience for one or more exercises of the RESID.
2. Not feasible, when both completion rate and correctness were low in both training sessions, and/or multiple major problems arose from the participants' experience for multiple exercises of the RESID.

Statistical analyses

Baseline characteristics (age, sex, level of ID), attendance, completeness, and correctness were analysed with descriptive statistics. Participants' experiences were analysed with descriptive statistics and feedback of the participants was described. Data was analysed with SPSS version 21 (IBM Corporation, New York).

Ethical Considerations

Participants and/or their legal representatives provided informed consent. The Medical Ethical committee of the Erasmus MC, University Medical Centre Rotterdam approved this study (MEC-2016-242).

RESULTS

Expert meeting

Five physiotherapists (out of six invited) and two physical-activity-instructors (out of four invited) participated in the expert meeting (n=7).

Conditions for RT exercises

The experts stated that from a motivational perspective, the less exercises the better. Because of the often short attention span the maximal duration of a session should be 60 minutes. Furthermore, special attention is required for the breathing instructions during exercising, because most adults with ID tend to hold their breath during exercising.

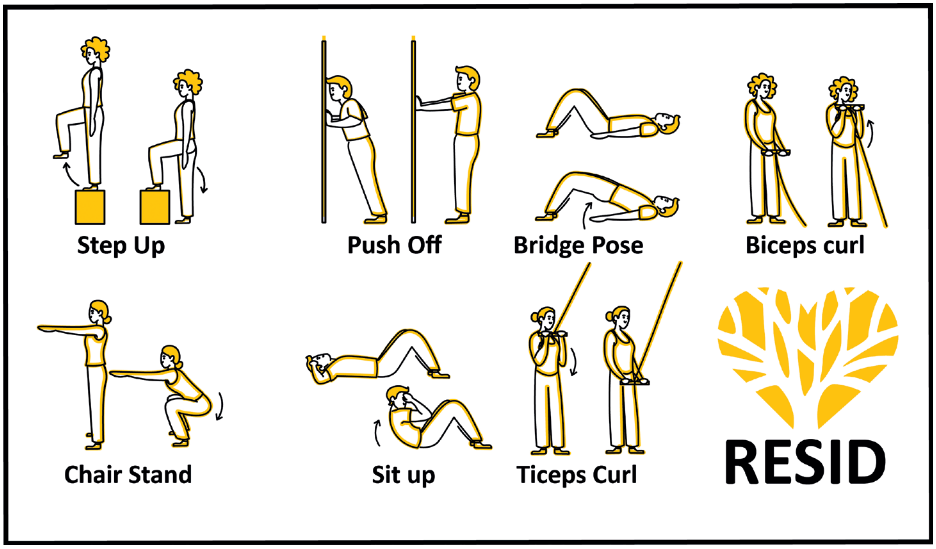
Motivating participants to perform the RT exercises

The experts raised motivation as the key topic with regard to the compliance with RT exercises Besides using music and simple games to enhance compliance, as mentioned in the ACSM guidelines (ACSM 2013), the experts added that maximal external motivation (from caregivers and/or family), humour during training, positive reinforcement and rewards should be used to stimulate enjoyment.

Selected exercises for the RESID

The experts stated that alternative exercises without weight machines are possible. The motor control issues of adults with ID however stress the need for easy and safe exercises. Because of the large variety in physical capabilities it was considered important that the exercises of the RESID can be tailored to the physical capabilities of an individual. The researchers used 46 exercises as a starting point for a group discussion. Consensus was reached on a set of seven exercises (figure 1). The seven exercises are considered a core set, which can easily be tailored to the capabilities of the individual (see appendix I for examples).

Figure 1: RESID exercises



Pilot study

Participant characteristics

Eleven participants (six male, ten mild ID) participated in the pilot study. The mean age was 28.7 (±8.1) years (range 19-44) (table 1).

Workout characteristics

The total attendance of both training sessions was 77% (17/22 sessions). Seven participants joined both of the training sessions (64%) (Table 1). One participant did not join either one of the sessions after providing informed consent (9%), because of a lack of motivation for exercising. Three participants skipped session two, because of other social appointments.

Two exercises of the RESID were tailored to the physical capabilities of the participants. Because the seated squat was too difficult to perform for eight participants, this exercise was altered by squatting with a ball behind their back against the wall. The push off was replaced by a push up for four participants (one with knee support), because the push off was too easy.

Completion rate

In the first session, all two sets of ten repetitions were performed for the step up, the biceps curl and the triceps curl (100% completion rate). One participant performed just one set of the push up, because of not liking the exercise this first session, resulting in a 90% completion rate for this exercise. One participant did not perform the seated squat, the abdominal curl and the bridge pose, because of back pain, resulting in a 90% completion rate for those exercises. In the second session, all participants performed all sets and repetitions of all exercises, resulting in an excellent completion rate (100%).

Table 1: Definition of feasibility of the RESID.

1. Feasible without adaptations necessary	The completion rate was good to excellent, the correctness was good to excellent, and no major problems arose from the participants' experience for all exercises of the RESID in neither one of the training sessions.
2. Feasible with adaptations necessary	Completion rate and correctness was less than good in both training sessions, and/or a major problem arose from the participants' experience for one or more exercises of the RESID in either one of the training sessions. The RESID should be re-evaluated or adjusted.
3. Not feasible	Both completion rate and correctness were low in both training sessions, and/or multiple major problems arose from the participants' experience for multiple exercises of the RESID in either one of the training sessions.

RESID: Resistance Exercise Set for adults with Intellectual Disabilities.

Correctness of exercise execution

In the first session, all participants performed the step up (100%) and the push off (100%) correctly . Only one participant could not correctly perform the seated squat (90%), abdominal curl (90%), bridge pose (90%), and the biceps and triceps curls (90%). In the second session, all participants performed all exercises correctly (100%), which results in an excellent completion rate.

Participants' experience

The exercises were liked by 70-100% of the participants from much to very much, and 70-100% of the participants wanted to perform the exercises again during their regular sports classes. Two participants found the seated squat and push off difficult to perform and one participant found the abdominal curl and bridge pose difficult. The biceps curl was the only exercise all participants wanted to perform again.

DISCUSSION

The purpose of this study was to design a feasible resistance exercise set for a total body workout for adults with mild or moderate ID (RESID), which can be performed without the use of weight machines. The completion rate of the exercises was excellent (90-100%), as well as the correctness of execution (89-100%).Overall, the participants liked the exercises and found them easy to perform. Therefore, the RESID seems feasible for adults with mild to moderate ID and has the potential to be used in RT-programs.

The RESID is a core set of exercises and can be tailored to the physical capabilities of an individual with ID, without changing the essence of an exercise (appendix I). This is in line with the recommendation of the experts in this study and the ACSM to individualise exercise training (ACSM 2013). Professional expertise is required to tailor the exercises of the RESID adequately.

This is the first study to develop a feasible, weight machine free, resistance exercise set for a total body workout specifically for adults with mild to moderate ID, which has the potential to be used in a RT-program. Strong aspects of this study were that the RESID was designed with the expert knowledge of physiotherapists and physical activity instructors experienced in working with adults with ID, within the guidelines for resistance training of the ACSM (ACSM 2013). Furthermore, the heterogeneity of the participants in the pilot (age, sex) provides confidence the results can be feasible for a larger group of adults with a mild and moderate ID, even more because the exercises can be tailored to the physical capabilities of the participants.

A limitation is that a small convenience sample of attendants of sport classes was used for this study. Therefore participants were probably more active and motivated to exercise. Furthermore, just one participant with a moderate ID was included. These aspects limit generalization of the results to the population of adults with ID. For safety reasons the exercises of were performed without weights. So, even though the performance (completion and correctness) of the exercises was excellent, it remains unclear how adults will perform the exercises with weights, and as part of an RT-program.

Future research should therefore study if the RESID is feasible in a larger group of adults with ID and if they are able and willing to perform the RESID in an RT-program with increasing weights at moderate or vigorous intensity. This will be our main question in our next study into the feasibility of a 24-week RT exercise program, with the RESID as selected exercises.

Conclusion

The RESID is a feasible resistance training exercise set for adults with mild or moderate ID, and has the potential to be used in different training settings without the need of expensive weight machines. The RESID seems a feasible core set of exercises in daily practice, with the possibility to tailor one or more exercises to the physical capabilities and physical fitness of each adult with ID. A physiotherapist, physical-activity-instructor or fitness-instructor needs to be available to closely supervise the training sessions and adapt exercises where necessary.

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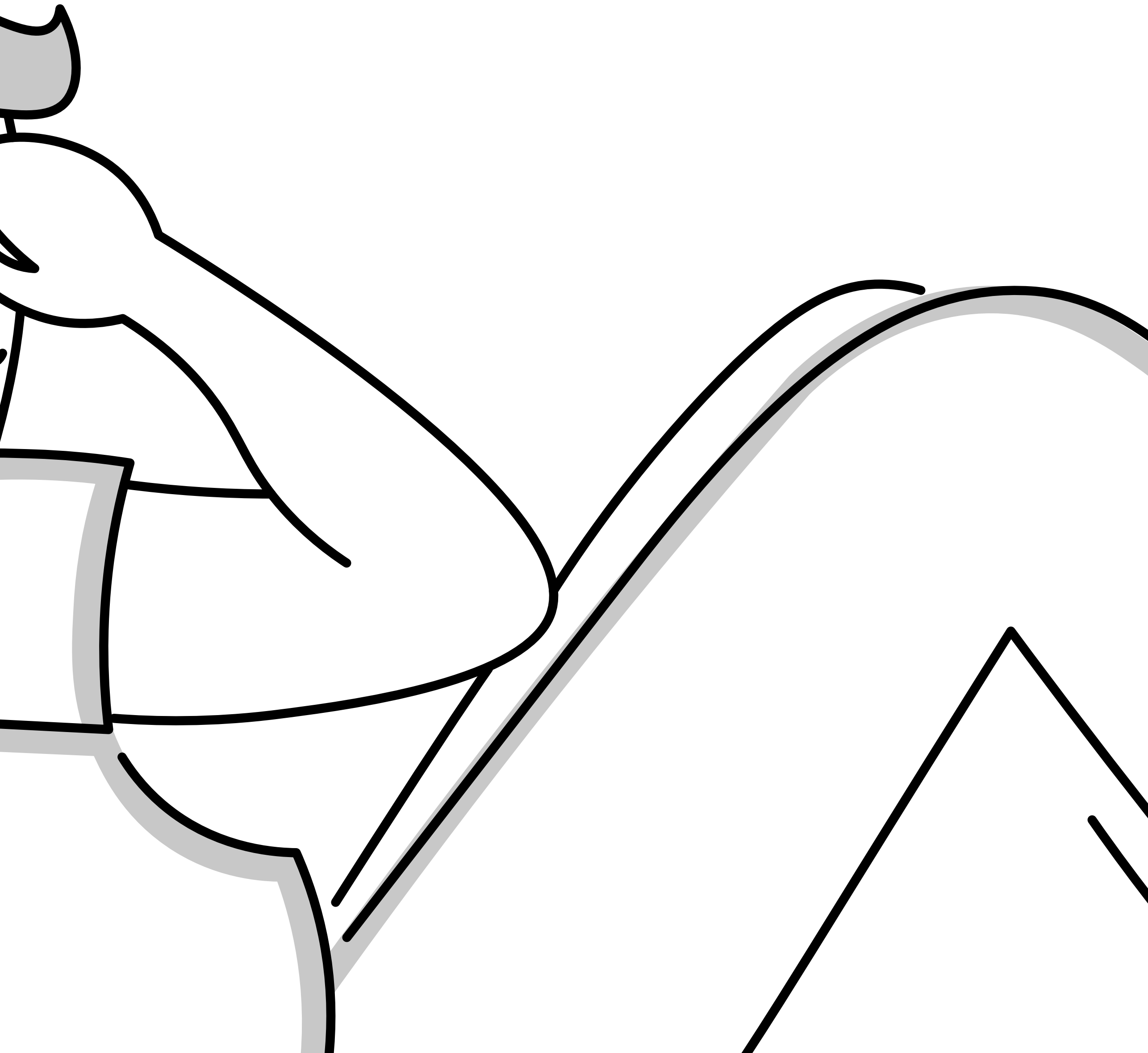

CHAPTER 3

The feasibility of vigorous resistance exercise training in adults with intellectual disabilities with cardiovascular disease risk factors

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ABSTRACT:

Background: The cardiovascular disease (CVD) risk is high in adults with intellectual disabilities. This CVD risk can potentially be decreased with a resistance training (RT) program at vigorous intensity, following previous research on successful High Intensity Training-programs. Our aim was to explore the feasibility of a vigorous RT-program for adults with intellectual disabilities with CVD risk factors.

Method: Twenty-four adults with intellectual disabilities with at least one CVD risk factor participated in a 24-week RT-program. The training intensity was increased from novice (50%1RM) to vigorous (75-80%1RM). Feasibility was based on the achieved training intensity at the end of the RT-program.

Results: Nineteen participants finished the RT-program. Feasibility was good as 58% (11 out of 19) of the participants worked out at vigorous intensity at the end of the program.

Conclusions: It is feasible for the majority of adults with intellectual disabilities with CVD risk factors to exercise at vigorous intensity.

INTRODUCTION

The prevalence of cardiovascular risk factors, such as type-2 diabetes mellitus, hypertension, dyslipidaemia and a high waist circumference, is high in adults with intellectual disabilities (de Winter et al. 2012, Room et al. 2016). These risk factors indicate an increased risk of cardiovascular disease (CVD), which is an important cause of mortality in adults with intellectual disabilities (Sobey et al. 2015, Oppewal et al. 2018). In the general population, these CVD risk factors can be positively influenced by physical activity (PA), thereby preventing the development of CVD (Colberg et al. 2010, Umpierre et al. 2011, Cornelissen et al. 2013, Ishiguro et al. 2016, Riebe et al. 2018). However, the proportion of adults with intellectual disabilities who participate in PA consistent with public health recommendations is low (Peterson et al. 2008, Hilgenkamp et al. 2012). Increasing PA through training and exercising may, therefore, be an effective strategy to reduce CVD risk and prevent the development of CVD in this population.

Traditionally, aerobic training (AT) is recommended to reduce CVD risk, but nowadays resistance training (RT) is also recommended (Colberg et al. 2010, Riebe et al. 2018). Both AT and RT can be customized to the capabilities of the participants to assure optimal participation, safety and effectiveness. However, RT has potential additional benefits compared to AT. Firstly, RT seems to be a more attractive way of exercising than AT for overweight and sedentary individuals (Holten et al. 2004). In line with that, compliance rates have been shown to be higher in RT-programs than in AT-programs in sedentary older adults (Dunstan et al. 2002, Hong et al. 2008). Secondly, RT has a double positive impact on the resting metabolic rate, because RT results in more muscle mass which requires more energy at rest, and RT causes micro-trauma in muscle tissue that requires energy for the muscle remodelling processes (Westcott 2012). Finally, an increase in muscle strength has positive effects on performing activities of daily living (ADL) and is beneficial for reducing sarcopenia (age-related loss in muscle mass), which is already prevalent at a young age in people with intellectual disabilities (Savage et al. 2011, Bastiaanse et al. 2012, Beltran Valls et al. 2014). These advantages of RT make it interesting to explore the potential of an RT-program for adults with intellectual disabilities at risk for developing CVD.

From studies in the general population we know that to effectively reduce CVD risk with an RT-program, it is necessary to train all large muscle groups at vigorous intensity (Riebe et al. 2018). Furthermore, a gradual increase in training intensity is advised for novice trainees, until vigorous intensity is reached (Riebe et al. 2018). Besides these general recommendations regarding RT, there are some special considerations to take into account when exercising with adults with intellectual disabilities. Adults with intellectual disabilities can have motivational problems, often use medication that might impede being physically active, often have motor control issues, generally have a shorter attention span and often do not want to continue exercising when there is a physical discomfort (Bossink et al. 2017, Riebe et al. 2018). These considerations influence the feasibility of an RT-program for adults with intellectual disabilities.

Studies regarding RT-programs specifically for adults with intellectual disabilities are scarce and no studies have been performed in adults with intellectual disabilities with CVD risk factors. Most studies performed a combination of RT and AT or RT and balance exercises (Carmeli et al. 2005, Calders et al. 2011, Mendonca et al. 2011, van Schijndel-Speet et al. 2016). Some studies only focused on people with Down Syndrome (DS) and not intellectual disabilities in general (Shields et al. 2008, Mendonca et al. 2011). People with DS have syndrome-specific mental and physical problems, such hypotonia and ligament laxity, which impairs the generalization of these results to people with intellectual disabilities in general. Also, most studies did not report training intensity (Podgorski et al. 2004, Machek et al. 2008, van Schijndel-Speet et al. 2016). The studies that included resistance exercise at moderate to vigorous intensity focused only on healthy adults with mild intellectual disabilities (Podgorski et al. 2004, Carmeli et al. 2005, Calders et al. 2011). It is therefore not known whether RT at vigorous intensity is feasible for adults with mild and moderate intellectual disabilities with CVD risk factors.

A feasibility study is most suited to address this question (Thabane et al. 2010, Eldridge et al. 2016). We expected that an RT-program would be feasible for adults with ID, as long as the RT-program can be fitted to the physical possibilities of each individual and because the short bouts during resistance training make it easier for the individuals with ID to focus on the exercise and makes it easier for the trainers to motivate them. Therefore, the aim of this study was to explore the feasibility of a 24-week RT-program progressing to a vigorous training intensity for adults with intellectual disabilities with CVD risk factors. Additionally, this study aimed to examine the dropout, attendance, safety and experience of the participants, as well as the experience of trainers of the 24-week RT-program.

METHODS

Study design

This multicentre observational feasibility study was part of the 'Healthy Aging and Intellectual Disabilities' (HA-ID) consortium; a consort of three care providers for people with intellectual disabilities in the Netherlands; Abrona (Huis ter Heide), Ipse de Bruggen (Zoetermeer) and Amarant (Tilburg) in collaboration with the Chair for Intellectual Disability Medicine of the Erasmus MC, University Medical Center Rotterdam (Hilgenkamp et al. 2011).

Participants

All participants lived and/or worked in a residential or community-based setting of the participating care providers for people with intellectual disabilities in the Netherlands.

Individuals with intellectual disabilities within the residential setting were invited to participate by their nurse practitioner if they were diagnosed with a mild (IQ = 50-69) or moderate (IQ = 35-49) intellectual disability, older than 18 years, and diagnosed with at least one CVD risk factor (type 2 diabetes mellitus, hypertension, hypercholesterolemia and/or overweight/obesity). Participants were excluded when physical problems inhibited exercising or when there was no medical clearance given by the physician. All participants or their legal representatives gave written informed consent. This study was performed in accordance with the Helsinki declaration (WMA 2013). The medical ethics committee of the Erasmus MC, University Medical Center Rotterdam, the Netherlands, approved this study (MEC-2016-574).

RT-program

Training sessions

The participants completed a 24-week RT-program, with two training sessions a week (48 sessions in total). Each session lasted approximately 60 minutes, and started with a five-minute warm-up of low intensity aerobic activity (cycle ergometer or treadmill), after which the resistance exercises were performed. Each session ended with cooling-down and stretching exercises for five minutes. The participants were supported and supervised by a physiotherapist or physical activity instructor during the entire program to ensure good posture, safety and support. An instruction session at the start of the program was provided for all trainers to make sure they understood the training protocol and exercises and were able to execute them correctly. The trainer-participant ratio was 1:1 (n=8), 1:2 (n=12), or 2:4 (n=4), depending on the participant's preferences to train individual or in a group, and depending on the organisational possibilities (training time and day, location, availability of trainers). The training sessions were performed at different locations, either at a local physiotherapy practice (n=13), at home (n=1), or at a local fitness centre (n=9). Both the trainer-participant ratios and training locations are feasible options in daily practice and therefore used in this study.

Exercises

The RT-program consisted of seven exercises (step up, push off/up, seating squat, abdominal curl, bridge pose, biceps curl and triceps curl). In our previous pilot study, these exercises had been recommended by experienced physiotherapists and physical activity instructors working with adults with intellectual disabilities and found feasible to perform (Weterings et al. 2018). However, the RT exercises were not set in stone; when necessary trainers could tailor the exercises to the physical capabilities of the participant (Weterings et al. 2018). Researchers were available to provide the trainers with feedback throughout the program when adapting an exercise of the RT- program to make sure the participant performed a complete workout as intended.

Progression in training intensity

Most participants were novice trainees and therefore the RT-program had five phases, with increasing training intensity in each phase (see table 1) (Riebe et al. 2018). Each phase consisted of at least eight sessions, so the bodies of the participants could adapt to the physical strain of the exercises in order to prevent injuries. To move to the next phase, at least five out of the seven exercises should be performed with good posture and breathing technique during eight training sessions. The first phase was the familiarization phase, in which the participants were introduced to the exercises, training posture and breathing techniques.

The training intensity was described as the percentage of an one repetition maximum (1RM), which is ‘the greatest resistance that can be moved through the full range of motion in a controlled manner with good posture’ (Riebe et al. 2018). For safety reasons there was no 1RM-measurement of each exercise at the start of the program, because training posture and breathing techniques were not trained yet. Instead, the trainer selected a weight for each exercise with which he/she expected that the participants could perform a maximum of 20 repetitions (exercising at 50%1RM), which was then set as number of repetitions during the familiarization phase. The participants were asked to work to tolerance or until the intended repetitions were reached. After this starting point, whenever participants performed two sets of the intended repetitions in a controlled manner with good posture and breathing technique, the training weight was increased by ?5% or the smallest amount possible for each exercise, without changing the number of repetitions (Riebe et al. 2018). As participants moved on to the next phase, the number of intended repetitions decreased to correspond with the level of %1RM (see table 1) while the training weight was increased, to make sure weight and intended repetitions corresponded with the 1RM-score of the previous training session. The trainers logged the intended training intensity, training weights and performed repetitions of each training session, exercise and set. Within each phase, The recovery between sets was between 30 seconds and 2 minutes depending on the training intensity (see table 1), in accordance with the ACSM guidelines for RT (Riebe et al. 2018).

Table 1: Training intensity per phase.

Phase	% of 1RM	No. of sets	No. of repetitions	Rest between sets
Familiarization	50	2	20	30 sec.
1	60	2	18	30 sec.
2	70	3	12	1 min.
3	75	3	10	1 min.
4	80	3	8	2 min.

1RM: the maximum amount of weight that a person can possibly lift for one repetition over the whole range of motion.

Measurements

Participants' characteristics

Age, sex, the presence of CVD risk factors (type 2 diabetes mellitus, hypertension, dyslipidaemia, and/or overweight/obesity), and diagnosis were derived from medical records. Behavioural therapists or psychologists categorized level of intellectual disabilities as mild (IQ = 50-69) or moderate (IQ = 35-49) for each participant. Body mass index (BMI) was calculated by weight (measured with Seca Robusta type 813, in kilogram) divided by squared height (measured with Seca 216 height rod, in meter). Waist circumference was measured with a flexible tape (in centimetre). All measurements were performed at the start of the RT-program.

The feasibility of training at vigorous intensity

The achieved training intensity at the end of the RT-program was used to define feasibility. Vigorous intensity was defined as training intensity of at least 75%1RM. Feasibility of the RT-program was characterized as low (<25% of participants reached ≥75%1RM), moderate (≥25% and <50% of participants reached ≥75%1RM), good (≥50% and <75% of participants reached ≥75%1RM), and excellent (≥75% of participants reached ≥75%1RM) (Hilgenkamp et al. 2013).

Dropout

The dropout was presented as the percentage of participants not finishing the 24-week RT-program. The researcher logged the dropout after consulting the participant, the trainer and the participants' caregiver. The results of the participants that dropped out were not further used for analyses of this study.

Attendance, safety, participants' experience and trainers' experience

The trainers logged attendance and adverse events of each training session. At the end of the program, a custom-made questionnaire was used to evaluate the participants' experience and trainers' experience. The participant's questionnaire contained questions for the participants about the experience, difficulty and acceptance of the RT-program. The participants responded mostly on a 5-point Likert scale, but some questions were open questions, so participants could give feedback in their own words. The trainers' questionnaire contained open questions regarding the RT-program and their take on the difficulty and acceptance of the RT-program by the participants.

Statistical analysis

The participant's characteristics were analysed for all participants with descriptive statistics. The training intensity, dropout, attendance, and participants' experiences of all participants who finished the 24-week RT-program were analysed with descriptive statistics. The trainers' experience was analysed with descriptive statistics. The additional comments of the participants and trainers were described qualitatively. The data was analysed by using SPSS version 24 (IBM Corporation, New York).

RESULTS

Participants' characteristics

Twenty-four participants (13 women/11 men) with mild (n=13) and moderate (n=11) intellectual disabilities started the RT-program. Seven participants had type-2 diabetes mellitus, seven had hypertension, five had dyslipidaemia and 18 were diagnosed as being overweight or obese. However, our baseline BMI measurement revealed that 22 participants were overweight/obese, one participant was slightly underweight and one had an average BMI (see table 2).

Table 2: Participants' characteristics.

Number of participants	24
Male	11 (45.8%)
Female	13 (54.2%)
Level of ID	
Mild	11 (45.8%)
Moderate	13 (54.2%)
Diagnoses	
Down syndrome	3 (12.5%)
Cerebral Palsy (GMFCS I)	2 (9.5%)
Age (years), mean ± SD [range]	44 ± 17 [23 - 75]
CVD Risk	
Diabetes-mellitus, type 2	7 (29%)
Hypertension	7 (29%)
Dyslipidaemia	5 (20%)
Overweight/Obese	22 (92%)
BMI ± SD [range]	33.9 ± 6.9 [17.4 – 44.2]
Underweight	1 (4%)
Normal	1 (4%)
Overweight	5 (21%)
Obese	5 (21%)
Severe obese	5 (21%)
Morbidly obese	7 (29%)
Waist circumference, mean ± SD [range] in cm.	115 ± 15 [82 – 144]

ID = intellectual disability; GMFCS = Gross Motor Function Classification Score; SD = standard deviation; CVD = cardiovascular disease; BMI = body mass index; cm = centimetre.

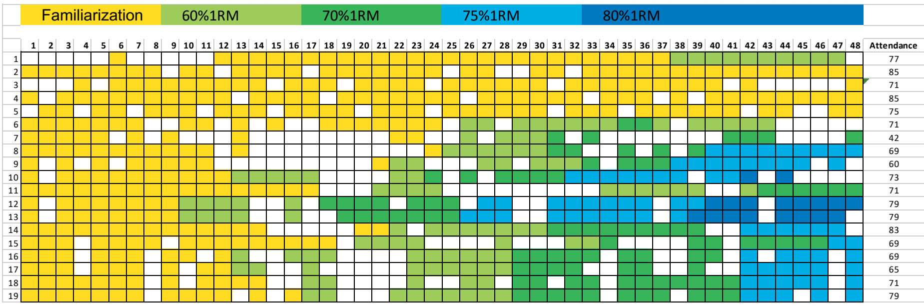
Feasibility of training at vigorous intensity

Nineteen participants finished the 24-week RT-program and 11 out of 19 participants (58%) worked out at vigorous intensity, of which eight at 75%1RM (42%) and three at 80%1RM (16%) at the end of the program. Therefore, the feasibility to train at vigorous intensity was good for adults with intellectual disabilities. Four participants did not exceed the lowest level of training intensity (familiarization, 50%1RM), due to difficulties with increasing training weight. They had difficulties performing an exercise every time the training weight was increased. This prevented the trainer to increase the training weight and thereby the intensity. Two participants worked out at 60%1RM and two participants at 70%1RM at the end of the program (see also figure 1).

Dropout

Five participants (21% of participants, 2 men) did not finish the 24-week RT-program. Three participants did not want to continue training due to motivational problems and two participants stopped due to injuries not related to the program (one participant tore his knee ligaments and one participant fractured his hip).

Figure 1: training intensity of the participants per session.



The participants who finished the RT-program are listed on the left. The coloured squares show the training intensity of each session and the blank squares show the training sessions that were missed. The attendance of each participant was noted on the right in percentages

Attendance

The overall attendance for the participants who finished the program was 73% (an average of 35 sessions per participant, range 40-91%, see figure 1). Training sessions were cancelled due to holidays (36% of the cancelations), absent trainers (11%) and cancelation due to force majeure (fire alarm and cancelation due to a storm warning, 11%). Without the cancelations due to holidays, absent trainers and force majeure the attendance would be 85%. Other reasons for cancellation were illness of the participant (15%), forgotten (4%), did not want to train (4%) and not reported (19%).

Safety

Other than some muscle soreness after training, no adverse events related to the RT-program were reported.

Participants' experience

Most participants (n=18) liked participating in the RT-program and would recommend joining the RT-program to other people. One participant did not understand this question and did not respond. For most participants (n=18), the 24-week duration was not too long and they would join again (see table 3). Eight participants noticed a better performance in daily life after the RT-program (see table 3 for their comments).

Trainers' experience

The trainers responded that all participants were able to perform the RT-program. Even though six participants needed continuous feedback on the execution of the exercises and three participants found it difficult to perform one or two of the exercises correctly. These exercises were performed too fast or not in the full range of motion. Two participants found it difficult to handle increasing weights during training. For six participants the trainers had to make adjustments to the exercises to meet their physical possibilities, for example some participants could not perform the bridge pose, which was adapted to a lateral pull down or a back raise; one participant used the leg raise instead of the abdominal curl and one participant could not perform push ups because of arm length differences and the seated horizontal push was used. Throughout the program close supervision of the trainers was necessary, to ensure good posture and breathing technique. One trainer had to split the training sessions from a group (1:2) to individual training (1:1), because of the negative interaction between the participants.

Table 3: Responses of the participants about their experience with the RT-program.

	Positive	Neutral	Negative	Remarks
Did you like to participate in the RT-program?	18	1		No remarks
Did you like to train at your achieved intensity?	17	2		No remarks
Would you join the RT-program again?	13		6	No remarks
Would you recommend joining the RT-program to other people?	13		5	1 participant did not understand the question
Duration of 24-week RT-program	14		1 too long 4 too short	No remarks
Did you like to train 2x per week?	14	3	2	No remarks
Did you notice a difference in daily life after/during the RT-program?	8	11		"Walking is easier" "My diabetes is stable for the first time" "It is easier to do my daily chores" "I can lift heavy boxes at work now" "Cycling is easier now" "I feel better after training"

RT: Resistance training.

DISCUSSION

The results of this study showed that 58% (11 out of 19) of the participants achieved vigorous training intensity of ≥75%1RM. Therefore, the feasibility to exercise at vigorous intensity was considered good for adults with mild or moderate intellectual disabilities with CVD risk factors. Although the feasibility was good, the overall number of training sessions at a vigorous intensity was lower than expected, which could limit potential health benefits. The step-by-step increase in training intensity, which is advised for novice trainees (Riebe et al. 2018), took half of the training sessions before vigorous training intensity could be reached. This, combined with the fact that 27% of the training sessions were cancelled and that many participants progressed more slowly than expected through the different intensity phases, led to a limited time of training at vigorous intensity or to not reaching vigorous intensity at all in 24 weeks.

Feasibility was anticipated to be negatively influenced by motivational problems and motor control problems many adults with intellectual disabilities experience, and because t adults with intellectual disabilities find it often difficult to continue exercising when there is a physical discomfort (like pushing through resistance, sweating, a raised heartbeat or breathing heavily) (Bossink et al. 2017, Riebe et al. 2018). Therefore, we tried to increase feasibility by working with trainers with a lot of experience in working with adults with ID. The trainers were experienced physiotherapists and physical activity instructors, and all trainers received an instruction session before the start of the program to ensure understanding of the protocol. These trainers were able to adapt exercises, when necessary, to the possibilities and limitations of the participants and ensured that the participants maintained a correct posture during exercising. They also motivated the participants to do their best and perform at the required intensity, all while making sure there was a positive atmosphere during training through humour and positive reinforcement. Furthermore, the RT-program started with a familiarization period tailored to each participant.

Eighteen participants liked the RT-program, thirteen would join again and just 4% of the training sessions were cancelled because the participant did not want to train. Therefore motivation of the participants for the RT-program seemed no problem. It seemed more difficult for adults with intellectual disabilities to perform the RT-program, considering the result that many participants progressed more slowly through the different intensity phases than expected. The participants needed more time to get used the exercises and the exercising, even though the trainers provided close support and supervision. Furthermore, three participants found one or two exercises too difficult to perform during the whole RT-program, two participants found it difficult to handle increasing weights and for six participants alternative exercises had to be used. Therefore, these motor control problems and the physical discomfort during training had a direct impact on the feasibility of the RT-program.

To increase the feasibility, many facilitators mentioned in the literature were used in this study to help the participants to continue training in the RT-program (Bossink et al. 2017, Riebe et al. 2018, Weterings et al. 2018). The trainers tried to create a positive and comfortable atmosphere during training; the participants received a diploma and medal at the end of the program; there was often social interaction with peers; there was always guidance during training from the trainer; and the RT-training was organized close to home.

There was a drop out of 21% (5 out of 24). Two studies, one on RT in adults with a mild intellectual disabilities (Calders et al. 2011) and one study on RT in adults with DS (Shields et al. 2008), had no drop outs and another study of older adults with borderline to profound intellectual disabilities had a comparable dropout of 20% (3 out of 15) (Podgorski et al. 2004). Adults with intellectual disabilities are a heterogenic population with many motivational, behavioural and physical problems. A dropout can occur, despite all efforts to support the participants during training. In our study, five participants dropped out, three due to motivational problems and two due to injuries not related to the RT-program.

For this feasibility study, we also wanted to know how many of the 48 sessions would be attended, to be able to anticipate the attendance in our following effect study. The average attendance was 73%, which was lower than the study of Calder et al. (>90% out of 40 sessions in 20 weeks) (Calders et al. 2011) and of Shields et al. (92.8% out of 20 sessions in 10 weeks) (Shields et al. 2008), but comparable to the study of Podgorski et al. (75% out of 48 sessions in 12 weeks) (Podgorski et al. 2004). A review on RT in adults with type-2 diabetes mellitus in the general population showed an attendance of 75-100% across studies (Umpierre et al. 2011) and a review on the effect of RT on CVD risk factors in overweight/obese children showed an attendance of 76-96% across studies (Dietz et al. 2012). The attendance of our study is comparable with the lower end of the studies mentioned in both reviews. Future studies should anticipate that around 25% of the training sessions will be cancelled and compensate with extra training sessions and/or try to increase attendance. There are some ways to increase the attendance. Future studies can reschedule a new training session when a participant cannot be present and need to make sure there will always be a trainer present. Furthermore, participants (and caregivers) should be reminded that the participant has a training session scheduled, to further increase the attendance.

The participants in this study had a large variation in age, type of CVD risk, level of intellectual disabilities, and sex; which is important in a feasibility study, because they should be representative of the intended population (Thabane et al. 2010). The number of participants in this study was sufficient to answer our research question on whether vigorous RT is feasible for adults with mild to moderate intellectual disabilities and CVD risk factors. Eight participants did report a positive difference in their daily life after the

RT-program. For example, one participant mentioned that walking was easier, another one said that cycling to work was easier, and another participant could better lift the heavy boxes at work.

There are some limitations to this study. The small number of participants and the diversity in age and risk factor limits the generalization of these findings to all adults with intellectual disabilities. It was therefore also not possible to perform subgroup analyses. Furthermore, the participants had actively joined this study, so they wanted to exercise in RT-program, limiting the generalization of the results to all adults with intellectual disabilities. The participant's experiences were derived through a questionnaire that we self-constructed, and therefore not based on an existing questionnaire. With this self-report participants might have provided more favourable answers, therefore results should be interpret with caution. Finally, it remains to be determined if the RT-program can increase muscle strength and if it can positively influence ADL-performance and/or CVD risk factors in adults with intellectual disabilities. Future studies should therefore make efforts to increase the total number of training sessions with vigorous training intensity (for example by training longer, use less phases to reach vigorous intensity after familiarization), thereby increasing the potential health benefits for adults with intellectual disabilities and CVD risk factors. Furthermore, future studies should also focus on an RT-program for adults with severe and profound intellectual disabilities and on adults with physical limitations who were excluded in this study.

Conclusion

It is feasible for the majority of adults with intellectual disabilities with CVD risk factors to exercise at vigorous intensity. Physiotherapists, physical activity instructors or fitness instructors experienced with working with people with intellectual disabilities can use this RT-program to train at vigorous intensity in daily practice for adults with intellectual disabilities, yet close supervision remains necessary during exercising. Vigorous intensity RT seems a promising non-pharmaceutical new option in the prevention of CVD in adults with intellectual disabilities.

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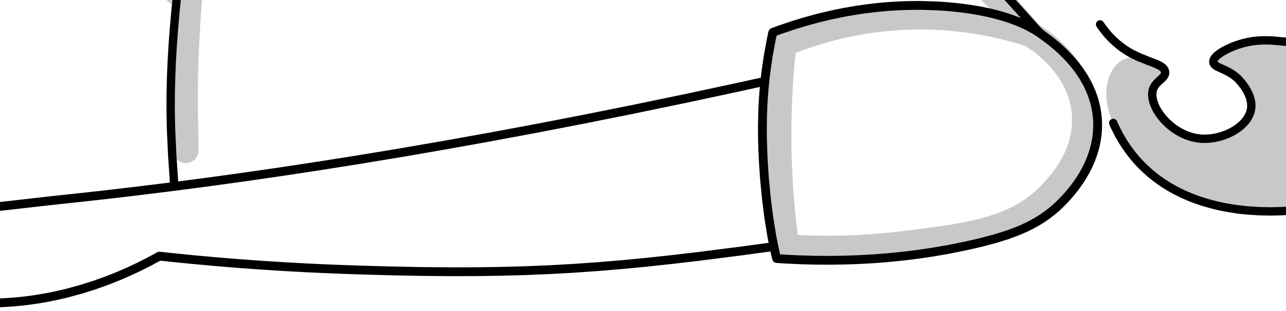

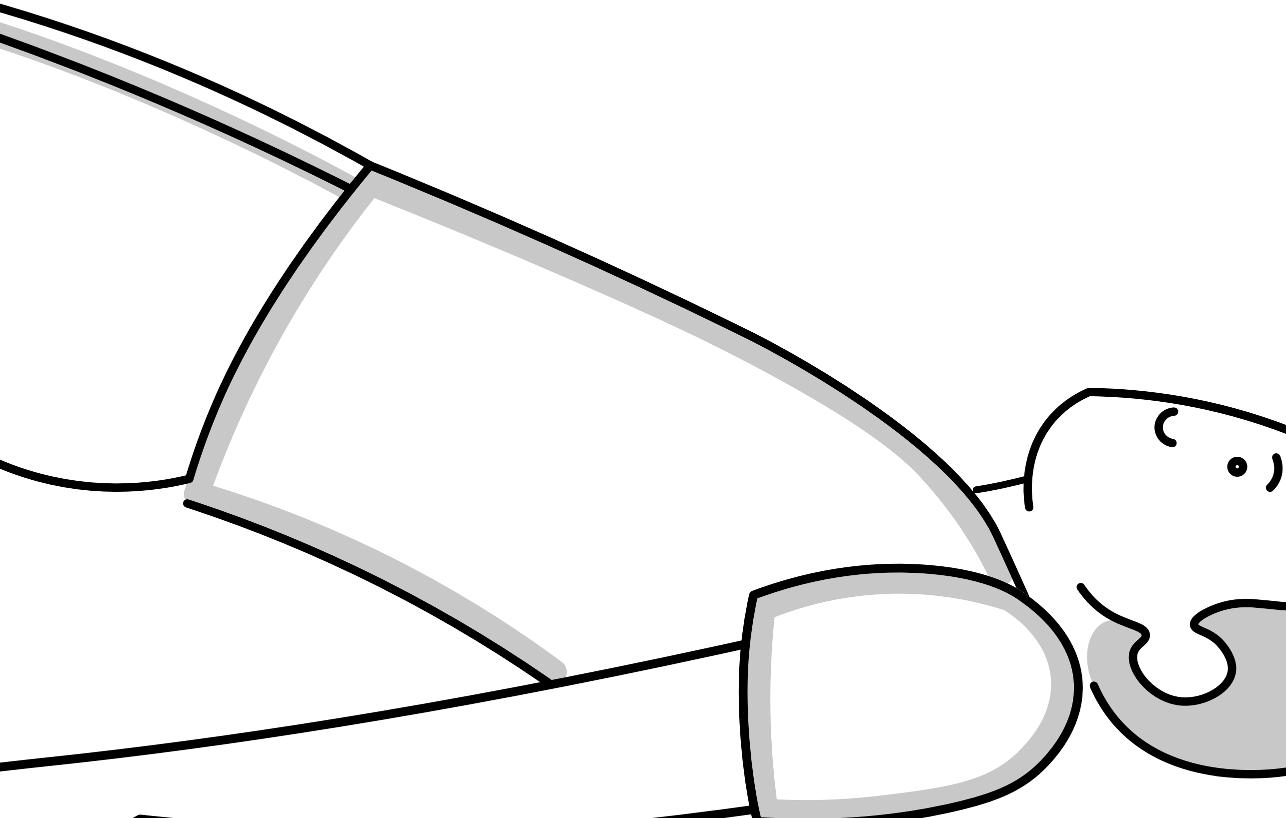

CHAPTER 4

The feasibility of supramaximal verification of peak oxygen uptake of a graded maximal treadmill test in adults with intellectual disabilities

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ABSTRACT

Purpose: To determine the feasibility of aerobic capacity measurement (VO_{2max}) of adults (18-50 years) with intellectual disabilities (ID) through supramaximal verification.

Methods: A cross sectional observational design was used, which consisted of 2 sessions. Prior to testing a 3-step familiarization process was performed (session 1), after which the VO2peak of a graded maximal treadmill test (GXTT) ($VO_{2peakGXTT}$) and a supramaximal exercise test (SET) ($VO_{2peakSET}$) were measured (session 2). The feasibility parameters used in this study were completion rate (based on the criteria for maximal effort), agreement between the $VO_{2peakGXTT}$ and the $VO_{2peakSET}$ (through intraclass-correlation-coefficient (ICCagreement)) and acceptance (through a custom-made questionnaire).

Results: 12 Participants performed the measurement procedure. The completion rate was 75% and the ICCagreement was 0.99 ($p<0.001$). The questionnaire showed that the measuring procedure was acceptable for the participants.

Conclusion: This study demonstrated that it appears feasible to perform a maximal treadmill test (GXTT) followed by a supramaximal exercise test for adults with ID. Therefore, it can be possible to verify the VO_{2peak} of the GXTT as a true VO_{2max} in adults with an ID.

INTRODUCTION

Intellectual disability (ID) is characterized by significant limitations in both intellectual functioning and adaptive behaviour, covering many everyday social and practical skills and originates before the age of 18 (AAIDD 2010). The prevalence of ID ranges between 1% and 1.25% of the world's population (Maulik et al. 2011).

Research on the physical fitness of adults with ID shows lower physical fitness in this population (Janicki et al. 1999, Graham et al. 2000, Frey et al. 2005, Hilgenkamp et al. 2012). Their physical fitness can be compared to the physical fitness of a general population 20 years older (Hilgenkamp et al.). Physical fitness is important for independence, well-being and health (Cowley et al. 2010, Bartlo et al. 2011). Research in adults with ID has identified that cardiorespiratory capacity, one component of physical fitness, is a major indicator for one's overall fitness (Fernhall 1993, Pitetti et al. 1993). However strong evidence is lacking on the validity of maximal and submaximal cardiorespiratory exercise tests in adults with ID, because little research has been done in this area (Vis et al. 2009, Hilgenkamp et al., Oppewal et al.). More research is needed on the validity of submaximal tests such as the 6 minute walk test (6MWT) and the Incremental Shuttle Walking Test (ISWT) in adults with ID (Vis et al. 2009, Hilgenkamp et al. 2012, Oppewal et al. 2013). Norm scores need to be established on the cardiorespiratory fitness of the adults with ID (Oppewal et al. 2013) that will help to determine the need for and effect of prevention and intervention programs, which attract more interest in the last decade (Rimmer et al. 2010, Calders et al. 2011, Oviedo et al. 2014, van Schijndel-Speet et al. 2016). Knowledge about the age-related decline of cardiorespiratory fitness can be used to identify threshold levels below which cardiorespiratory fitness will become dangerously low, which is associated with an increased risk for all-cause mortality. Oppewal et al. 2013). Furthermore since the cardiorespiratory fitness levels are low across the entire population with ID, there is a need to investigate possible determinants for low cardio respiratory fitness levels, including the influence of specific (genetic) syndromes (Oppewal et al. 2013). So there is a need for a valid maximal test to measure the cardiorespiratory fitness of adults with ID to be able to determine health risk factors, evaluate therapy in clinical practice and to validate submaximal cardiorespiratory exercise tests. A graded maximal treadmill test (GXTT) is a maximal cardiorespiratory exercise test and has been used in adolescents and younger people with ID (Fernhall et al. 1990, Fernhall et al. 1998), but because of the following number of limitations these findings cannot be generalized to adults with ID (Oppewal et al.).

First, the studies into cardiorespiratory fitness of people with ID using maximal cardiorespiratory exercise tests mainly focused on children and adolescents with Down syndrome (Fernhall et al. 1990, Pitetti et al. 1993, Mendonca et al. 2010). However, people with Down syndrome (DS) could significantly differ in cardiorespiratory fitness compared to adults with ID without Down syndrome, because of syndrome specific chronotropic incompetence, joint laxity and muscle hypotonia (Mendonca et al. 2010) thereby limiting the generalizability of the findings to adults with ID (Oppewal et al. 2013).

Second, the available research shows different GXTT protocols have been used, although they all refer to the same articles for the feasibility and reliability (Fernhall et al. 1987, Fernhall et al. 1990). The measurement procedure almost always includes a familiarization process to optimize maximal testing results, but protocols differ in using a fixed starting speed or an individually based treadmill speed or in using different inclinations in different timeframes (Fernhall et al. 1987, Pitetti et al. 1995, Rintala et al. 1997, Draheim et al. 1999, Tsimaras et al. 2003, Cowley et al. 2010, Mendonca et al. 2011). This raises questions whether the feasibility of the measurement procedure is still applicable to all protocols.

Third, these studies used mostly secondary criteria such as Respiratory Exchange Ratio (RER; $=VCO_2/VO_2$) and maximal or peak heart rate (HR_{max}), to check if the peak oxygen uptake measured during a test (VO_{2peak}) was a true VO_{2max} measurement. Recent studies showed that some participants who met the secondary criteria did not meet their maximal effort, while other participants did not meet any the secondary criteria while a maximum effort is given. Therefore, it is not known whether a maximal cardiorespiratory exercise test reflects true maximal capacity in individuals with ID.

A better way to provide a criterion for establishing VO_{2max} is to use a supramaximal exercise test (SET) after the graded exercise test in a single session test protocol (Thoden 1991, Rossiter et al. 2006). A SET is only valid with the GXTT being completed as well (Rossiter et al. 2006). Supramaximal means a workload above the peak workload attained during a GXTT. A feasible and safe supramaximal exercise protocol consists of performing at a 110% workload of the maximum workload of the GXTT. The SET test has a duration of about 3-5 minutes (Rossiter et al. 2006), whereas GXTT will take on average 10 to 15 minutes. If consistent VO_{2peak} values are found in both exercise tests, the VO_{2peak} can be considered a valid indicator of VO_{2max} attainment. A recent study by Astorino et al. (2009) found supramaximal testing was suited for sedentary adults in the general population (Astorino et al. 2009), which is promising considering most adults with ID lead a sedentary lifestyle (Hilgenkamp et al. 2012). The SET provides opportunities to verify the scores of GXTT protocol in adults with ID establishing true VO_{2max}-scores as a more valid gold standard for adults with ID. This would be a solid starting point for the much-needed research into the validity of submaximal exercise tests for adults with ID. Before a large group of participants with ID is subjected to this onerous burden test for validation or norm scoring, it is important to study the feasibility of this test as a first step. To this date it is not known if adults with ID are able and willing to perform both the GXTT and the SET until exhaustion.

This study explored three aspects (completion rate, agreement and acceptability) of feasibility of measuring VO_{2max} with a GXTT followed by a SET. Prior to testing there was a 3-step familiarization process for the participants to get acquainted with the test equipment and procedure. A feasibility study is most suited for this research question (Bowen et al., Thabane et al. 2010). Therefore the aim of this study was to examine the feasibility of a graded maximal treadmill test combined with a supramaximal exercise test in adults (18-50 years) with ID.

MATERIALS AND METHODS

Participants

A convenience sample was used for this study. All participants had to live and/or work in a residential setting for people with ID in the Utrecht region of the Netherlands. Individuals with ID within the residential care setting were invited to participate if they were diagnosed with ID, between 18 and 50 years old and able to follow test instructions. Participants were excluded when significant ambulatory problems inhibited treadmill walking or when there was no medical clearance given by the doctor. All participants and their legal representatives gave written informed consent. The medical ethics committee of the Erasmus Medical Centre at Rotterdam, the Netherlands, approved this study (MEC-2014-603).

Design

This feasibility study consisted of 2 sessions. Session 1 consisted of the 3-step familiarization process and session 2 consisted of the test protocol. The test protocol (session 2) started with the GXTT (with a total duration of about 10-15 minutes) followed by a recovery period of at least 10 minutes or until the heart rate (HR) dropped below 120 beats/minute, before performing the SET (with a duration of about 3-5 minutes at a workload of at least 110% of the GXTT) (Rossiter et al. 2006). There were at least 24 hours between the 2 sessions to prevent fatigue hampering the outcome of the test.

OUTCOME

Baseline characteristics

The baseline characteristics were derived from the medical records (age, gender and level of ID). In the first session overall physical fitness was tested with the "VB-fitscan" (ID-fitscan), which consists of measuring weight, length, waist circumference, 30-second chair stand (30sCS), 5-times-chair-stand (5tCS), grip strength, static balance, and walking speed. All tests have been found feasible and reliable for people with ID (de Jonge 2010, Hilgenkamp et al. 2012).

Feasibility

The three feasibility parameters for this study were completion rate, agreement between VO_{2peak} scores of the GXTT and SET and acceptance of the measurement procedure by the participants and the investigators (described in more detail below). The overall feasibility of the measurement procedure was interpreted as [1] feasible without adaptations necessary, [2] feasible with adaptations necessary or [3] not feasible (see table 1 for detailed definitions) (Thabane et al. 2010).

Table 1: Definition of feasibility

Feasible without adaptations necessary	Both the completion rate and the agreement between $VO2_{peakGXTT}$ and $VO2_{pekSET}$ were good to excellent and no major problems arose from the questionnaire.
Feasible with adaptations necessary	Completion rate or agreement scored less than good to excellent and/or a major problem arose from the questionnaire; the measurement procedure should be re-evaluated or adjusted.
Not feasible	Both completion rate and the agreement between $VO2_{peakGXTT}$ and $VO2_{pekSET}$ scored less than good and/or multiple major problems arose from the questionnaire.

Completion rate

Completion rate was defined twofold; both tests (GXTT and SET) had to be completed and the criteria of maximal effort had to be met for both tests (Rowland 1993). Completion was recorded after the GXTT and the SET separately as completed 'yes' or 'no'. The Rowland criteria were used to define maximal effort (Rowland 1993). The criteria are divided in subjective and objective criteria (see table 2). A participant had to meet at least 2 subjective and 1 objective criteria for each test before a test was considered successfully completed. The predicted maximum heart rate ($HR_{predicted}$) was calculated with the specific formula for people with ID with or without Down Syndrome (DS): $HR_{predicted}$: 210 - (0.56 age) - (15.5 DS), with non-DS coded as 1 and DS coded as 2 (Fernhall et al. 2001). The completion rate was characterised as low (<25%), moderate (≥25% and <50%), good (≥50% and <75%), and excellent (≥75%) (Hilgenkamp et al. 2012).

Table 2: Criteria for maximal effort

Subjective Criteria	Objective Criteria
Unsteady walking	HR_{peak} > 95% $HR_{predicted}$*
Sweating	RER_{peak} > 1.00 (Fernhall & Otterstetter, 2003)
Facial flushing	VO_2 plateau in the last minute
Clear unwillingness to continue despite encouragement	

HR=heart rate; RER= respiratory exchange ratio; $HR_{predicted}$: 210 - (0.56 x age) - (15.5 x DS), with non-DS (Down syndrome) coded as 1 and DS coded as 2 (Fernhall et al., 2001)

Agreement of VO2peak

Only the scores of the participants who completed the test and met the criteria of maximal effort were included. Agreement was assessed through calculation of the normalized VO_{2peak}. Normalized VO_{2peak} was calculated as $VO_{2peakGXTT}$/kg or $VO_{2peakSET}$/kg and expressed as millilitres per minute per kilogram (mL/kg/min). The Intraclass-Correlation-Coefficient ($ICC_{agreement}$, model 2.1) was calculated for normalized VO_{2peak} to analyse agreement (Portney et al. 2009). The $ICC_{agreement}$ scores was be interpreted as Poor < 0.40, Fair 0.40 - 0.70, Good 0.70 – 0.90 and excellent > 0.90 (Coppieters et al. 2002).

Acceptability

A custom-made questionnaire was used to evaluate the participants' acceptance of the familiarizations process and the test. There were questions about the experience, the difficulty and the acceptance of the participants of the total measurement procedure. The participants could mostly respond on a 5-point Likert scale. Some questions were open questions so participants could give feedback in their own words (see Appendix II for the complete questionnaire).

Materials

The same treadmill, a Johnson Health Tech JET-7000N (Johnson Health Tech. Co., Ltd., Taichung, Taiwan) was used for practicing and testing. The VO_2 was measured with a calibrated mobile gas analysis system (Cortex Metamax B3, Cortex Medical GmbH, Leipzig, Germany) and MetaSoft® Studio Software (Cortex Medical GmbH, Leipzig, Germany). The Cortex Metamax is a valid and reliable system for measuring ventilatory parameters during exercise (Medbo et al. 2002, Brehm et al. 2004). HR was measured with a Polar T31 heart rate monitor (Polar Nederland b.v., Almere, the Netherlands).

Procedure

Familiarization process

The familiarization process consisted of 3 steps for the participants to get acquainted with the materials and testing procedure in order to optimize maximal testing results (Fernhall et al.). In step 1 the testing procedures, testing equipment and the testing environment was explained and demonstrated. During step 2 the participants walked on the treadmill at a comfortable speed of between 3 and 6 km/h for up to 5 min. While walking at the highest speed, the treadmill incline was gradually increased in order for the subjects to become accustomed to the changes in grade. During step 3 the whole test protocol (GXTT and SET) was practiced at the testing speed on the treadmill with the measurement gear, mask, and heart rate monitor in place. Only when a participant was familiar with the current step, the next step was introduced.

GXTT and SET

The test was performed on a motorized treadmill, because walking is an activity adults with ID are most familiar with and is found valid and reliable for people with DS and adolescents with ID (Fernhall 1993, Pitetti et al. 1993, Mendonca et al. 2010). To maximize testing results a semi-individualized protocol was used so participants were required to walk at a speed depending on their individual capability (Fernhall et al.). At the start of the GXTT the walking speed was set at comfortable pace for the participant for 2 minutes. Then speed was increased to a fast walk for 2 minutes. The fast walk speed was derived from the average speed from three 5-meter walk test trials walking as fast as possible. The treadmill grade was increased by 2.5% every 2 minutes until 12.5%, after which the speed was further increased by 0.8 km/h every minute until volitional exhaustion. Peak exercise parameters were defined as the values achieved during the last 30 seconds of the test.

The SET starts with a 30-second warm-up walk at comfortable pace, after which the speed was increased to a fast walk for another 30 seconds. Then the grade was increased 1% every 10 seconds to at least 110% of the maximum attained setting during the GXTT or until 15%. After which the grade was kept at 15% and the speed was further increased by 0.9 km/h every minute until volitional exhaustion. Peak exercise parameters were defined as the values achieved during the last 30 seconds before stopping. During the whole test (GXTT and SET) the participants were positively encouraged as much as needed to continue walking.

Table 3: Baseline characteristics of participants

Participants			
Gender n (% male)		**12 (75%)**	
Level of ID n (% male)	Borderline	-	
	Mild	4 (75%)	
	Moderate	8 (75%)	
	Severe	-	
	Baseline characteristics		
	Mean	*SD*	*Range*
Age (y)	30.1	8.0	20 – 45
Body Mass (kg)	83.4	35.1	53.9 – 182.0
Height (cm)	177.2	11.0	156.0 – 198.0
BMI (kg/m^2)	26.0	7.9	18.3 – 46.4
Waist Circumference (cm)	94.7	20.4	71.0 – 138.0

Table 3: Continued

Participants			
	ID-fitscan (n=12)		
	Mean	*SD*	*Range*
30sCS (number)	14.6	3.1	9 – 20
5tCS (sec)	9.75	1.94	8.13 – 14.06
Max. grip strength right (N)	40.7	11.1	18 – 52
Max. grip strength left (N)	38.4	11.6	18 – 51
Static Balance (av. time in sec)	9.12	2.32	2.00 – 10.00
Walking Speed (km/hr)	5.3	1.2	2.6 – 7.0
	GXTT (n=12)	SET (n=11)	
	Mean (SD) *Range*	*Mean (SD)* *Range*	
HR_{peak} (beats/min	164 (29) 112 – 218	170 (23) 128 – 212	
Time (min:sec)	16:26 (4:41) 4:11 – 23:16	6:29 (2:30) 4:23 – 12:34	
Peak Speed (km/hr)	8.6 (3.2) 4.0 – 15.0	8.9 (2.4) 5.8 – 12.8	
$VO2_{peakSET}$ (L/min)	2.48 (0.85) 1.10 – 4.16	2.54 (0.81) 1.26 – 4.03	
$VO2_{peak}$/kg (mL/min/kg)	32.2 (12.4) 15.6 – 51.0	33.6 (11.2) 16.4 – 49.4	
VCO_{2peak} (L/min)	2.94 (1.11) 1.19 – 5.33	2.82 (0.94) 1.23 – 4.33	
VE_{peak} (L/min)	83.71 (31.67) 35.91 – 156.48	85.80 (25.98) 35.89 – 125.83	
RER_{peak}	1.18 (0.20) 0.94 – 1.66	1.13 (.027) 0.85 – 1.80	

ID= Intellectual Disability; SD= Standard Deviation; BMI= Body Mass Index; 30cCS= 30 second chair stand; 5tCS= 5 times chair stand; SD= standard deviation; kg= kilogram; cm= centimeters; m= meters; sec= seconds; N= newton; y= years, GXTT= Graded Maximal Treadmill Test, SET= Supramaximal Exercise Test, Norm= normalized, SD= standard deviation, VO_{2peak}= peak volume of oxygen, RER= respiratory exchange ratio, HR= heart rate, VCO_{2peak}= maximal volume of carbon dioxide, VE= expiratory ventilation, L= liter, min= minute, sec=second, km= kilometer, hr=hour

Data analysis

Completion rate and the achievement of the criteria for maximal effort were analysed with descriptive statistics. VO_{2peak} was analysed with descriptive statistics. Associations between the normalized $VO_{2peakGXTT}$ and the $VO_{2peakSET}$ were examined with the use of the

ICCagreement (model 2.1) (Portney et al. 2009). Bland and Altman (BA) plots were used to display the Limits of Agreement (LoA) between $VO_{2peakGXTT}$ and the $VO_{2peakSET}$ (Bland et al. 1986). In a BA plot the difference between the values of the two tests are plotted against the average of the two tests in order to graphically evaluate the agreement using +/- two standard deviations of the mean difference to define acceptable upper and lower limits. The questionnaire was analysed with descriptive statistics and remarks of the participants were described. The results of the ID-fitscan were used to describe the participant characteristics. All statistical analyses were performed using SPPS for Mac (version 21 SPPS Inc., Chicago, Ill).

RESULTS

Participants

This study included 16 participants. Four participants were excluded after initial inclusion; two for not obtaining medical clearance and two for the inability to make an appointment for performing the tests within the test period. A total of 12 adults with ID (9 male) participated in this study. None of the participants had DS. The diversity of the group is described with the baseline characteristics, results of the ID-fitscan of the participants and results on both the GXTT and SET in table 3.

Completion rate

The completion rate was 75%, which is defined as excellent (9 out of 12 participants) (Hilgenkamp et al. 2012). Eleven participants performed both the GXTT and the SET and 9 of the remaining 11 participants met the criteria for maximal effort (see table 4). One participant did not want to perform the SET, because of discomfort of the mask and fatigue and 2 participants did not meet any of the objective criteria for maximal effort.

Agreement of VO2peak

The ICCagreement between the normalized $VO_{2peakGXTT}$ and the normalized $VO_{2peakSET}$ was excellent; ICC= 0.99 (95%CI 0958 – 0.998; p=< 0.000). The LoA was displayed in a Bland Altman Plot with the difference of the $VO_{2peakSET}$ from the $VO_{2peakGXTT}$ presented in percentage. The differences ranged between -6.4% to 5.7% with a mean difference of 0.95% (figure 1). The differences of the VO_{2peak} of the participants who did not meet the criteria of maximal effort were -14.8% and 13.4%.

Table 4: Criteria for maximal effort

	GXTT		SET*	
Subjective Criteria	*Yes*	*No*	*Yes*	*No*
Unsteady walking	0	12	0	11
Sweating	11	1	11	
Facial flushing	10	2	10	1
Clear unwillingness to continue despite encouragement	12	0	11	0
Objective Criteria	*Yes*	*No*	*Yes*	*No*
HR_{peak} > 95% $HR_{predicted}$*	3	8	4	7
RER_{peak} > 1.00	10	2	8	3
VO_2 *plateau in the last minute*	0	12	0	11

HR_{peak}= peak heart rate; RER_{peak}= peak Respiratory Exchange Ratio; $HR_{predicted}$: 210 - (0.56 age) - (15.5 DS) (Fernhall et al., 2001); VO_2 = *volume of oxygen*
** 1 participant had problems with the heart rate monitor, so no HR-score for the GXTT could be measured and another participant did not perform the SET. Therefore only 11 scores were counted for HR and for the SET scores.*

4

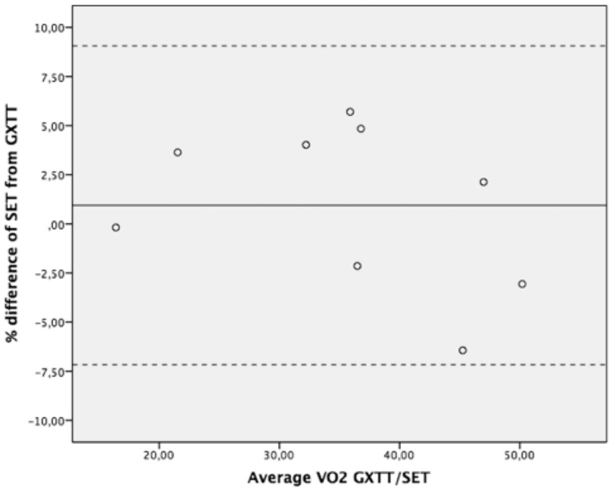

Figure 1: Bland Altman Plot of percentage of difference of $VO_{2peakSET}$ compared to $VO_{2peakGXTT}$ and the average VO_{2peak} of the GXTT and SET in ml/min/kg (N=11). N=9; LoA= -7.2 – 9.1 % (mean ±1.96*SD); Mean difference= 0.95%; Standard Deviation of the difference= 4.1%; Range of -6.4% - 5.7%.

Acceptability

The questionnaire showed no major problems in acceptance of the total test procedure for the participants (see table 5). The participants mostly enjoyed performing the test and would come again to perform the test another time (10 out of 12). The participants frequently had problems with the breathing mask. Eight participants commented that they did not like the mask. One participant refused to wear the mask any longer halfway through testing and stopped the test. Another participant removed the mask between the GXTT and the SET but finished testing. The increase in walking speed after reaching the maximal inclination was difficult for 2 participants. It was not the increase in speed itself, but they commented their locomotion skills made it difficult to keep up with the increase in speed. The instructors responded that the test was acceptable for most participants (11 out of 12).

Table 5: Summary of responses of questionnaire

	Acceptance	*Annoying*	*Remarks*
Breathing mask	4	8	"The mask is annoying." "Reminds me of narcosis"
Heart rate monitor	11	1	-
Walking speed	10	2	"The increasing speed was hard to maintain with my locomotion skills."
Treadmill inclination	12	0	-
Familiarization process	11	1	"Very useful"
Total test	10	2	"When can we do it again?"
Instructors opinion on participants' acceptance	11	1	Because of familiarization, only 2 participants needed extra instruction before testing and 1 participant stopped during the actual test.

DISCUSSION

The purpose of this study was to determine three aspects of feasibility (completion rate, agreement and acceptability) of measuring VO_{2max} with a GXTT followed by a SET. Prior to testing there was a 3-step familiarization process for the participants to get acquainted with the test equipment and test protocol. The completion rate was excellent (75%), the agreement was excellent (ICC 0.99, p=< 0.000) and the questionnaire showed good acceptance. The results of this study show that it is feasible to measure the VO_{2max} through the GXTT and SET and no major adaptations to the measuring procedure are necessary.

The completion rate of the measurement procedure was 75% of the participants. The predetermined standards for acceptable completion rate were ≥50% as good and ≥75% as excellent (Hilgenkamp et al. 2012). This standard was an arbitrary choice. A completion rate of 75% was considered excellent for this study, because of the large heterogeneity of adults with ID and the possible motivational problems of adults with ID (Hilgenkamp et al. 2012). The completion rate of 75% can be used in for future research when calculating the necessary sample size.

It is difficult to verify if a participant has performed with maximal effort in adults with ID. Criteria were therefore used to establish maximal effort. These criteria were divided in subjective and objective. The objective criteria contain thresholds of HR, RER and VO_2 plateau. As stated in the introduction these thresholds have been questionable for establishing VO_{2max}, because of between-subject differences in maximal attainable physiological values (Rossiter et al. 2006, Midgley et al. 2009). Regarding the criteria, it is interesting that only 3 participants reached the $HR_{predicted}$ in both the GXTT and the SET and 1 participant reached it once. One participant even remained more than 30 beats per minute below $HR_{predicted}$. The equation of the predicted maximal heart rate as described by Fernhall (Fernhall et al. 2001) was based on group-level calculations and seems not suited for individual use in people with ID to rely on because of the huge variations in actual maximal heart rates. This is also in line with the study of Oppewal et al. (2014) into heart rate recovery in older adults with ID (Oppewal et al. 2014). Regarding the other criteria, none of the participants reached a VO_2-plateau during the tests. The RER_{peak} was >1.0 for 8 of the participants in both tests with a lower $RER_{peakSET}$ for all participants compared to the $RER_{peakGXTT}$. Although these criteria were questionable ways to establish VO_{2max} (Rossiter et al. 2006, Midgley et al. 2009), they seem to give a good impression of whether the performance was with considerable effort. We found participants meeting an objective criterion had a much smaller percentage of difference of VO_{2peak} between the GXTT and the SET.

The ICCagreement between the VO_{2peak} on the GXTT and the SET was very high with a score of 0.99. This is very promising, but it has to be taken into account that the wide range of VO_{2peak}/kg scores amongst the participants, the small differences of the GXTT and the SET and the low number of 9 participants may have caused an overestimation of the ICCagreement. The BA Plot of the LoA showed an average difference in VO_{2peak}/kg scores of 0.95% of the participants' performance on the GXTT and a range of -6.4% to 5.7%. Some scores are above the coefficient of variation of 3.9% Midgley et al. (2006) found in a study of distance runners (Midgley et al. 2006), but the coefficient of variation for adults with ID needs to be determined in future studies. This coefficient of variation can be used as verification criterion threshold of the VO_2 scores of the GXTT and the SET.

Overall the acceptance of the measurement procedure was good, but the use of a breathing mask was annoying for 8 out of 12 participants. A breathing mask had to be used to measure the ventilation of the participants. So even though we used a user-friendlier mask, which covers mouth and nose instead of a mouthpiece with a nose clip, the mask was still considered uncomfortable. However, the breathing mask is in our opinion the best choice to measure the ventilation. Introduction and familiarization of the breathing mask seems important to ensure acceptance and therefore performance of the participants.

Strengths

This study is the first to study the feasibility of cardiorespiratory fitness (VO_{2peak}) measurement through supramaximal testing for adults with ID living in a residential setting. Previous studies of maximal testing in adults with ID are not focused on residential living setting,(Fernhall et al. 1987, Nasuti et al. 2013) concern younger adults with DS(Fernhall et al. 2009, Mendonca et al. 2010), and/or have no supramaximal verification of VO_{2peak} (Fernhall et al. 1987, Kittredge et al. 1994, Nasuti et al. 2013).

A strong aspect of this study is the careful familiarization process to allow the participants to become familiar and comfortable with the testing procedures. The use of a familiarization process is necessary for a good result as recommended in previous studies (Fernhall et al. 1987, Mendonca et al. 2010). The equipment is unknown to most participants (especially the breathing mask and the treadmill), most of them are not familiar with test protocols and most of them are not used to performing with maximal effort. The questionnaire on acceptability showed high appreciation of the familiarization process. A familiarization process helps the participants to feel confident with their ability to perform the test, resulting in better performances of the test.

This study shows the performance of the SET after the GXTT gives more insight in whether the performance represented a true maximal effort and thus a true VO_{2max} score. The criteria for maximal effort seem to give a first impression in this study, but to be more certain the GXTT scores and the SET scores can be compared and maximal effort can be confirmed (Rossiter et al. 2006). As a previous study by Midgley et al. (2006) showed a coefficient of variation for distance runners (Midgley et al. 2006). Future research also needs to establish a coefficient of variation as the maximum verification criterion threshold for adults with ID. For this study the threshold is unknown. This could implicate that although 9 participants' tests met the criteria of maximal effort, they were still not all with maximal effort.

Limitations

Major limitations of this study are the use of a convenience sample and the exclusion of participants with significant ambulatory problems. The adults with ID who participated in this study were physically active and most of them had an active lifestyle. The participants were probably more used to getting tired and were more likely to perform with maximal effort. Most of the adults with ID however, lead a sedentary lifestyle (Hilgenkamp et al. 2012, Dairo et al. 2016) and are not used to sweating, to feeling their heartbeat and to getting exhausted. Therefore the findings of this study cannot be generalized to all adults with ID, but this study is a promising start and more research is needed to see if VO_{2max} can be measured in other groups of adults with ID.

For an accurate measurement it is necessary to choose the right speed for the fast walk. An underestimation of the fast walk speed results in a test time above the desired test time and could lead to muscle fatigue instead of volitional exhaustion. So despite the protocol being initiated as a walking test protocol; some participants had to run instead of just walking before volitional exhaustion was reached. For the GXTT and SET a certain amount of time and effort is needed for a good score (Astorino et al. 2009, Midgley et al. 2009). Although the wide range of VO_{2max} scores underline using a semi-individualized protocol, the average test time for both the GXTT and the SET in this study were above the intended duration of 10-15 and 2-5 minutes respectively. Possibly the chosen fast walk speed of most participants underestimated their possibilities, which led to an increase in test time. In this study muscle fatigue could have caused an underestimation of the true VO_{2max}. Some extra time in the familiarization process should be spend in getting the participants familiar with walking on the treadmill and choosing a fast enough walk speed, but not too fast, so the test time stays between the preferred timeframe.

The treadmill had a maximum inclination of 15%, after which it was necessary to increase the speed. It was noted that some participants had problems with their locomotion skills when the speed was increased, which could have led to an early stop of the test. For future research it would be advised to use a treadmill with higher inclination possibilities.

Future directions/clinical implications

This study was performed in adults with a mild to moderate ID living in a residential setting with a more active lifestyle. The feasibility of the measurement procedure in adults with ID with a more sedentary lifestyle as well as in adults with a more moderate to severe ID remains to be determined. Furthermore, it is not known what the maximal difference between the VO_{2peak} scores of the GXTT and the SET can be to still reflect a true VO_{2max} in adults with ID. The Bland Altman plot on LoA showed a range of differences of VO_{2peak} between -6.4% and 5.7% in this study, but the verification criterion threshold of the VO_2 scores for adults with ID still needs to be determined. The findings of this feasibility study are promising for future research aiming to use the measurement procedure with supramaximal verification of the VO_{2peak} of the GXTT, to establish true maximal effort and therefore a true VO_{2max} in adults with ID. There is a growing interest in having population appropriate representation or female specific results. Future research should take into account that among adults with ID the female-to-male ratio varied between 0.7 and 0.9 (Maulik et al. 2011). A study of Hilgenkamp (2014) on subgroups in older adults with ID showed there was no sex difference in participation, but there was a difference in physical fitness between male and female, age and level of ID (Hilgenkamp et al. 2014). Future research can establish valid norm scores on the cardiorespiratory fitness of the adults with ID. This study is just the first step, which could in time lead to the potential gold standard of the test procedure to validate the much-needed, more practical, less onerous submaximal exercise tests. These submaximal exercise tests for adults with ID can then be used to determine health risk factors and evaluating prevention, intervention and therapy programs in clinical practice.

Conclusion

This is the first study investigating maximal and supramaximal cardiorespiratory testing in adults with ID. We show that it seems feasible to perform the measurement procedure of a familiarization process and a maximal treadmill test (GXTT) followed by a supramaximal exercise test (SET) in adults with mild to moderate ID. Therefore, it can be possible to verify the VO_{2peak} of the GXTT as a true VO_{2max} in adults with an ID. The results of this feasibility study encourage further research on measuring the cardiorespiratory fitness of adults with ID.

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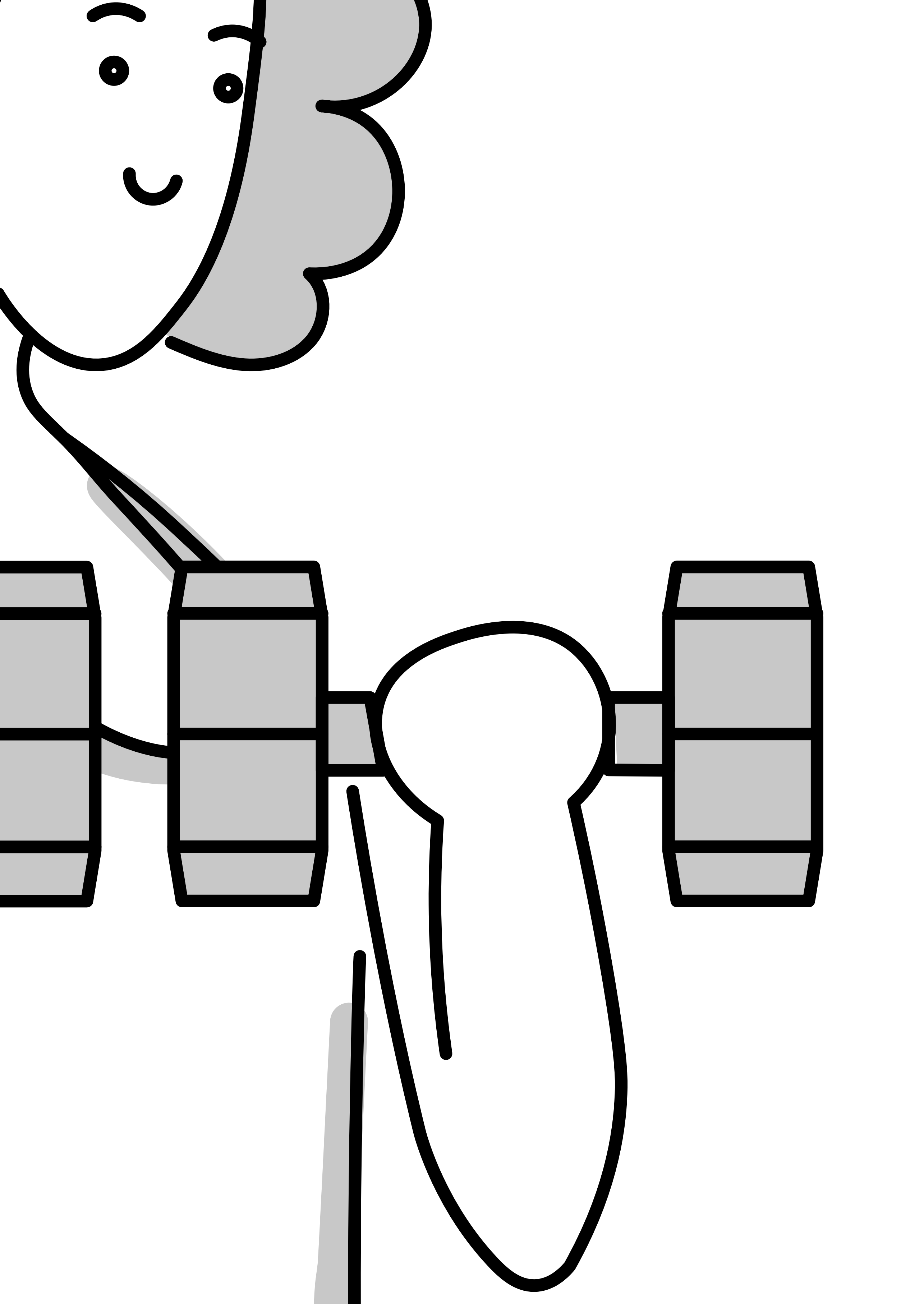

CHAPTER 6

The responsiveness of muscle strength tests in adults with intellectual disabilities

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ABSTRACT

Background: Muscle strength is both a strong predictor for future negative health outcomes, and a prerequisite for physical fitness and daily functioning of adults with ID. Therefore, it is important to be able to monitor the muscle strength of adults with ID over time. The aim of this study is to assess the responsiveness of five field tests that measure muscle strength and endurance (grip strength, hand held dynamometry of leg extension and arm flexion, 10RM-test of the seated squat and the biceps curl, 30-second Chair stand and the 5-times Chair stand) in adults with ID after a 24-week resistance-exercise training (RT) program.

Method: The responsiveness of the five muscle strength and endurance tests was assessed by correlating the change scores of the five tests with the slope of the training progression of specific exercises within the RT-program, namely the step up, seated squat, biceps curl and triceps curl.

Results: The 10RM-test of the seated squat was significantly correlated with the step up ($R=0.53$, $p=0.02$) and the seated squat ($R=0.70$ $p=0.00$). None of change scores on the other tests was significantly correlated with the training progression of the exercises.

Conclusion: The 10RM test of the seated squat could potentially be used to evaluate the effects of an RT-program in adults with ID. Responsiveness of the grip strength, hand held dynamometry, 10RM-test of the biceps curl, 30-second Chair stand and the 5-times Chair stand could not yet be confirmed.

BACKGROUND

Adults with intellectual disabilities (ID) generally have lower muscle strength and muscle endurance compared to the general population (Hilgenkamp et al., 2012, Graham and Reid, 2000, Cuesta-Vargas and Hilgenkamp, 2015). Sarcopenia (the age-related loss in muscle mass) is already highly prevalent in adults with ID aged 50-64 years old (Bastiaanse et al., 2012, Carmeli et al., 2012). Lower muscle strength has been found to be predictive for a decline in the performance of both basic and instrumental activities of daily living (ADL), for a decline in mobility and for a higher mortality risk in adults with ID (Oppewal et al., 2014, Oppewal and Hilgenkamp, 2019b). With muscle strength being both a strong predictor for future negative health outcomes, and a prerequisite for physical fitness and daily functioning of adults with ID, it is important to be able to monitor the muscle strength of adults with ID over time.

Monitoring muscle strength and muscular endurance requires exercise tests with good measurement properties, such as feasibility, reliability, validity and responsiveness. Tests normally used in the general population cannot simply be used in adults with ID, because adults with ID often have motivational, behavioural and/or physical problems, as well as cognitive limitations that hamper the execution of some tests (Riebe et al., 2018, Bossink et al., 2017). It requires the expertise of trainers to motivate the participants to exercise/work out/perform at their best, but even then it can sometimes be difficult (Weterings et al., 2020a). Previous studies with adults with ID have used different tests to measure muscle strength and muscle endurance, such as a one-repetition-maximum (estimation) test (1RM-test) (Calders et al., 2011, Dijkhuizen et al., 2018, Machek et al., 2008, Mendonca et al., 2011, Shields and Taylor, 2010), the maximal voluntary contraction test measured with a hand-held dynamotor (HHD) (Lin and Wuang, 2012), the grip strength (GS) test measured with a hand dynamometer (Hilgenkamp et al., 2012, van Schijndel-Speet et al., 2016), the 30-second chair stand test (30sCS) (Dijkhuizen et al., 2018, Hilgenkamp et al., 2012, Podgorski et al., 2004) and the 5-times chair stand test (5tCS) (used by the Healthy Athletes program of the Special Olympics)(Oppewal and Hilgenkamp, 2019a) to measure muscle endurance. All these tests have been found feasible and reliable in measuring muscle strength or endurance in adults with ID (Oppewal and Hilgenkamp, 2019a, Dijkhuizen et al., 2018, Horvat et al., 1993, Surburg et al., 1992, Dunn, 1978) and the GS and 30sCS are also predictive for a decline in mobility (Oppewal and Hilgenkamp, 2019a). However, the responsiveness of all these muscle strength and endurance tests is unknown.

Responsiveness is defined as 'the ability of an instrument or test to detect change over time in the construct to be measured' (Mokkink et al., 2010). Assessing whether an individual's status has changed over time is often the most important objective of measurements in clinical practice and research (Vet et al., 2015). If a test is not responsive, it cannot determine whether the muscle strength of a group or an individual changed over time.

Therefore, the aim of this study is to assess the responsiveness of five field tests that measure muscle strength and endurance (GS, HHD, 10RM-tests, 30sCS and 5tCS) in adults with ID. We will assess the responsiveness of these tests over a 24-week resistance-exercise training program (RT-program). It is expected that there will be a significant positive correlation between the change scores of the five field tests and the progression (change scores) on the exercises (step up, seated squat, biceps curl, triceps curl) of this 24-week resistance-exercise training program.

METHOD

Study design

This study was part of a multicentre feasibility study of vigorous resistance-exercise training for adults with ID with cardiovascular disease (CVD) risk factors (Weterings et al., 2020a), which was conducted by the 'Healthy Ageing and Intellectual Disabilities' (HA-ID) consortium. This consortium consists of three care provider organizations for people with ID in the Netherlands, Abrona (Huis ter Heide), Ipse de Bruggen (Zoetermeer) and Amarant (Tilburg), and the Chair for Intellectual Disability Medicine of the Erasmus MC, University Medical Center Rotterdam (Hilgenkamp et al., 2011)

Participants

The participants lived and/or worked in a residential or community-based setting of the participating care provider organizations. They were invited to participate in the RT-program by their nurse practitioner if they were diagnosed with at least one CVD risk factor (type-2 diabetes-mellitus, hypertension, hypercholesterolemia and/or overweight/obesity), were 18 years or older, had a mild (intelligence quotient (IQ) = 50-69) or moderate (IQ = 35-49) ID, and when a training facility was present nearby. Participants were excluded when physical problems inhibited exercising or when their physician had not provided medical clearance (Weterings et al., 2020a). This study was performed in accordance with the declaration of Helsinki (WMA, 2013) and all participants or their legal representatives provided written informed consent. The medical ethics committee of the Erasmus MC, University Medical Center Rotterdam, the Netherlands, approved this study (MEC-2016-574). All participants who finished the RT-program were included in the analysis for this study.

Overview study procedures

The participants completed a 24-week RT-program, with two training sessions a week (Weterings et al., 2020a). Each session consisted of seven exercises (step up, seating squat, abdominal curl, bridge pose, biceps curl and triceps curl), which were found feasible for adults with ID (Weterings et al., 2018). A physiotherapist or physical activity instructor with experience with working with adults with ID supervised all sessions. The RT-program consisted of five training intensity phases. The training intensity was defined by the percentage of 1RM (%1RM). The 1RM is the maximum amount of weight that a person can possibly lift for one repetition over the whole range of motion (Riebe et al., 2018). The RT-program started with a familiarization phase at 55% of 1RM (2 series of 20 repetitions), then a training phase at 60% of 1RM (2 series of 18 repetitions), then at 70% of 1RM (3 series of 12 repetitions), 75% of 1RM (3 series of 10 repetitions) and finally a training phase at 80% of 1RM (3 series of 8 repetitions) (Weterings et al., 2020a). For each training session, the trainers logged the weight and the number of repetitions for all series of all exercises. For the step up the height of the step that was used was logged as well.

At baseline, the participants performed the GS, the HHD of the arms and legs, the 30sCS and the 5tCS. After the familiarization phase, the 10RM-test was performed for the seated squat and the biceps curl. The 10RM-tests were performed after the familiarization phase to assure safety and a good execution of the test. The duration of the familiarization phase differed for all participants and ended when the RT-exercises were performed with good posture and technique (see description below) for eight consecutive sessions (Weterings et al., 2020a). At the end of the RT-program all the strength and muscular endurance tests were repeated within a two to five day interval after the last training session. The test administrator was a physiotherapist with fifteen years of experience in working with adults with ID performed all measurements (S.W.). During all measurements the participants were verbally motivated as much as possible.

MEASUREMENTS

Participant characteristics

Age, sex, level of ID, CVD risk factors (type 2 diabetes, hypertension, dyslipidaemia, and/or overweight/obesity), and diagnosis of Down syndrome and Cerebral Palsy were collected from medical records. Body mass index (BMI) was calculated by dividing weight (measured with Seca Robusta type 813, in kilogram) by squared height (measured with Seca 216 height rod, in meter). Waist circumference was measured with a non-stretchable measurement tape over the unclothed abdomen at the narrowest point between the costal margin and iliac crest in a standing position with the arms along the body (in centimetres).

Muscle strength tests

Grip strength test

The GS was measured with a hand dynamometer (Jamar hand dynamometer, Sammons Preston Rolyan, Bolingbrook, IL). The participant was sitting in a chair with the elbow in a 90-degree angle and the hand palm in a vertical position. The test was performed three times for both hands with 1-minute rest between the attempts of the same hand. The maximum score of the six attempts was the test score (Oppewal and Hilgenkamp, 2019a).

HHD-test

The Maximal Voluntary Contraction of both the arms and legs was measured with a Handheld Dynamometer (HHD-test) (Microfet 2, Hoggan Health Industries) for both the flexion and extension movement, using the break-method (Bohannon, 1988, Burns and Spanier, 2005). The HHD measurements of the elbow flexion and extension were performed sitting behind a table with the elbow resting on a table at a 90-degree-angle. Knee flexion and extension measurements were performed in a prone position with both hands resting under the head. The knee was placed vertical in a 90-degree angle. The HHD placement was at the most distal point of the lower arm and leg (van der Ploeg et al., 1984). The HHD-test was found feasible and reliable in adults with ID (Weterings et al., 2020b). In the general population, feasibility, reliability and the sensitivity to change were also good (Bohannon, 1997, van der Ploeg et al., 1991).

10RM-test

In this study, the participants performed the 10RM-test of the seated squat (10RM-Seated squat) and the biceps curl (10RM-Biceps curl). We used the 10RM-test instead of the 1RM-test, because of the higher risk for injury with the 1RM-test in frail groups, like elderly and chronically ill people (Garber et al., 2011). The 10RM-test has been found feasible and reliable for people with high risk of CVD and health conditions in the general population (Williams et al., 2007). Additionally, the 10RM-test seemed more suited for adults with ID, because they are often not used to perform vigorous exercises (Bossink et al., 2017). The weights for the 10RM-tests were selected in consultation with the trainer, choosing the weight for which the participant was expected to be able to perform 8-12 repetitions. With that weight, the participant was asked to perform the exercise until exhaustion, with a maximum of 30 repetitions. When a participant reached 30 repetitions, he/she was asked to stop and perform the 10RM-test again with a higher weight after a resting period of at least 5 minutes. The number of performed repetitions was then used to estimate at which percentage of 1RM the test was performed (see table 1 for the percentages). The weight and the percentage of 1RM were used to calculate the score for the 10RM-test. For example, a participant performed 14 repetitions with 12 kg for the biceps curl; 14 repetitions can be compared to 70%1RM so the score for the 10RM-Biceps curl = 12 kg *(100/70) = 17.1 kg.

Table 1: The number of repetitions and presented as the percentage of 1RM (Garber et al., 2011, Jongert et al., 2004)

No. of repetitions	Percentage of 1RM
6-8	80
9-11	75
12-14	70
15- 16	65
17-19	60
20-24	55
25-30	50

1RM = one repetition maximum; no. = number

Muscle endurance tests

30sCS and 5tCS

For the 30-second chair stand test (30sCS), participants were instructed to stand up and sit down again as fast as possible in 30 seconds, without using their hands. The number of repetitions was the test score. For the 5-times chair stand test (5tCS), participants were instructed to stand up and sit down again as fast as possible for 5 times, without using their hands. The time to complete five stances was the test result. The test administrator recorded time at one-hundredth of a second with a stopwatch. In both tests the participants started sitting on a chair with the knees in a 90-degree angle and the feet on the floor (Oppewal and Hilgenkamp, 2019a).

Training progression

The responsiveness of the five muscle strength and endurance tests was assessed by correlating the change scores of the five tests with the slope of the training progression of the step up, seated squat, biceps curl and/or triceps curl. We selected these specific exercises from the total of seven exercise performed within the RT-program, because the tests measure the muscle strength and endurance of the muscle groups targeted with these specific exercises.

To determine the progression in the training sessions, we calculated the average slope. The 1RM-score of each exercise of all training session was estimated. We used the number of performed repetitions to determine the training intensity of each exercise and training session (see table 2), which was then used to calculate the 1RM-score (see below for the specific calculations for each exercise). All the 1RM-scores were used as data points to create a slope of the training progression throughout the program of each participant for each exercise.

Step up

The 1RM-score of the step-up was calculated by multiplying the total training weight (bodyweight plus added training weight in kg) with the height of the step (in m). This result was then multiplied with 100 divided by the training intensity percentage to get the 1RM-score of the step-up in Kgm. (the 1RM score = ((body weight + training weight) * height * 100) / %1RM) (Zatsiorsky et al., 2021).

Seated Squat

For the seated squat, 80% of the bodyweight is used during training (de Leva, 1996). Therefore the 1RM-score was calculated by multiplying the total weight (0.8 times bodyweight plus added training weight) with a 100 divided by the training intensity percentage to get the 1RM-score in kg. Some participants used a leg press. For them, the estimated-1RM was calculated by multiplying added training weight with a 100 divided by the training intensity percentage to get the 1RM-score in kg.

Biceps curl and Triceps curl

For both the biceps and triceps curl, the estimated-1RM was calculated by multiplying training weight (the total weight held in the left and right arm) with a 100 divided by the training intensity percentage to get the 1RM-score in kg.

Statistical analyses

The participant's characteristics were analysed with descriptive statistics for all participants who finished the RT-program. The results of the muscle strength and endurance tests were analysed with descriptive statistics and a t-test to test for differences between before and after the RT-program. The results of the 1RM-scores of each exercise of the first and last training session were analysed with descriptive statistics, a paired samples t-test to test for differences and the effect sizes (ES) were calculated with Cohen's d. ES were considered low (<0.2), moderate (≥0.2 - <0.8) or large (≥0.8) (Cohen, 1988).

To assess the responsiveness of the tests, a linear mixed model (LMM) was used to compare the slope of the 1RM-score on the exercises (training progression) of each participant with the change scores of the muscle strength and endurance tests for each participant. The assumptions of normality were checked and were not perfectly met for some data, however this was considered to not influence our model fit. The responsiveness for each test was calculated by averaging the individual correlation scores between the tests and the training progression of the exercises. We calculated the individual correlations because we anticipated that if the results were pooled, some correlations could disappear because of the large heterogeneity of the participants and their training results.

For the lower extremities, the change scores of the 30sCS, the 5tCS and the HHD-test of the legs were compared with the slope of the training progression on the step up and seated squat, from the start until the end of the RT-program. Furthermore the change scores of the 10RM-Seated squat were compared with the slope of the training progression of the step up and seated squat starting after the familiarization phase until the end of the RT-program.

For the upper extremities, the change scores of the GS and the HHD-test of the arms were compared with the slope of the training progression of the biceps curl and the triceps curl at the start and end of the RT-program. Furthermore the change scores of the 10RM-Biceps curl were compared with the training progression of the biceps curl starting after the familiarization phase until end of the RT-program.

During the training, some participants changed from a seated squat to a leg press. When this happened, we saw that the estimated-1RM of the seated squat showed a sudden drop. This did not resemble the actual training experiences of the participants and trainers. It is most likely caused by the formula used to calculate the estimated-1RM, by either overestimating the amount of weight lifted by the seated squat or by underestimating the force needed during the leg press performance. Therefore, we corrected for this sudden drop in the LMM by equalising the first 1RM-score of the leg press with the last 1RM-score of the seated squat and progressing from that point on. All LMM analyses were performed in R Studio (R Studio, Boston) and the descriptive statistics and T-tests were analysed in SPSS 25 (IBM corporation).

Table 2: Participants' characteristics.

Number of participants, n	19
Male, n (%)	10 (45.8%)
Female, n (%)	9 (54.2%)
Level of ID	
Mild, n (%)	9 (45.8%)
Moderate, n (%)	10 (54.2%)
Diagnoses	
Down syndrome, n (%)	3 (12.5%)
Cerebral Palsy (GMFCS I), n (%)	2 (9.5%)
Age (in years), mean ± SD [range]	42 ± 18 [23 - 75]
CVD risk factors	
Type-2 diabetes mellitus, n (%)	5 (29%)
Hypertension, n (%)	6 (29%)
Dyslipidaemia, n (%)	4 (20%)
Overweight/Obese, n (%)	16 (92%)
BMI mean ± SD [range]	33.9 ± 6.9 [17.4 – 44.2]
Waist circumference (in cm.), mean ± SD [range]	114 ± 14 [82 – 139]

ID = intellectual disability; GMFCS = Gross Motor Function Classification Score; SD = standard deviation; CVD = cardiovascular disease; BMI = body mass index; cm = centimetre.

RESULTS

Participant's characteristics

Nineteen participants (10 men, 9 women) out of 24 (12 men, 13 women) finished the RT- program and were included in the analyses. Nine participants had a mild ID and ten had a moderate ID. Five participants had type-2 diabetes mellitus, six had hypertension, four had dyslipidaemia and sixteen were overweight or obese (table 2).

The muscle strength tests

The results of the muscle strength tests are shown in table 3. The duration of the familiarization phase differed per participant and had an average of 22.1 (±9.5) sessions (ranged between session 10 and session 46).

Table 3: Results of the muscle strength and endurance tests at baseline and post-intervention.

Exercise Test	Baseline measurement, mean ± SD	Post-intervention measurement, mean ± SD	Change score	Effect size Cohen's d [confidence interval]	P-value
GS (in kg.)	35.0 ± 9.5	35.0 ± 11.0	0.0 ± 3.5	0.00 [-0.64 – 0.64]	1.00
HHD elbow flexion left (in N.)	215 ± 69	233 ± 80	18 ± 33	0.24 [-0.4 – 0.87]	0.04*
HHD elbow flexion right (in N.)	207 ± 74	227 ± 80	19 ± 25	0.26 [-0.38 – 0.89]	0.00**
HHD elbow extension left (in N.)	150 ± 47	154 ± 56	4 ± 25	0.08 [-0.56 – 0.71]	0.51
HHD elbow extension right (in N.)	147 ± 40	156 ± 54	10 ± 25	0.21[-0.45 – 0.82]	0.10
HHD knee flexion left (in N.)	158 ± 58	150 ± 55	-7 ± 29	-0.14 [-0.78 – 0.50]	0.30
HHD knee flexion right (in N.)	167 ± 58	156 ± 54	-11 ± 27	-0.20 [-0.83 – 0.45]	0.08
HHD knee extension left (in N.)	264 ± 110	267 ± 109	2 ± 36	0.03 [-0.61 – 0.66]	0.80
HHD knee extension right (in N.)	270 ± 107	268 ± 104	3 ± 46	-0.02 [-0.65 – 0.62]	0.78
10RM-test$_{SS}$ (1RM in kg.) after familiarization	166.5 ± 60.4-	203.2 ± 89.8	36.7 ± 73.8	0.48 [-0.18 – 1.11]	0.04*
10RM-test$_{BC}$ (1RM in kg.)$^{+}$ after familiarization	18.3 ± 6.7	23.4 ± 9.5	5.3 ± 5.9	0.62 [-0.04 – 1.26]	0.00**

Table 3: Continued

Exercise Test	Baseline measurement, mean ± SD	Post-intervention measurement, mean ± SD	Change score	Effect size Cohen's d [confidence interval]	P-value
30sCS (no.)	13.7 ± 4.8	14.7 ± 6.2	-1.1 ± 3.5	0.18 [-0.46 – 0.81]	0.17
5tCS (sec.)	11.53 ± 6.12	11.17 ± 4.88	-0.36 ± 5.89	-0.07 [-0.70 – 0.50]	0.79

*SD = standard deviation; HHD = maximal voluntary contraction measured with a hand held dynamometer; N. = Newton; RM = repetition maximum; GS = grip strength; 30sCS = thirty-second chair stand; 5tCS = five-times chair stand; no. = number of repetitions; sec. = seconds; Paired t-test: * sign p<0.05, ** sign p<0.01, +based on 18 participants; one participant could not perform the 10RM-test$_{BC}$ at the end of the program.*

The training progression of the exercises

The average 1RM-scores of the first training session and the last training session of the participants of each exercise are shown in table 4.

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Table 4: The 1RM-scores of the exercises of the RT-program at the first and last training session.

	Group average 1RM-score of first training session	Group average 1RM-score of the last training session	Progression (in %)	Effect size Cohen's d [confidence interval]	P-value
Step Up (in kgm)	35.0	51.0	35%	0.87 [-0.075 - 1.806]	.001**
Seated Squat (in kg)	172.3	195.0	13%	0.5 [-0.413 - 1.413]	0.03*
Biceps Curl (in kg)	8.0	20.0	150%	1.83 [0.754 - 2.895]	.000**
Triceps curl (in kg)	5.3	17.5	230%	2.18 [1.046 - 3.318]	.000**

*RM = repetition maximum; % = percentage; kgm = kilogram*meter; kg = kilogram; paired t-test: *sign p<0.05, ** sign p<0.01,*

Table 5: Correlations between the change scores of the muscle strength tests and the average progression of the 1RM-scores of the exercises.

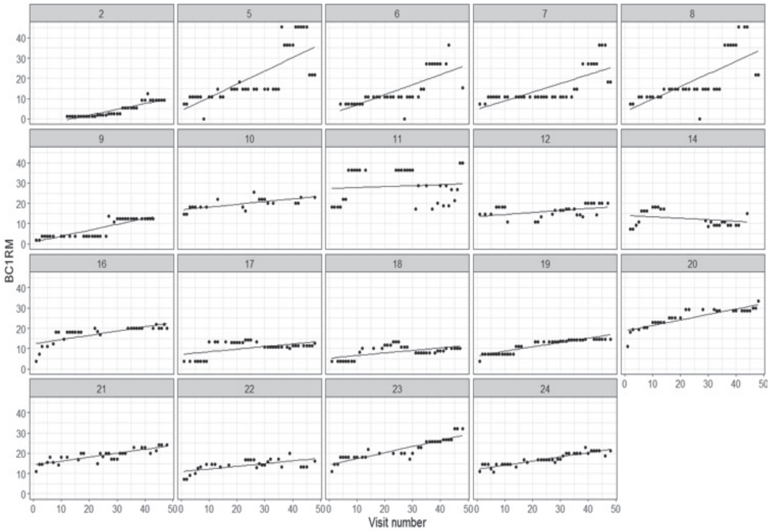
Test	Exercise	Correlation mean [conf. interval]	*P-value*.
GS	Biceps Curl	-0.35 [-0.69 – 0.13]	0.14
	Triceps Curl	0.14 [-0.33 – 0.56]	0.56
HHD elbow flexion left	Biceps Curl	-0.19 [-0.61 – 0.30]	0.44
HHD elbow flexion right	Biceps Curl	-0.11 [-0.54 – 0.36]	0.65
HHD elbow extension left	Triceps Curl	-0.05 [-0.50 – 0.41]	0.82
HHD elbow extension right	Triceps Curl	0.00 [-0.46 – 0.45]	0.99
HHD knee extension left	Step Up	-0.37 [-0.71 – 0.10]	0.12
	Seated Squat	-0.06 [-0.50 – 0.41]	0.82
HHD knee extension right	Step Up	-0.52 [-0.79 – -0.09]	0.02*
	Seated Squat	-0.06 [-0.50 – 0.40]	0.80
10RM-test$_{SS}$+	Step Up	0.53 [0.10 – 0.79]	0.02*
	Seated Squat	0.71 [0.31 – 0.88]	0.00**
10RM-test$_{BC}$+	Biceps Curl	-0.45 [-0.76 – 0.03]	0.06
30sCS	Step Up	-0.11 [-0.54 – 0.36]	0.65
	Seated Squat	0.17 [-0.30 – 0.58]	0.47
5tCS	Step Up	0.03 [-0.43 – 0.48]	0.90
	Seated Squat	0.05 [-0.41 – 0.49]	0.84

Sign = significance; 30sCS = thirty-second chair stand; 5tCS = five-times chair stand; HHD = maximum voluntary contraction measured with hand held dynamometer.
*+ After familiarization. * paired t-test: sign p<0.05, ** sign p<0.01,*

The responsiveness

The correlations between the training progression of the exercises and the change score on the tests are shown in table 5. The 10RM-Seated squat was significantly correlated with the step up (R=0.53, p=0.02) and the seated squat (R=0.70 p=0.00). Furthermore the HHD knee extension of the right leg was significant negatively correlated with the step up (R=-0.52, p=0.02) but not significant with the seated squat (p=0.80). None of change scores on the other tests were significantly correlated with the training progression on the other exercises. In figure 1 an example of the training progression of the seated squat for each participant is shown. For each participant, the 1RM-score of each training session is plotted; the line represents the average slope of training progression of the whole RT-program. This slope is correlated with the change score of the tests for each participant.

Figure 1: The estimated-1RM of each training session of each participant of the biceps curl with the average slope of the training progression



This example is representative for the other exercises.

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DISCUSSION

For this study we assessed the responsiveness of the GS, HHD-test, 10RM-tests, 30sCS and 5tCS by correlating the changes in performance on these tests with the slope of the progression of the exercises of a 24-week RT-program in adults with ID. In this study only the 10RM-Seated squat seems to be a responsive test to measure the progress in muscle strength over the RT-program.

All four exercises within the RT-program showed a significant progression, with 150% and 230% for the biceps and triceps curl respectively (effect sizes of 1.83 and 2.13). The progression of the step up (35%) and seated squat (13%) were smaller (effect sizes of 0.87 and 0.5). These effect sizes are larger than the effect size of 0.26 Shields et al. (2008) found for the leg press after a 10-week RT-program for adults with DS (Shields et al., 2008). A study by Calders et al (2011) showed a similar progression of 33% for the lower body strength in adults with ID after a 20-week RT-program (Calders et al., 2011).

The 10RM-Seated squat (change score of 36.7 kg) and the 10RM-Biceps curl (change score of 5.3 kg.) showed a significant improvement. Another study in young adults with ID by Machek et al. (2008) found an increase in the predicted-1RM of seated dip of 53.74 kg and an increase of 25.6 kg for the biceps curl after a 12-week RT-program (Machek et al., 2008). Furthermore, we found a significant improvement in the HHD elbow flexion test (both left and right) with change scores of 18 and 19 N.. The HHD knee extension showed no significant improvement. In contrast, a study by Lin et al (2012) found a significant progression in the HHD knee extension test with a change score of 3.42 Pounds after a 6-week RT-program in adolescents with Down syndrome (Lin and Wuang, 2012). The scores of the other HHD tests, the GS, 30sCS and 5tCS did not change in our study. Calders et al. (2011) did find significant improvements for the 30sCS and GS after a 20-week RT-program in adults with a mild ID (Calders et al., 2011).

We hypothesized that the heterogeneity of the participants in our study might impair finding significant results. For example, using the average of the correlations might eliminate differences between responders and non-responders of the RT-program. Therefore, we used the individual correlations of each participant instead and calculated the average of those individual correlations. Nevertheless for most tests we found non-significant low correlations. There are very few articles on the responsiveness of muscle strength tests in general, so it is difficult to compare the results of this study with other literature, than the previous-mentioned intervention studies. These studies mostly mention the progression of their RT-program or mention the results on the tests, but no study mentions the correlation between the results of the RT-program and the results of the tests. We therefore have to hypothesize what could be the reason why the responsiveness of the tests in this study showed mostly low non-significant correlations. There are some potential reasons for this lack of responsiveness.

First, it could be that most of these tests are not so responsive to measure changes in muscle strength and therefore less suited to be used to evaluate the progression of muscle strength within an RT-program. Responsiveness studies focus on the agreement between change scores, in our case two measurements of the tests and the slope of the exercises, with its own measurement error (often indicated by the MDC of a test) for each measurement or calculation. The MDC of a single score is large for the GS (6.5-8 kg. (Kim et al., 2014, Blankevoort et al., 2013), the HHD-test (10-17N.) (Buckinx et al., 2017) and the chair stand (2 repetitions) (Hesseberg et al., 2015) in the general (older) population. These large MDC's lower the potential correlations for their responsiveness, as explained above. It could, therefore, be difficult for these tests to show progression after the RT-program, despite the significant increase in 1RM-scores in the training program. In this study, only the responsiveness of the 10RM-Seated squat was significantly correlated with the step up and seated squat. The correlations of the 10RM-Seated squat were 0.5 for the step up and 0.7 for the seated squat. There are, to date, no guidelines on what is an acceptable correlation for responsiveness (Vet et al., 2015). Normally 0.7 is the minimum correlation to be acceptable, but in responsiveness studies lower scores are often found (Vet et al., 2015).

Second, it could also be argued that the GS, HHD, 30sCS and 5tCS tests are not suited to measure the increased muscle strength, because these tests measure different aspects of muscle performance. The GS measures strength in the hands and that is not trained specifically. The 30sCS and 5tCS measure muscle endurance. These two tests are highly dependent of the speed with which the test is performed It could be speculated that though the leg muscles were potentially getting stronger, it was still difficult for adults with ID to speed up the sitting and standing, because speed requires another type of muscle control, coordination and cognitive attention, which can be difficult for adults with ID (Riebe et al., 2018). The HHD measures isometric muscle strength of the arms and legs in a 90-degree angle, but the exercises of the RT-program are performed over a full range of motion of the muscles. Training and measuring muscle strength is dependent on the angle in which it is performed and trained (Riebe et al., 2018). Only the 10RM-tests are performed exactly like some of the exercises of the RT-program. They are the only ones that partly show a correlation, even though they were not performed at the start of the program.

Limitations

This study was performed within a feasibility study regarding the feasibility of a progressive RT-program in adults with ID with CVD risk factors. This study sample is not a representative sample of the whole, diverse population of adults with ID and therefore more research into the responsiveness of muscle strength tests is necessary in adults with ID. With 19 participants, this study included just a small heterogeneous sample of adults with ID with at least one CVD risk factor. The heterogeneous sample is preferred for feasibility testing, as it reflects the differences of the adults with ID in daily life, but a heterogeneous sample lowers the internal validity of a study. So, the results of this study should be interpreted with caution.

Furthermore, the modelling of the training progression by the 1RM-scores of the exercises in RT-program could have impeded the actual progression of the participants' muscle strength. The training intensity differed during the RT-program for most participants and exercises and even exercise execution differed between participants and even within the RT-program of individual participants. All these different factors made it more complex to model the training results into a standardised 1RM-score. Furthermore, this modelling was based on assumptions when calculating the 1RM-scores, and this could have impeded with the true training progression of the participants in the RT-program.

Important factors that could influence both the testing results and the progression during the RT-program were the motivational, behavioural and/or physical problems, and cognitive limitations of the participants (Bossink et al., 2017, Riebe et al., 2018). It requires the expertise of the trainers to motivate the participants to train at their best, but even then it is hard to interpret if they actually performed the exercises and tests as best as they could. For example, sometimes a participant stopped training after one or two lifts after a training weight was increased (always in small steps), stating this was way too heavy to lift, where the participant easily performed the required 10-20 repetitions the series before. This could also be a problem in the general population, but even more so for adults with ID.

Recommendations

There is a need for more uniform measurements with good measurement properties (Robertson et al., 2017). This is the first study into the responsiveness of muscle strength tests in adults with ID. More studies are necessary to find the appropriate muscle strength tests to monitor changes in muscle strength of adults with ID. The ID-fitscan is a first attempt to get uniformity in fitness tests for adults with ID, which have been found reliable and valid (Oppewal and Hilgenkamp, 2019a). However, the responsiveness of the muscle strength and endurance tests used in the ID-fitscan (GS, 30sCS and 5tCS) is still questionable, which could make them unfit to evaluate RT-programs in adults with ID. Only the responsiveness of the 10RM-Seated squat showed a significant correlation of 0.53 and 0.70 and with the step up and seated squat. Future research should also study potential individual factors influencing the responsiveness of the tests, as there are large differences between participants in the test results and the change scores of the RT-program as shown in figure 1. Furthermore, more research is needed into the floor and ceiling effects and smallest detectable change, which have still never been investigated, and are necessary to interpret the results of muscle strength testing.

Conclusion

The 10RM-Seated squat could potentially be used to evaluate the effects of a RT-program in adults with ID with CVD risk factors. Furthermore, it is still questionable that the GS, HHD-test, 10RM-Biceps curl, 30sCS and 5tCS could be used to evaluate the effects of a RT-program in adults with ID. Interestingly, the 1RM-scores of all four exercises, both the 10RM tests and the HHD-test of the elbow flexion were all significantly improved. This stresses the need for more research into the interpretation of the results of RT-programs and the way muscle strength can be measured in adults with ID.

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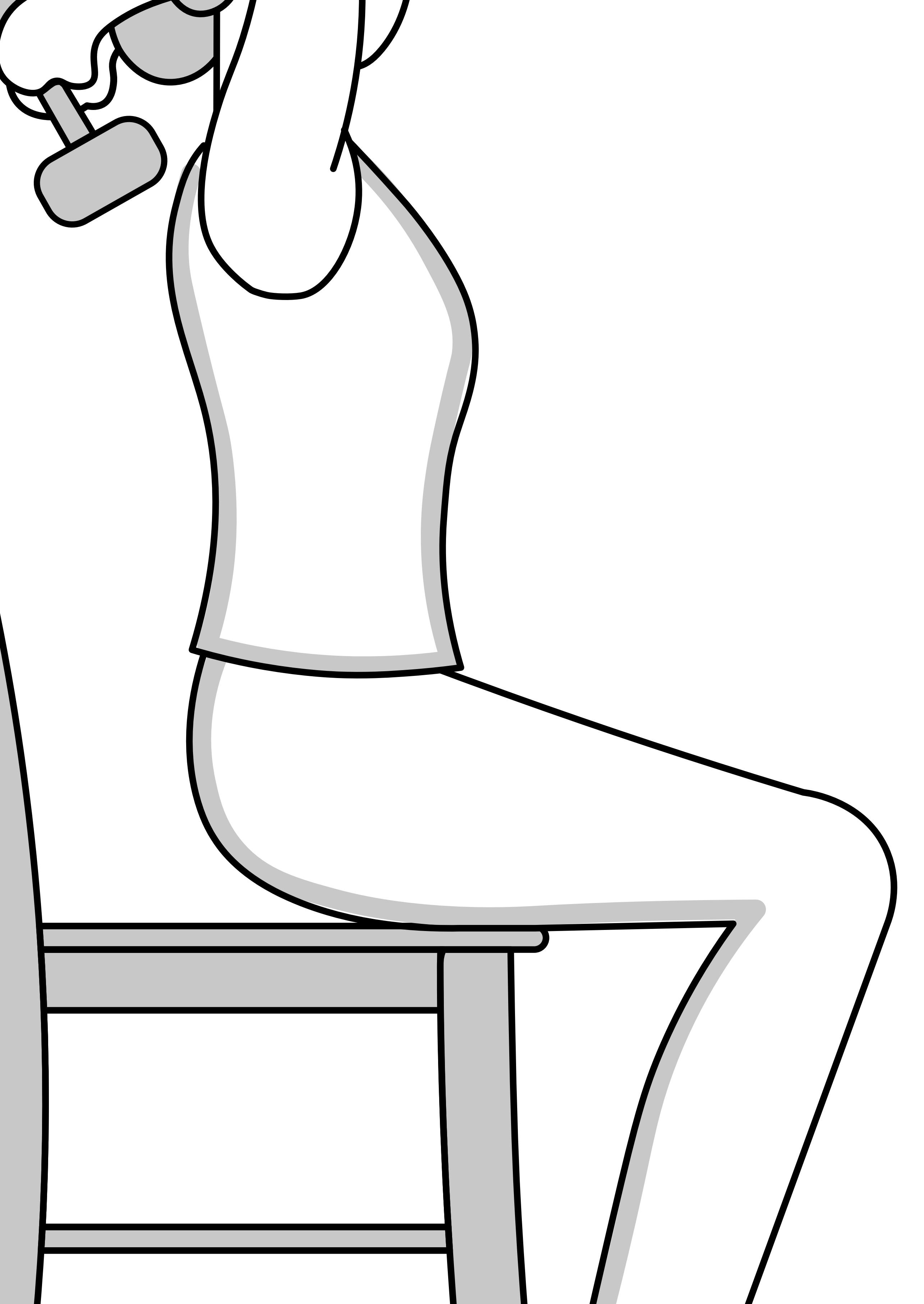

CHAPTER 7

General Discussion

GENERAL DISCUSSION

PRINCIPAL FINDINGS

This thesis showed that it is feasible for adults with mild and moderate ID with cardiovascular disease (CVD) risk factors to perform a 24-week progressive resistance exercise-training (RT) program at vigorous intensity. This is a first step in developing a non-pharmaceutical intervention for adults with ID with CVD risk factors (type-2 diabetes mellitus, hypertension, dyslipidaemia and being overweight/obese). This RT-program was developed in accordance with the most recent guidelines on RT for people with CVD risk factors in the general population (Williams et al. 2007, Colberg et al. 2010, Piepoli et al. 2016, American College of Sports Medicine 2022) and adapted to the needs and possibilities of adults with ID.

First, we developed the Resistance Exercise Set for adults with ID (RESID) together with experienced physiotherapists and physical activity instructors. The RESID consists of seven resistance exercises focussing on the total body without the use of weight machines (Chapter 2). Second, we asked these experts which facilitators they consider to be important for a successful delivery of an RT-program for adults with ID. These facilitators were: close monitoring during training; training sessions with a maximum of 60 minutes, no more than two sessions a week, maximal external motivation (from caregivers and/or family), fun during training, positive reinforcement and the use of rewards (Chapter 2). Third, based on the RESID and facilitators we developed a 24-week progressive RT-program to train the whole body at vigorous intensity, which we then tested for feasibility in 24 participants with ID. Nineteen participants of three ID care provider organizations finished the RT-program. Eleven of these 19 (58%) participants trained at vigorous intensity at the end of the program. Based on these results we concluded that the RT-program is feasible (Chapter 3).

This thesis also focused on the psychometric properties of physical fitness tests for adults with ID. We found that it is feasible to perform a supramaximal exercise test (SET) after a graded maximal treadmill test (GXTT) to measure the cardiorespiratory fitness in adults with ID. Nine participants out of twelve (75%) performed the complete measurement procedure. We found that for most participants the VO_{2peak} of the GXTT was confirmed (ICC=0.99) by the SET as a true VO_{2max} if the secondary criteria were met (Chapter 4). Looking at muscle strength measures, we also found that it is feasible and reliable to measure muscle strength with a handheld dynamometer (HHD). We found excellent reliability (ICC > 0.9). However, we found the standard error of measurement (SEM) to be rather large (15.2 – 21.1 N.) (Chapter 5). Finally, we studied the responsiveness (the ability to measure changes over time) of the muscle strength and muscle endurance tests used in the RT-program. The seated squat-10RM test showed moderate responsiveness with the progression during the RT-program on the seated squat exercise (R=0.70 p=0.00) and could be used to evaluate changes in muscle strength within an RT-program. The grip strength, the 30-second chair stand, the 5-times chair stand, the HHD and the arm-flexion-10RM-test did not show any change over time, even though we did see a progression on the execution of the exercises in the RT-program for these muscle groups. These tests are therefore questionable to be used to evaluate changes in muscle strength within an RT-program (Chapter 6).

METHODOLOGICAL CONSIDERATIONS

Testing issues

It was difficult to establish the responsiveness of the fitness tests used in this thesis. We had to make some assumptions in calculating the progression of the RT-program. The results of each training session were expressed as a 1RM-score for which training weight and intensity were used to calculate the 1RM. In some exercises, the total training weight consisted of a part of one's own body weight and additional training weights, like for the seated squat and the step up. For these exercises, we had to make assumptions on the percentage of body weight we needed to use to calculate the total training weight. Although there are no clear guidelines for these calculations, we used the available research for these calculations (Zatsiorsky et al. 2021, de Leva 1996). These assumptions could have impeded with the actual training progression made by the participants, which could have impacted the comparison of the results of the fitness tests with the progression of the participants on the exercises of the RT-program. Future research should find clear guidelines how to calculate training progression, especially when the body weight is being used during exercising as part of the training weight. These guidelines could help to understand the relation between exercise progression and progression in fitness test and could aid in finding responsive fitness tests for RT-programs.

Bias

For a feasibility study, only a rather small group of participants is needed, as long as the total target population is represented in the study. So, for our study, all adults, of both sexes, with both mild and moderate ID, of all age groups should be represented in the study, as well as a representation of all CVD risk factors to have a good representative study sample. The characteristics of the participants in this thesis represented this variation well. However, that variation, together with the limited number of participants, creates challenges for the validity of the results of this thesis for the specific subgroups, as well as for generalizability to adults with other levels of ID.

A problem with almost all intervention studies is that only participants who are motivated for the intervention will participate, creating a bias for the target population to which you want to generalize the results. This is even more so for studies regarding exercise programs, because people's motivation to start exercising will have a major impact on their decision to participate. To target only the potential participants for our study, they were invited to participate when visiting their local nurse practitioner to participate, and we provided them with detailed and easy-read information. Furthermore, it is important to keep participants motivated during exercising. Therefore, we also focused on finding the most feasible workout and asked experts in a focus group for motivational factors to motivate the participants during exercising.

CLINICAL IMPLICATIONS

The RESID

The RESID is a feasible resistance training exercise set for adults with ID and provides a full-body workout. It has the potential to be used in different training settings and does not require expensive weight machines. The RESID is a core set of exercises that can be tailored to the physical and cognitive capabilities of the participant. Therefore, we recommend the RESID for use by physiotherapists and physical activity instructors in an exercise program for almost all adults with ID (see Appendix I).

Exercise execution

The guidelines on CVD exercise programs in the general population show that the best results are achieved when exercising at vigorous intensity (American College of Sports Medicine 2022), which is a challenge for novice trainees. Furthermore, the guidelines state that novice trainees should not start at vigorous levels of intensity to prevent injuries (American College of Sports Medicine 2022). Because our participants were not familiar with RT, we had to start at a low intensity, and slowly increase the intensity of the exercises to vigorous. This progressive increase in intensity made the RT-program complex for the trainers, with a familiarization phase and four intensity phases, decreasing repetition and increasing weights at the beginning of each new phase, while increasing weights within each phase as well, when the intended number of repetitions was met. Furthermore, for this thesis we relied on the expert opinion of the trainers when to increase the intensity. The training intensity was only increased if the trainers were confident it was safe to do so. This made the RT-program complex for both trainees and trainers, which could have hampered the training progression, and feasibility of the study. The physiotherapists and physical activity instructors should be more aware of the progression difficulties for adults with ID and try to find ways to overcome these difficulties during exercising In daily practice increasing the results of exercising on physical fitness in adults with ID.

Previous studies on RT-programs did not report the actual training intensity of the training sessions or did not report the intensity that they aimed to train at. Most used a fixed number of sets and repetitions (Shields et al. 2008, Tamse et al. 2010) or decreased the number of repetitions after a certain number of sessions (Suomi 1998, Calders et al. 2011, Fornieles et al. 2014). Furthermore, previous studies about RT training included healthy participants, adults with DS or Special Olympic athletes. The participants in our study were novice trainees with ID with at least one CVD risk factor (type-2 diabetes mellitus, hypertension, dyslipidaemia and being overweight or obese).

Future studies should find ways to provide more guidance for the trainers throughout the RT-program with a more standardized RT-program, because guiding the adults with ID during exercising requires full attention. Furthermore, future studies should find (digital) ways, like for instance an app to calculate the progression in weights automatically, to assist trainers with the increase of the training weights during exercising of the participants. These efforts make the execution of the program more standardised or repeatable and make sure that there can be a more optimal training dose during the RT-program.

Professional guidance is necessary

Close monitoring of the participants was necessary to exercise throughout the RT-program. The trainer had to be physically next to the trainee during exercising to make sure the exercises were performed with the right posture, technique and dosage, as other studies on RT in adults with ID showed as well (Shields et al. 2008, Calders et al. 2011, Dijkhuizen et al. 2018). The physiotherapists or physical activity instructors specialised in adults with ID have the necessary knowledge and skills to motivate participants to perform the exercises in the RT-program, and assure safety. These trainers are specialised in dealing with the motor control issues and motivation problems of adults with ID and can adapt the RT-program to the possibilities of the participants while still adhering to the structure of the program. The individualised step-by-step positive approach helped to motivate the adults with ID through experiencing successes during training.

Motivation from caregivers and family is key

Researchers, physiotherapists and physical activity instructors should involve caregivers and family from the beginning when setting up exercise programs and/or health programs to increase their involvement and positive attitude towards the exercise program. In our study the caregivers received an information letter with all the information about the RT-program and that was enough to include the intended number of participants in our study. Still, during the inclusion period of the study we noticed some reluctance from caregivers and family of the participants to let the adults with ID join the RT-program, for instance because they thought it would be too complex for a participant, or they were afraid the participant with behavioural problems would get to strong for them to handle. This reluctance may hamper inclusion when a higher number of participants is needed in future studies. Reluctance of caregivers is a known barrier for adults with ID to participate in exercise (Bossink et al. 2017). Future studies should invest time in discussing the worries, reluctance or questions of caregivers and family that might prevent them from providing full support for adults with ID to go exercising. Furthermore, it is necessary to invest in informing the caregivers and family of the importance of exercising and their important role in providing positive support for adults with ID before the start of the RT-program.

The psychometrics of physical fitness testing

It is important to study the psychometric properties of a test before using the test to be sure you measure what you want to measure. In this thesis we provided some insight in the psychometrics of a number of tests for adults with ID. First, there is still a lack of a valid test to measure the cardiorespiratory fitness of adults with ID. Our study showed it is feasible to use a GXTT with an SET and it can be used as ‘gold standard’ in the search of a more practical submaximal test in adults with ID or in validation studies of currently used tests like the 6-minute-walk-test, the 2-minute-step-test or the shuttle-walk-test. As for now there are many questions whether these tests actually measure the aerobic capacity of adults with ID. Second, the HHD is an easy and reliable way of measuring muscle strength in adults with ID, which can be used in daily practice as long as a strict protocol will be followed how to measure the muscle strength. Future studies should investigate ways to lower the SEM, which would make it more suited to measure small changes in muscle strength. Furthermore, the HHD can be used for measuring the strength of many muscles in the body, not just the flexion and extension of the arms and legs, but also in shoulder and hip muscles. Future studies should investigate the feasibility and reliability of measurements for other muscles. This could help physiotherapists in their daily practice, measuring the muscle strength of specific muscles in adults with ID. Third, only the 10RM test of the legs showed moderate responsiveness and seems to be the best option to measure change in muscle strength within an RT-program.

DIRECTIONS FOR FUTURE RESEARCH

CVD risk factors

The next step for future research is to study the health effects of this RT-program on the CVD risk factors, type-2 diabetes mellitus, overweight/obesity, hypertension and dyslipidaemia in adults with ID. Furthermore, the effects of the RT-program on muscle mass need to be determined, as muscle mass is an important factor to increase rest metabolism, which helps in lowering CVD risk and getting healthier. As the support of caregivers is important for adults with ID to exercise, future research should involve caregivers and family more before the start and during the RT-program, to improve inclusion to the study and improve adherence to the RT-program. It is important to explain why resistance exercise training could benefit the health of the adults with ID and discuss any worries, reluctance or questions (Bossink et al. 2017). Furthermore, the effect of the RT-program on prevention of developing CVD risk factors for adults with ID should be studied in the future, as to prevent adults with ID developing (pre-) diabetes and/or (pre-)hypertension and/or dyslipidaemia.

Other health outcomes

Future research should also study the effect of the RT-program on other health outcomes than CVD risk. An RT-program could have positive effects on physical activity levels and on the ability to perform activities in daily life (ADL) and quality of life in adults with ID. Furthermore, it is interesting for future research to study the effects of the RT-program on the prevention of sarcopenia in older adults with ID. There are more health outcomes that could be improved as well, such as arthrosis, osteoporosis and sleep. Moreover, there are studies that indicate that being physically active and exercise can positively influence challenging behaviour in adults with ID (Ogg-Groenendaal et al. 2014). It would be interesting to study the effects of an RT-program on behavioural problems in adults with ID in future research. Especially because adults with ID have three to five times more often behavioural problems (Ogg-Groenendaal et al. 2014), which is a major burden for their relatives, friends, and caregivers (Ogg-Groenendaal et al. 2014). Improving these health and behavioural outcomes could also motivate caregivers to support adults with ID to participate in an RT-program. The important next step is to study the effectiveness of this program in reducing CVD risk and other health outcomes.

Cost-effectiveness

From a financial perspective, there are a number of considerations when implementing the RT-program. The RT-program can only be performed with close monitoring of physiotherapists or activity instructors, which has to be funded by the care providers. However, the potential health benefits, such as reduced CVD risk, less sarcopenia, improved independence in daily activities, and a reduction of behavioural problems, could potentially cause a reduction in medicine usage, medicine costs, and lower the amount of care and support needed from caregivers in daily life. These costs and benefits have to be determined and calculated by studying the cost-effectiveness and the Quality Adjusted Life Years (QALY's). The QALY is used in health economic evaluations to quantify the health effect of a medical intervention or a prevention program and ultimately to help payers, like care providers, allocate healthcare resources. Therefore, it would be interesting for future research to study the cost-effectiveness and QALY's of the RT-program within the healthcare system for adults with ID.

Exercise programs to benefit all adults with ID

For this study into the development of an RT-program, adults with severe and profound ID were excluded, as the participants had to be able to follow the instructions for the exercises by the trainer. There are already some studies into exercise programs to increase muscle strength of people with severe and profound ID and visual impairments (Bossink et al. 2017, Dijkhuizen et al. 2019), but future research should also focus on developing specific exercise programs to increase physical fitness and health of all people with ID targeting their specific health issues and physical capabilities.

Furthermore, we only included participants who were able to perform all the exercises, excluding adults with ID who had difficulty standing and walking or were in a wheelchair. This was based on the ACSM and other guidelines stating that all large muscle groups need to be trained in order to have an effect on the CVD risk factors for adults in the general population. It is also important for future research to focus on developing an RT-program for all adults with ID in wheelchairs or other motor problems, like standing and walking difficulties.

Measuring and monitoring physical fitness

Fitness tests like the ID-fitscan, the HHD and 10RM tests could help monitor the physical fitness of adults with ID. Physical fitness has been shown to be strong predictor of a range of health issues, many of which are highly prevalent in adults with ID (de Winter et al. 2012, American College of Sports Medicine 2022, Oppewal et al. 2018, Hilgenkamp et al. 2012). All these health problems impede with their ADL abilities, quality of life and their ability to engage in society. Future research should develop norm scores and cut-off points to detect adults with ID at risk for developing health problems. Adults with ID could then be screened and monitored by health professionals in daily practice to detect potential health problems. When the cut-off points show the adults with ID are at risk of developing health problems, the health-professionals can then discuss taking preventative actions with the adults with ID and their caregivers to prevent the health problems.

The challenge is to stay physically active

Over the last 15 years, we gained much knowledge on the health risks of people with ID, the high risk of CVD, the high levels of polypharmacy, and the very low levels of physical fitness (Bastiaanse et al. 2012, de Winter et al. 2012, Hilgenkamp et al. 2012, Cooper et al. 2015, Schoufour et al. 2018). Increasing PA is advised in the new international consensus guidelines in the Management of type-2 diabetes mellitus in adults with ID (Taggart et al. 2021). Many healthcare professionals and professional caregivers try to improve the PA levels of people with ID, but on many fronts the problems are still there. This thesis is a first step towards finding an exercise program to improve PA levels, through exercising, and health for adults with ID with CVD risk factors, and could potentially be used as an alternative for medications. If this intervention is proven effective, there is the challenge of sustaining PA levels and improved health, after the intervention. Many studies show, that interventions only work as long as the interventions are in place (van Brussel et al. 2011). Adults with ID should find a routine in daily life that is in line with the PA guidelines. Their average daily activities are very often below the PA guidelines, which makes it almost necessary for them to keep on exercising after the intervention (Kuijken et al. 2020). Future studies should focus on ways for adults with ID to sustain their improved physical activity and keep on exercising in their daily life after an intervention.

A healthy lifestyle in daily life

It is important to realise that a healthy lifestyle is more than just improving PA levels. Health is a state of physical, mental and social well-being (WHO 1948). This means besides getting regular exercise, eating a balanced diet, avoiding tobacco and drugs and getting plenty of rest, and be included in society is part of a healthy lifestyle. Daily caregivers are identified as the most important and influential stakeholders to implement all these healthy lifestyle components in daily support of adults with ID, but they are not properly facilitated to support a healthy lifestyle (Kuijken et al. 2019). Recently there have been some studies into how to facilitate professional care givers and create a supportive environment for a healthy lifestyle of adults with ID (Steenbergen et al. 2017, Kuijken et al. 2020, Vlot-van Anrooij et al. 2020). They showed that there is often a lack of a shared vision and a policy in which all health professionals and caregivers know their roles and responsibilities (Steenbergen et al. 2017, Kuijken et al. 2019, Vlot-van Anrooij et al. 2020, Vlot-van Anrooij et al. 2020). A healthy lifestyle should be promoted by everybody within the care organisations and be part of their mission and vision (Steenbergen et al. 2017, Vlot-van Anrooij et al. 2020). This also means that a multidisciplinary approach should be incorporated where general practitioners, physicians specialised in ID medicine, physiotherapists, nurse practitioners, dieticians, behavioural therapists and caregivers together with the adults with ID themselves try to promote their health.

Implementation is key

There is a gap in knowledge in the best way to implement a healthy lifestyle and increase PA. Implementing new insights, treatments, strategies or even de-implementing non-working treatments is a profession in itself. It could for example, be helpful if healthcare professionals and professional caregivers could have time to reflect on their own work and work habits and discuss alternatives, preferably with the aid of a specialist. Furthermore, future studies should study the best ways to implement all existing knowledge, not just as a follow up in an intervention study, but as a whole new implementation study.

CONCLUSION

This thesis showed that it is feasible for adults with ID with CVD risk factors to perform a vigorous RT-program. Furthermore, we showed that there are feasible, reliable and responsive muscle strength tests, which can be used to measure and monitor muscle strength. These results support physiotherapists and physical activity instructors to train adults with ID and monitor the muscle strength, and we introduced an RT-program that potentially could help adults with ID to get healthier. This thesis is the first step towards an evidence-based exercise intervention to reduce CVD risk for adults with a mild or moderate ID with CVD risk factors. It provides a solid starting point for future research into improving the health of adults with ID. Future studies should not just focus on the unanswered questions of the potential to improve CVD risk with an RT-program, but also on improving sarcopenia, activities in daily live, fall risk and even behavioural problems. The exercise set and RT-program give physiotherapists and other movement specialists new options in their efforts to support adults with ID into healthy aging.

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SUMMARY

SUMMARY

CHAPTER 1
General Introduction

People with intellectual disabilities (ID) generally have more health problems than the general population. Multiple studies show for example that the prevalence of cardiovascular disease (CVD) risk factors, such as type-2 diabetes mellitus, hypertension, dyslipidaemia and being overweight/obesity, is high in (older) adults with ID. The standard therapy for people with ID when diagnosed with CVD risk factors is still providing medication. However, in the general population increased physical activity (PA) and exercise are found to improve CVD risk factors. Increasing PA levels and exercising can reduce or even eliminate the need for medication to improve CVD risk factors. However, the effect of PA or exercise on CVD risk factors has not yet been widely studied in adults with ID.

Resistance-exercise training (RT) is a form of PA. Clear guidelines for RT to improve cardiovascular health exist for the general population. There is also a clear dose response relationship: more exercise sessions improve the cardiovascular health more, and a vigorous RT-program improves cardiovascular health more than a moderate RT-program. RT also has potential beneficial effects on reducing sarcopenia and may improve the ability to perform daily activities. RT could be feasible for adults with ID, because it requires short bouts of exercise of a limited number of repetitions followed by a resting period. However, it is not known if a vigorous RT-program is feasible for adults with ID. Therefore, the aim of this thesis is to study if individuals with ID with CVD risk factors can perform a RT-program at vigorous intensity. The second aim of this thesis is to find feasible, reliable and responsive fitness tests to be able to measure and evaluate the physical fitness of adults with ID.

CHAPTER 2
A pilot study on a resistance exercise set for a total body workout

The American College of Sports Medicine (ACSM) recommends the use of weight machines during training, because they offer a guided execution of a movement. However, in daily practice, many adults with ID are less likely to have access to training facilities with machines. Therefore, the focus of this study was to design and to determine the feasibility of a Resistance Exercise Set for a total body workout for adults with mild or moderate ID (RESID), without the use of weight machines, and which can be tailored to their physical capabilities. In an expert meeting with seven physiotherapists and physical activity instructors, potential exercises were discussed to come to a final selection of seven exercises for a total body workout, the RESID, with alternative exercises for tailoring. To test the feasibility of the RESID, 11 participants (5 women, 10 mild ID, 1 moderate ID, mean age 29 ± 8 years [range 19-44]) performed the RESID twice during regular sport classes. Feasibility was defined by the amount of exercises completed and the correct execution of the exercises. The completion (90-100%) and execution (90-100%) were excellent for all exercises. Therefore, the RESID was found

to be a feasible resistance training exercise set for adults with mild or moderate ID for use in daily practice. A physiotherapist, physical activity instructor or fitness instructor needs to be available to closely supervise the execution of the exercises and adapt the exercises when necessary.

CHAPTER 3
The feasibility of vigorous resistance exercise training program

In chapter 3 we studied the feasibility of a progressive RT-program, with increases in intensity from novice to vigorous intensity, for adults with ID with CVD risk factors. Feasibility was defined by achieving vigorous training intensity at the end of the RT-program. Twenty-four adults with ID (13 women, 11 mild ID, 13 moderate ID, mean age 44 ± 17 years [range 23 – 75]) with at least one CVD risk factor (type-2 diabetes mellitus, hypertension, dyslipidaemia and being overweight/obesity) participated in the 24-week RT-program. All CVD risk factors were represented in this study. The training intensity was gradually increased from novice (50%1RM) to vigorous (75%–80%1RM). Participants moved to the next phase of training intensity when at least five out of the seven exercises were performed with good posture and breathing technique during eight training sessions. Nineteen participants completed the RT-program, with an overall attendance of 73%. The feasibility was good as 58% (11 out of 19) of the participants exercised at vigorous intensity at the end of the program. In a questionnaire, the trainers responded that close supervision of the trainers was necessary throughout the RT-program to ensure good posture and breathing technique. This study showed that it was feasible to perform an RT-program at vigorous intensity for adults ID with CVD risk factors. Physiotherapists, physical activity instructors or fitness instructors, with experience in working with people with ID, can use this RT-program to train at vigorous intensity with adults with ID in daily practice, yet close supervision remains necessary during exercising.

CHAPTER 4
Feasibility of Supramaximal Verification after a Graded Maximal Treadmill Test

Cardiorespiratory fitness (CRF), a component of physical fitness, has been identified as a major indicator for overall fitness and an important predictor of future health. However, strong evidence is lacking on the validity of maximal and submaximal cardiorespiratory exercise tests in adults with ID. In chapter 4, we therefore studied the feasibility of a graded maximal treadmill test (GXTT) followed by a supramaximal exercise test (SET) to measure the maximal CRF, expressed as maximal oxygen uptake (VO_{2max}), in adults with a mild or moderate ID. The SET is used to check if the peak oxygen uptake measured during the GXTT (VO_{2peak}) was a true VO_{2max} measurement. Twelve participants (3 women, 4 mild ID, 8 moderate ID, mean age 30 ± 8 years [range 20 - 45])) performed the measurement procedure. Nine out of 12 participants completed the measurement procedure (75%), which we defined as excellent. The ICCagreement of the normalized VO_{2peak} of the GXTT and normalized VO_{2peak} of the SET was 0.99 ($P < .001$). A custom made questionnaire showed that the measuring procedure was acceptable for the participants. Ten participants (83%) would perform the test another time, but also eight participants (67%) responded they did not like wearing the mask during testing. This study demonstrated that it is feasible to perform a GXTT followed by a SET for adults with ID. Therefore, it can be possible to verify the VO_{2peak} of the GXTT as a true VO_{2max} in adults with ID.

CHAPTER 5
The feasibility and reliability of hand held dynamometry

Muscle strength is both a strong predictor for future negative health outcomes, and a prerequisite for physical fitness and daily functioning of adults with ID. A potential feasible way to measure muscle strength in daily practice may be with a hand-held dynamometer (HHD). Therefore, the aim of this study was to determine the feasibility and reliability of HHD strength measurement in adults with ID. In this study 21 (11 women, 10 mild ID, 11 moderate ID, mean age 44 ± 17 years [range 23-75]) out of the 24 participants who started the RT-program performed the HHD-measurements twice for arm flexion and leg extension, with both the left and right side. Feasibility was defined as performing the measurements according to the measurement protocol. The test-retest reliability was calculated with the intraclass correlation coefficient (ICCagreement), the standard error of measurement (SEM) and the minimal detectable change (MDC). The feasibility of the HHD measurements was good to excellent for elbow flexion (66.7% - 81.0%) and moderate to good (42.9% - 72.4%) for knee extension for adults with ID. The reliability was excellent with all ICC's ≥0.90. The SEM's were between 15.2 and 28.1 Newton (N.) resulting in MDC's between 45.2 and 53.5 N., which is a rather large change score in clinical practice. Therefore, in chapter 5 we showed that is feasible and reliable to measure the muscle strength with the HHD, but the SEM's and MDC's are rather large. These large SEM's and MDC's means that these tests can only detect a fairly large change in muscle strength as a true change in muscle strength, which makes the tests less suited to evaluate training programs and therapies in clinical practice in which only small changes can be expected.

CHAPTER 6
The responsiveness of muscle strength tests

With muscle strength being both a strong predictor for future negative health outcomes, and a prerequisite for physical fitness and daily functioning of adults with ID, it is important to be able to monitor the muscle strength of adults with ID over time. Therefore a muscle strength test should be able to detect change over time in muscle strength, which is called the responsiveness of a test. The aim of this study in chapter 6 was to assess the responsiveness of five field tests that measure muscle strength and endurance, namely grip strength (GS), HHD of leg extension and arm flexion, 10RM-test of the seated squat and the biceps curl, 30-second Chair stand (30sCS) and the 5-times Chair stand (5tCS) in adults with ID after the 24-week RT-program. The responsiveness of the five muscle strength and endurance tests was assessed by correlating the change scores on the five tests with the slope of the training progression, of specific exercises within the RT-program, namely the step up, seated squat, biceps curl and triceps curl. Nineteen participants (9 women, 9 mild ID, 10 moderate ID, mean age 42 ± 18 years [range 23 - 75]) performed all tests at baseline and at the end of the RT-program. The 10RM-test of the seated squat was significantly correlated with the step up ($R=0.53$, $p=0.02$) and the seated squat ($R=0.70$ $p<0.00$). Furthermore the HHD knee extension of the right leg was significant negatively correlated with the step up ($R=-0.52$, $p=0.02$) but not significant with the seated squat ($p=0.80$). None of change scores on the other tests were significantly correlated with the training progression on the other exercises. Therefore, we found that only the 10RM test of the seated squat could potentially be used to evaluate the effects of an RT-program in adults with ID. It is still questionable whether the GS, HHD-test, 10RM-test of the biceps curl, 30sCS and 5tCS could be used to evaluate the effects of an RT-program in adults with ID. Interestingly, the participants improved significantly on all four exercises in the RT-program, and in both the 10RM-test performances and the HHD-test of the elbow flexion, but no significant correlations were found. This stresses the need for more research into the interpretation of the results of RT-programs and the way muscle strength can be measured in adults with ID.

CHAPTER 7
General Discussion

This thesis showed that it is feasible for adults with ID to perform a total body workout with an exercise set for adults with ID (RESID), which can be tailored to their physical capabilities. This thesis also showed that it is feasible for adults with CVD risk factors to perform a 24-week RT-program at vigorous intensity. Furthermore, we showed that there are feasible, reliable and responsive muscle strength tests, which can be used to measure and monitor muscle strength. These results support physiotherapists and physical activity instructors to preform RT with adults with ID and monitor their muscle strength. Hereby, we introduced an RT-program that potentially could help adults with ID to get healthier. This thesis is the first step towards an evidence-based exercise intervention to reduce CVD risk for adults with a mild or moderate ID with CVD risk factors. The next step for future research is to study the health effects of this RT-program. This includes CVD health, but it would also be interesting to study the effect of the RT-program on muscle strength, muscle mass, sarcopenia, activities of daily living, arthrosis, osteoporosis, sleep and challenging behaviour. Additionally, it would be necessary for future research to study the cost-effectiveness and the quality-adjusted life-year (QALY's) of the RT-program within the healthcare system for adults with ID. Future research should also focus on finding responsive fitness tests to measure physical fitness and developing norm scores and cut-off points for the existing and new fitness tests. This would help health professionals to monitor and even prevent these health problems. People with ID at risk could then be screened and monitored to detect health problems. Moreover future research should also study ways to sustain the improved fitness and search for ways to keep adults with ID exercising within the national guidelines of PA in their daily life. The exercise set and RT-program give physiotherapists and other movement specialists new options in their efforts to support adults with ID into healthy aging.

SAMENVATTING

SAMENVATTING

HOOFDSTUK 1
Inleiding

Mensen met een verstandelijke beperking hebben gemiddeld meer gezondheidsproblemen. Zo hebben ze bijvoorbeeld een hoog risico op het krijgen van hart- en vaatziekten, doordat risicofactoren zoals diabetes, hoge bloeddruk, verhoogd cholesterol en het hebben van overgewicht of obesitas veelvuldig voorkomen. Het hebben van deze risicofactoren is op zichzelf ook al een gezondheidsprobleem. De standaard behandeling voor deze risicofactoren is nog steeds het geven van medicatie, maar onderzoek onder de algemene bevolking heeft aangetoond dat meer bewegen en sporten deze risicofactoren ook kan verminderen. Bewegen en sporten kan zelfs tot zo'n verbetering leiden dat er geen medicatie meer nodig is. De invloed van bewegen en sporten op deze risicofactoren is echter nog niet veel onderzocht bij volwassenen met een verstandelijke beperking .

Krachttraining is een vorm van bewegen die kan worden ingezet om de risicofactoren op het krijgen van hart- en vaatziekten te verbeteren. Er bestaan duidelijke algemene richtlijnen over hoe krachttraining eruit moet zien. Hoe meer je traint en hoe intensiever je traint, hoe groter het gezondheidseffect zal zijn. Krachttraining heeft ook een gunstig effect op de spiermassa, die door inactiviteit en het ouder worden afneemt (sarcopenie). Daarnaast kan het ook een positief effect hebben op het kunnen uitvoeren van activiteiten in het dagelijks leven en mogelijk ook op gedrag. Krachttraining als trainingsvorm zou haalbaar kunnen zijn voor volwassenen met een verstandelijke beperking, omdat het korte series van inspanning vereist, die gevolgd worden door een rustperiode. Het is echter onbekend of een intensief krachttrainingsprogramma, zoals wordt voorgeschreven in de richtlijnen, ook echt haalbaar is voor volwassenen met een verstandelijke beperking . Het doel van dit proefschrift is om de haalbaarheid van een intensief krachttrainingsprogramma te onderzoeken bij volwassenen met een verstandelijke beperking , die risicofactoren hebben op het krijgen van hart- en vaatziekten. Het tweede doel van dit proefschrift is om de haalbaarheid, betrouwbaarheid en responsiviteit te onderzoeken van fitheidstesten, waarmee de fysieke fitheid van volwassenen met een verstandelijke beperking kan worden gemeten en geëvalueerd.

HOOFDSTUK 2
Een pilot studie naar een set krachttrainings-oefeningen voor het hele lichaam

Voor krachtraining bij volwassenen met een verstandelijke beperking adviseert het American College of Sports Medicine (ACSM) om fitnessapparaten te gebruiken, omdat deze een goede uitvoering van oefeningen het beste ondersteunen. Helaas hebben volwassenen met een verstandelijke beperking in de dagelijkse praktijk lang niet altijd toegang tot fitnessapparaten. Daarom hebben we ons in dit proefschrift gericht op het ontwerpen van een set van krachttrainingsoefeningen voor volwassenen met een verstandelijke beperking voor het hele lichaam, waarbij geen fitnessapparaten nodig zijn. Deze set hebben we de Resistance Exercise Set for Adults with Intellectual Disabilities (RESID) genoemd. De RESID is ontworpen samen met zeven ervaren fysiotherapeuten en bewegingsagogen en bestaat uit zeven basisoefeningen. Voor iedere oefening is er ook een lijst met alternatieve oefeningen ontworpen, zodat de set aangepast kan worden naar de fysieke mogelijkheden van de volwassenen met een verstandelijke beperking . Vervolgens hebben we deze set van oefeningen (RESID) getest op haalbaarheid bij volwassenen met een lichte of matige verstandelijke beperking. Hieraan hebben elf deelnemers (waarvan 5 vrouwen, 10 personen met een lichte verstandelijke beperking , 1 persoon met een matige verstandelijke beperking , gem. leeftijd 29 ± 8 jaar [19-44 jaar]) deelgenomen. Zij hebben de RESID uitgevoerd tijdens twee van hun reguliere sportlessen. Bij de uitvoerbaarheid van de RESID werd gekeken naar het aantal oefeningen dat volledig uitgevoerd kon worden en naar de juiste uitvoering van de oefeningen. Voor alle oefeningen was het resultaat dat 90-100% volledig werd uitgevoerd, waarbij ook 90-100% goed werd uitgevoerd. De conclusie van het onderzoek is dan ook dat de RESID voor volwassenen met een lichte tot matige verstandelijke beperking uitvoerbaar is in de dagelijkse praktijk. Voorwaarde daarbij is dat een fysiotherapeut, bewegingsagoog of fitnessinstructeur aanwezig moet zijn tijdens de training om te letten op de correcte uitvoering van de oefeningen en om eventueel alternatieve oefeningen uit te kiezen.

HOOFDSTUK 3
De haalbaarheid van een intensief krachttrainings-programma

In hoofdstuk 3 hebben we met de krachtoefeningen (RESID) een progressief krachttrainingsprogramma ontwikkeld voor volwassenen met een verstandelijke beperking, met een risicofactor voor hart- en vaatziekten. Progressief betekent dat de trainingsintensiteit toeneemt gedurende het programma, in dit geval van beginnersniveau naar een intensief niveau. In dit proefschrift hebben we de haalbaarheid van dit programma onderzocht. De haalbaarheid van het krachttrainingsprogramma hebben wij gedefinieerd door te kijken naar het aantal deelnemers dat intensief traint aan het eind van het krachttrainingsprogramma, waarbij de haalbaarheid goed was als meer dan 50% van de deelnemers aan het eind van het programma intensief zou trainen.

Het krachttrainingsprogramma duurde 24 weken en er zijn 24 deelnemers gestart met de training. Ze hadden een lichte of matige verstandelijke beperking (waarvan 13 vrouwen, 11 met een lichte verstandelijke beperking , 13 met een matige verstandelijke beperking, de gemiddelde leeftijd was 44 ± 17 jaar [23-75 jaar]) en ze moesten minimaal één risicofactor voor hart- en vaatziekten (diabetes, hoge bloeddruk, verhoogd cholesterol, overgewicht/obesitas) hebben. Voor een haalbaarheidsonderzoek is het belangrijk dat ook alle risicofactoren vertegenwoordigd zijn in het onderzoek, wat in dit onderzoek ook zo was. De trainingsintensiteit werd stapsgewijs verhoogd van beginner (50%1RM) naar intensief (75%-80%1RM). 1RM staat voor repetitie-maximum van één, oftewel het gewicht dat een persoon maximaal één keer kan tillen. Bij 50%1RM kunnen ze het trainingsgewicht twintig keer tillen en bij 80%1RM kunnen ze dat acht keer. De krachttraining wordt zo steeds intensiever. Deelnemers gingen een stap intensiever trainen als zij minimaal vijf van de zeven oefeningen goed konden uitvoeren op de beoogde intensiteit gedurende acht trainingen; dus met de juiste techniek, uitvoering en ademhalingstechniek. Negentien deelnemers hebben het krachttrainingsprogramma volbracht, met een gemiddelde aanwezigheid van 73%. De haalbaarheid was goed, aangezien 58% (11 van de 19) van de deelnemers op het eind van het programma intensief aan het trainen was. In een vragenlijst na afloop gaven de trainers aan dat ze gedurende het hele krachttrainingsprogramma de deelnemers nauwlettend in de gaten moesten houden, om er zeker van te zijn dat de oefeningen met de juiste techniek en ademhaling werden uitgevoerd.

Dit onderzoek toont aan dat een intensief krachttrainingsprogramma haalbaar is voor volwassenen met een lichte tot matige verstandelijke beperking , die risicofactoren hebben voor het krijgen van hart- en vaatziekten. Fysiotherapeuten, bewegingsagogen en fitnessinstructeurs die ervaring hebben met het werken met volwassenen met een verstandelijke beperking, kunnen krachttrainingsprogramma gebruiken in de dagelijkse praktijk, zolang ze nauwlettend toezien op een goede uitvoering van de oefeningen.

HOOFDSTUK 4
De haalbaarheid van een supramaximale in spanningstest als verificatie na een stapsgewijze maximale loopbandtest.

Cardiorespiratoire fitheid (het uithoudingsvermogen) is een belangrijke maat voor de algehele fysieke fitheid, en daarmee ook een voorspeller voor de toekomstige fitheid. De gouden standaard voor het meten van cardiorespiratoire fitheid is een maximale inspanningstest op een loopband of een fietsergometer, waarmee de maximale zuurstofopname (VO_{2max}) gemeten wordt. De validiteit van maximale inspanningstesten is echter nog niet vastgesteld bij volwassenen met een verstandelijke beperking . In hoofdstuk 4 hebben we daarom de haalbaarheid onderzocht van een stapsgewijze maximale loopbandtest (GXTT), gevolgd door een supramaximale inspanningstest (SET) bij volwassenen met een licht tot matige verstandelijke beperking . De SET wordt daarbij gebruikt om te toetsen of de piek in zuurstofopname (VO_{2piek}) aan het eind van de GXTT een echte VO_{2max} is. Aan dit onderzoek deden twaalf deelnemers mee (waarvan 3 vrouwen, 4 met een lichte verstandelijke beperking en 8 met een matige verstandelijke beperking , de gemiddelde leeftijd was 30 ± 8 jaar [20-45 jaar]). Negen van de twaalf deelnemers hebben de meetprocedure volledig uitgevoerd (75%), wat wij gedefinieerd hebben als een uitstekende haalbaarheid. De 'intraclass-correlation-coefficient' (ICCagreement) van de genormaliseerde VO_{2piek} van de GXTT en de genormaliseerde VO_{2piek} van de SET was 0.99 ($P < .001$). De op maat gemaakte vragenlijst toonde aan dat de metingen voor de deelnemers acceptabel waren om uit te voeren. Tien deelnemers (83%) zouden de volgende keer weer deelnemen aan de metingen. Acht deelnemers

(67%) gaven echter aan dat ze het niet prettig vonden om gedurende de meetprocedure het masker te dragen waarmee de zuurstofopname wordt gemeten. Dit onderzoek toont aan dat het haalbaar is om bij volwassenen met een lichte of matige verstandelijke beperking een GXTT uit te voeren, gevolgd door een SET. Het is op deze manier dus mogelijk om te verifiëren of de gemeten VO_{2piek} van de GXTT ook een echte VO_{2max} is bij volwassenen met een verstandelijke beperking .

HOOFDSTUK 5
de haalbaarheid en betrouwbaarheid van de hand held dynamometer

The feasibility and reliability of hand held dynamometry

Spierkracht is ook een belangrijke voorspeller voor toekomstige gezondheidsproblemen en voor het kunnen uitvoeren van activiteiten in het dagelijks leven. Een potentieel haalbare manier om in de dagelijkse praktijk de spierkracht te meten, is door gebruik te maken van een 'hand held dynamometer' (HHD), in dit onderzoek de Microfet. Het doel van dit onderzoek was om de haalbaarheid en betrouwbaarheid van de HHD-metingen te onderzoeken bij volwassenen met een verstandelijke beperking . Aan dit onderzoek deden 21 van de 24 deelnemers mee die gestart waren met het krachttrainingsprogramma (waarvan 11 vrouwen, 10 met een lichte verstandelijke beperking en 11 met een matige verstandelijke beperking , de gemiddelde leeftijd was 44 ± 17 jaar [23-75 jaar]). De kracht werd met de HHD gemeten voor het buigen van de arm en het strekken van de knie,voor zowel links als rechts. De deelnemers voerden alle metingen twee keer uit. De haalbaarheid was gedefinieerd als het percentage deelnemers dat de metingen kon uitvoeren volgens het meetprotocol. De test-hertestbetrouwbaarheid werd berekend door de 'intraclass-correlation-coefficient' (ICCagreement), de 'standard error of measurement' (SEM) en de 'minimal detectable change' (MDC). De SEM zegt iets over de foutmarge van iedere meting en de MDC betekent de kleinste echt verandering die gemeten kan worden (daarbinnen kan het ook een meetfout zijn). De haalbaarheid van de HHD-metingen waren goed tot uitstekend voor de armflexie (66.7% - 81.0%) en matig tot goed voor de knie-extensie (42.9% - 72.4%). De betrouwbaarheid was uitstekend: alle ICCagreement ≥0.90. De SEM's waren tussen de 15.2 en de 28.1 Newton (N), wat resulteerde in MDC's tussen de 45.2 en de 53.5 N. Dit betekent, dat een verschil van boven de 45.2-53.5 N goed gemeten kan worden met deze test en dat is een vrij groot verschil voor gebruik in de dagelijks praktijk. In hoofdstuk 5 laten we dus zien dat het gebruik van de HHD haalbaar en betrouwbaar is bij volwassenen met een verstandelijke beperking , maar dat de SEM's en MDC's nogal hoog zijn. Deze hoge waardes voor de SEM's en MDC's betekenen dat alleen grote verschillen in spierkracht gemeten met de HHD kunnen worden aangemerkt als echte verandering van spierkracht. De HHD-test is daardoor minder geschikt om trainingsprogramma's en therapieën in de dagelijkse praktijk mee te evalueren, als er maar kleine veranderingen in spierkracht te verwachten zijn.

HOOFDSTUK 6
De responsiviteit van spierkrachtmetingen

Het is voor het meten van spierkracht niet alleen belangrijk om een haalbare en betrouwbare test te hebben. Het is ook belangrijk dat een test veranderingen van spierkracht in de loop van de tijd kan meten, bijvoorbeeld om spierkracht te kunnen monitoren of om het effect van een behandeling/training te kunnen meten. Dit heet de responsiviteit van een test. Het doel van het onderzoek in hoofdstuk 6 was om de responsiviteit te bepalen van vijf veldtesten die spierkracht en spieruithoudingsvermogen meten bij volwassenen met een verstandelijke beperking , in een krachttrainingsprogramma van 24 weken. Het ging om de knijpkracht (GS), de HHD van de armflexie en knie-extensie, de 10RM-test van de 'seated squat' en de 'biceps curl', de 30-second Chair stand test (30sCS) en de 5-times Chair stand test (5tCS). De responsiviteit van de spierkracht- en spieruithoudingsvermogenstesten hebben we bepaald door de veranderscores van deze testen te correleren met de gemiddelde trainingsprogressie van vier corresponderende oefeningen, namelijk de step up, de seated squat, de biceps curl en de triceps curl. Negentien deelnemers (waarvan 9 vrouwen, 9 met een lichte verstandelijke beperking en 10 met een matige verstandelijke beperking, met een gemiddelde leeftijd van 42 ± 18 jaar [23-75 jaar oud]) hebben alle testen aan het begin en het eind van het trainingsprogramma uitgevoerd. De 10RM-test van de seated squat correleerde significant met de step up (R=0,53, p=0,02) en de seated squat van het trainingsprogramma (R=0,7, p<0,00). Verder was de HHD van de knie-extensie significant negatief gecorreleerd met de step up (R=-0,52, p=0,02), maar niet significant met de seated squat (p=0,80). Geen van de veranderscores van de overige testen correleerde significant met de gemiddelde trainingsprogressie van de overige oefeningen. De conclusie van dit onderzoek is dan ook dat alleen de 10RM-test van de seated squat mogelijk gebruikt kan worden om het effect van een krachttrainingsprogramma te evalueren bij volwassenen met een verstandelijke beperking . Het is onzeker of de knijpkracht, de HHD-tests, de 10RM-test van de biceps curl, de 30sCS en de 5tCS gebruikt kunnen worden om het effect van een krachttrainingsprogramma te evalueren. Het is wel interessant dat de deelnemers significant hogere scores hadden bij alle vier de oefeningen en bij zowel beide 10RM-testen als bij de HHD-test van de armflexie, maar dat hierbij geen significante correlatie werd gevonden. Deze resultaten laten zien dat er meer onderzoek gedaan moet worden naar de resultaten van krachttrainingsprogramma's en naar de manier waarop spierkracht gemeten kan worden bij volwassenen met een verstandelijke beperking .

HOOFDSTUK 7
Discussie

Dit proefschrift laat zien dat het haalbaar is voor volwassenen met een verstandelijke beperking om hun spierkracht te trainen met de set oefeningen voor het hele lichaam (RESID), waarbij de oefeningen aan te passen zijn aan hun fysieke mogelijkheden. Ook is het haalbaar voor volwassenen met een verstandelijke beperking met risicofactoren voor het krijgen van hart- en vaatziekten, om een intensief krachttrainingsprogramma van 24 weken uit te voeren. Daarnaast laat dit proefschrift zien dat er haalbare, betrouwbare en responsieve spierkrachttesten zijn, die gebruikt kunnen worden om spierkracht te meten en monitoren bij volwassenen met een verstandelijke beperking. De resultaten bieden fysiotherapeuten en bewegingsagogen ondersteuning om met volwassenen met een verstandelijke beperking een krachttrainingsprogramma te gaan doen en ondersteunen hen bij het monitoren van spierkracht van volwassenen met een verstandelijke beperking. We introduceren hiermee een krachttrainingsprogramma dat volwassenen met een verstandelijke beperking mogelijk kan helpen gezonder te worden. Zo zetten we een eerste stap naar een evidence-based-oefenprogramma dat het risico op het krijgen van hart- en vaatziekten kan helpen verminderen bij volwassenen met een lichte of matige verstandelijke beperking met risicofactoren op het krijgen van hart- en vaatziekten. Vervolgonderzoek moet gaan uitwijzen wat de gezondheidswinst van dit krachttrainingsprogramma is voor volwassenen met een verstandelijke beperking. Dit gaat in eerste instantie over de gezondheidswinst door het verminderen van de risicofactoren voor het krijgen van hart- en vaatziekten, maar het is ook interessant om te kijken naar het effect van dit krachttrainingsprogramma op de spierkracht, de spiermassa, sarcopenie, activiteiten van het dagelijks leven, artrose, osteoporose, slaap en gedrag. Daarnaast is het voor toekomstig onderzoek goed om te gaan kijken naar de kosteneffectiviteit en naar de zogenaamde ‘quality-adjusted life year’ (QALY’s) van dit krachttrainingsprogramma binnen het zorgsysteem voor volwassenen met een verstandelijke beperking . Het onderzoek moet zich ook gaan richten op het vinden van responsieve fitheidstesten en op het ontwikkelen van normscores (hoe scoort iemand ten opzichte van anderen) en cut-off points (bij welke score moet er een behandeling gaan volgen) voor bestaande en eventueel nieuw te ontwikkelen fitheidstesten.

De resultaten van dat onderzoek kunnen zorgprofessionals helpen bij de preventie en het monitoren van eventuele gezondheidsproblemen bij volwassenen met een verstandelijke beperking. Om gezondheidsproblemen op tijd te signaleren zouden de volwassenen met een verstandelijke beperking met een verhoogd risico vaker gescreend en gemonitord kunnen worden. Verder moet toekomstig onderzoek zich richten op manieren waarop volwassenen met een verstandelijke beperking eventuele gezondheidswinst en fitheid kunnen vasthouden (en eventueel zelfs nog verder verbeteren) in hun dagelijks leven tot minimaal het niveau van de nationale richtlijn voor gezond bewegen. De set spierkrachtoefeningen (RESID) en het krachttrainingsprogramma geven fysiotherapeuten en bewegingsagogen nieuwe mogelijkheden om volwassenen met een verstandelijke beperking te ondersteunen om gezond oud te worden.

DANKWOORD

DANKWOORD

Een goede vriend zei enige tijd geleden tegen mij: “Jeetje, Stijn, dat jij nu gaat promoveren”. Ik heb zelf ook nog steeds diezelfde verbazing, want dat had ik niet gedacht toen ik in 2003 mijn studie fysiotherapie afrondde. Ik was niet zo'n fanatieke student. Wat was ik toen blij dat ik mijn studie erop had zitten. Ik was helemaal klaar met studeren, opgelucht dat ik eindelijk gewoon kon gaan werken. Er stond toen toevallig net een vacature in de Volkskrant voor een baan bij Abrona, een instelling voor mensen met een verstandelijke beperking. Dat leek me wel wat. Ik had totaal geen ervaring met de doelgroep, maar mijn stage bij de methylschool in revalidatiecentrum Roessingh vond ik - tot mijn eigen verbazing - heel erg leuk. Ik dacht: bij Abrona woont de volwassen versie van die kinderen, dus dat zou weleens wat kunnen zijn. Vanaf dat moment heeft het werken met mensen met een verstandelijke beperking me gegrepen en dat is na bijna twintig jaar niet veranderd. Het hielp natuurlijk mee dat ik terechtkwam in een baan met super fijne collega's. En nu, bijna twee decennia later, werk ik nog steeds met veel plezier als fysiotherapeut bij Abrona.

In 2008 besloot Abrona mee te doen aan GOUD: Gezond OUDer worden met een verstandelijke beperking. Dat gebeurde samen met de instellingen Amarant en Ipse de Bruggen, onder leiding van prof. Heleen Evenhuis van het Erasmus MC en senior onderzoeker Ruud van Wijck vanuit de universiteit Groningen. Ik was meteen enthousiast en wilde graag meehelpen. Wat was dat een mooi onderzoek en wat was het interessant om mee te helpen en om de metingen te doen. Thessa, Ruud, en later ook, Marieke en Alyt: bedankt voor die mooie tijd. Ik was en ben super trots op de GOUD-onderzoeken, die wereldwijd nog steeds echt uniek zijn. Ik raakte daardoor geïnspireerd om meer te doen met fysiotherapie. Maar wat? Daar heb ik goede gesprekken over gehad met mijn leidinggevende, Ineke Bootsman. Ik heb me eerst aangemeld als bestuurslid van de NVFVG, de Nederlandse Vereniging voor Fysiotherapeuten die werken met Verstandelijk Gehandicapten. Dat was een heel interessante periode, waarin we echt aan het pionieren waren. Ik wil dan ook alle bestuursleden, en met name Aleid, Mariet, Marije, Jan en Helmi, bedanken voor die mooie tijd. Ik merkte toen wel dat ik tegen een kennisgrens aan liep en dat leidde tot mijn keuze om na bijna tien jaar werken weer te gaan studeren.

In 2012 ben ik gestart met de opleiding Fysiotherapiewetenschap bij het Universiteit Utrecht. Deze keer deed ik een studie wél op het juiste moment; ik ben nog nooit zo fanatiek geweest. Ik wil Thessa bedanken voor alle steun en tips tijdens mijn studie en de begeleiding bij mijn afstudeerproject, dat ook als artikel is opgenomen in dit proefschrift. Het werkte heel motiverend dat ik in een enthousiaste, slimme en fanatieke studiegroep terechtkwam, met onder andere Sandra, Remco, Elja, Niek, Davy en Roel.. Niek, Roel en Davy, onze borrels in The Basket waren de perfecte afsluiting van elke studiedag en vormden bovendien het begin van een echte vriendschap. Bedankt voor

alle inspirerende discussies, enthousiaste verhalen en de mooie tijd. Het is fijn dat we elkaar nog regelmatig spreken.

Aan het eind van mijn studie, in 2015, zat ik te dubben wat ik nu met mijn nieuwe kennis wilde gaan doen. Het heeft zo moeten zijn dat er toen plotseling een nieuw onderzoek voorbij kwam binnen GOUD, naar de haalbaarheid van intensieve krachttraining. Dat onderzoek leek op mijn lijf geschreven en ik ben er dan ook vol voor gegaan. In 2016 kon ik beginnen met het onderzoek en dat leidt nu, zes jaar later, tot dit proefschrift.

In die zes jaar is er zo veel gebeurd, heb ik zoveel meegemaakt en moet ik dus ook heel veel mensen bedanken. Dit proefschrift was er niet geweest zonder de fijne begeleiding van mijn beide copromotoren, Alyt en Thessa. Ik ben jullie ontzettend dankbaar voor alle inhoudelijke discussies en jullie eindeloze geduld om mijn teksten te lezen en te corrigeren. Jullie wisten een prettige werkomgeving te creëren met jullie enthousiasme en vertrouwen. Alyt, als mijn dagelijkse begeleider heb ik heel fijn met je samengewerkt. Het voelde ook echt als sámenwerken. Je was betrokkenen en we hebben veel goede gesprekken gehad over hoe we dit onderzoek het beste konden opzetten. Verder kon ik met mijn twijfel ook altijd snel bij je terecht. Thessa, wij kennen elkaar al sinds het begin van GOUD, zoals ik hierboven al beschreef. Jouw enthousiasme is altijd erg aanstekelijk en je gaf mij veel vertrouwen. Tijdens mijn proefschrift begeleidde jij mij meer op afstand, eerst vanuit Chicago en later vanuit Las Vegas. Je wist het toch altijd voor elkaar te krijgen om mijn teksten snel te lezen en om via Skype af te spreken, vaak tijdens jouw ontbijt. Wat mij betreft is het hoog tijd dat jij samen met Dederieke als duo-hoogleraar Geneeskunde voor Verstandelijk Gehandicapten (GVG) wordt geïnstalleerd. Natuurlijk wil ik ook mijn officiële promotor bedanken. Sita, jij kwam pas op het laatst in beeld als mijn promotor, maar hebt mij erg geholpen om het laatste artikel en mijn proefschrift tot een goed einde te brengen. Hartelijk dank daarvoor.

Verder wil ik natuurlijk iedereen binnen de vakgroep GVG bedanken voor de mooie tijd op de negentiende verdieping van het Erasmus MC. Dederieke, Sylvie, Marieke, Mylene, Sandra en Marlies: bedankt voor alle steun, de gezelligheid, de lunchwandelingen, de nuttige feedback tijdens onze vakgroepvergaderingen, de gezellige tijd bij alle congressen en alle taken die horen bij het werken binnen een vakgroep. Dederieke, wat is het jou, samen met anderen uiteraard, goed gelukt om GVG uit te bouwen tot de geweldige en grote onderzoeksgroep die het nu is. Verder wil ik de studenten Geneeskunde Ikra en Florien bedanken voor het meewerken aan het onderzoek en hun bijdrage aan de trainingen en artikelen. Daarnaast wil ik iedereen bedanken bij de Huisartsgeneeskunde, waar de GVG een onderdeel van is.

Ik wil natuurlijk ook alle deelnemers heel erg bedanken voor het meedoen aan dit onderzoek. Wat hebben jullie goed getraind en wat was het gezellig. Super bedankt allemaal, ook de begeleiders die jullie gesteund hebben in die periode. Veder wil ik

iedereen binnen GOUD, Abrona, Amarant, en Ipse de Bruggen bedanken voor alle steun in de afgelopen jaren. Dank aan alle bewegingsagogen en fysiotherapeuten, waaronder Karin, Thomas, Denny, Marloes, Marloes, Suzanne, Olaf, Karin, Kirsten, Leendert, Renske, David, Amy, Jan Albert, en alle anderen die mij geholpen hebben door met de deelnemers te gaan trainen. Zonder jullie was het niet gelukt. Verder wil ik ook alle bestuurders, gedragsdeskundigen, artsen, doktersassistenten en praktijkverpleegkundigen bedanken voor hun interesse, ondersteuning, het meedenken en het meehelpen om deelnemers voor het onderzoek te benaderen. Voordorp Fitness uit Utrecht wil ik ook graag bedanken voor het gebruik van de faciliteiten van het fitnesscentrum, waar we met een aantal deelnemers konden trainen.

Ik wil mijn direct leidinggevenden bij Abrona, Ineke en Jeannette, bedanken. Ineke, ik wil jou als inspirerend leidinggevende bedanken voor de fijne, jarenlange samenwerking en voor de steun bij mijn studie en later bij het onderzoek. Jeannette, als mijn nieuwe leidinggevende ben jij pas sinds het slot van mijn onderzoek betrokken. Jij bedankt voor het meedenken om dit proefschrift eindelijk tot een goed einde te brengen.

Mijn lieve (oud-)collega's van de afdeling fysiotherapie bij Abrona wil ik natuurlijk bedanken. Bianca, Colinda, David, Geke, Inge, Jan, Kelly, Maud, Petra, Stefan, Valdelice en Viviane: zonder jullie steun, aanpassingsvermogen en enthousiasme had ik dit onderzoek niet kunnen doen. In eerste instantie was ik tweeëneenhalf jaar afwezig als fysiotherapeut, maar daarna was ik, met enige regelmaat, weer even afwezig, om te werken aan dit proefschrift. Ik ben erg blij met jullie als collega's en ik ben trots om samen met jullie de fysiotherapie binnen Abrona vorm te geven.

Natuurlijk wil ik ook mijn vrienden en familie bedanken. De hele Wombats-familie voor alle gezelligheid en sportiviteit op de vrijdagavonden. Mijn vrienden Job, Thomas, Douwe, Jaap en Onno voor alle gezelligheid, interesse, gesprekken en leuke activiteiten samen. De afgelopen jaren heeft vooral corona ervoor gezorgd dat we elkaar minder gezien hebben, maar ik koester jullie vriendschap.

Mijn schoonfamilie wil ik uiteraard bedanken. Koos en Christien, Liesbeth en Pim: bedankt voor alle warmte en gezelligheid alle jaren en de interesse die jullie altijd hebben gehad, ook in de vorderingen van dit proefschrift. En wat een plezier om mijn nichtjes en neefje te zien opgroeien.

Lieve Monique, ik ben er trots op dat jij mijn moeder bent. Jij hebt er altijd voor gezorgd dat we een fijn thuis hadden en hebben. Je bent een scherpe denker, die er, net als ik, van houdt om alles in een breed kader te plaatsen. We kunnen over allerlei onderwerpen dan ook pittige discussies hebben. Heel erg bedankt voor al je opbeurende woorden en aanmoedigingen tijdens dit onderzoek. En ik vind het nog altijd zo fijn dat je in Dick een maatje hebt gevonden waarmee je samen oud kunt worden. Dick, jij bedankt voor alle goede zorgen en je interesse in mijn activiteiten.

Lieve Marleen, wij hebben samen al zoveel meegemaakt .Het bracht ons iedere keer nog dichter bij elkaar. De scheiding van onze ouders, het overlijden van onze vader, Cees, in 2004, en het verschrikkelijke noodlot dat Eldert, jouw partner, trof in de periode dat ik met mijn onderzoek bezig was. Wat een verschrikkelijke tijd was dat en wat een verdriet dat Eldert er niet meer is. Ik vind het zo ontzettend knap hoe jij het leven weer hebt opgepakt en weer zingeving hebt kunnen vinden. Ik kan me bovendien geen betere zus wensen.

Lieve Janne, jij was er al bij toen ik mijn diploma Fysiotherapiewetenschap kreeg en dus ook toen ik begon met dit onderzoek. Hoe gezellig en interessant het op het Erasmus ook was, ik ging iedere avond snel naar huis om jou weer te mogen knuffelen en omje te horen praten en lachen. Wat ben jij toch een slimme, vrolijke en lieve meid. Toen ik in 2017 op mijn verjaardag in Belfast was voor een congres, kreeg ik het mooiste filmpje. Jij zong daarop zo mooi en blij een verjaardagsliedje voor mij, samen met mama. Ik heb eigenlijk nooit heimwee, maar toen wilde ik meteen naar huis - gelukkig was dat die middag al. Wat ben ik ongelooflijk blij dat jij er bent.

Lieve Otto, in 2018 had ik mijn laatste meting er net een week opzitten, toen jij op de wereld kwam. Ik kan me al niet meer voorstellen dat jij er niet altijd bij bent geweest. Jij bent zo'n vrolijk en enthousiast kereltje. Als baby was je al veel aan het lachen en eigenlijk is dat nooit opgehouden. Iedere keer als ik een weekend ging werken aan dit proefschrift en jullie uit logeren gingen bij opa en oma, miste ik jullie aanwezigheid en gezelligheid enorm. Wat een stilte heerst er dan. Ik ben nog steeds elke dag blij dat jij er bent.

En tot slot natuurlijk mijn lieve Sarah. In mijn eerste studiejaar Fysiotherapiewetenschap was jij jouw promotieonderzoek aan het afronden. Wat hebben we toen veel naast elkaar zitten werken; dat hielp enorm met de studiediscipline. Daarna was jij al doctor, maar moest ik nog even door. Ik had wel de smaak te pakken. Afgelopen jaar was ik druk om mijn promotieonderzoek af te ronden en was jij weer bezig met een master. Nu is het voor ons wel even genoeg geweest met studeren, hoop ik. We zijn al ruim zeventien jaar samen, waarvan acht jaar getrouwd. Ik geniet nog iedere dag van ons samenzijn. Ik hoop nog lang met jou te genieten van alles wat wij leuk vinden: softbal, series kijken, eten, vakanties en natuurlijk van Janne en Otto. Ik ben iedere dag weer blij dat wij samen zijn.

CURRICULUM VITAE

CURRICULUM VITAE

Stijn Weterings is geboren op 21 juni 1976 in Nijmegen. Na het behalen van zijn vwo-diploma aan het Frencken College in Oosterhout (1994) begon hij aan een studie Werktuigbouwkunde aan de Universiteit Twente. Deze studie heeft hij niet afgrond; in de eerste helft van 1997 is hij gaan werken en reizen. In 1997 is Stijn gestart met de opleiding Fysiotherapie op de Saxion Hogeschool in Enschede. In 2002 liep hij stage bij de kinderrevalidatie -afdeling van Roessingh, onderdeel van de toenmalige Metylschool Twente, thans het onderwijscentrum Roessingh. Als afstudeeropdracht schreef hij een handleiding voor fysiotherapeuten bij een myofeedback-aangestuurde treinbaan, die ontwikkeld was door de research- en developmentafdeling van het revalidatiecentrum Roessingh in Enschede. In maart 2003 is hij als fysiotherapeut gaan werken bij Abrona, een instelling voor mensen met een verstandelijke beperking in Huis ter Heide, waar hij tot op heden werkzaam is. Van 2006 tot 2012 is Stijnbestuurslid geweest bij de NVFVG, de Nederlandse Vereniging voor Fysiotherapeuten die werken met Verstandelijk Gehandicapten, waarvan hij in zijn laatste jaar voorzitter was. In 2012 is hij begonnen met de eenjarige premasteropleiding Fysiotherapiewetenschap en aansluitend aan de tweejarige master Fysiotherapiewetenschap aan de Universiteit Utrecht. In 2015 studeerde hij af, met als masterafstudeeronderwerp de haalbaarheid van supramaximale verificatie bij een maximale inspanningstest op de loopband bij mensen met een verstandelijke beperking. In 2016 is hij vanuit Abrona met zijn promotietraject begonnen. Het onderzoek vond plaats binnen het GOUD-consortium (Gezond OUDer met een verstandelijke beperking), waarvan Abrona partner is, samen met Amarant en Ipse de Bruggen en het Erasmus MC. Zijn onderwerp was het onderzoeken van de haalbaarheid van intensieve krachttraining bij mensen met een verstandelijke beperking, die risicofactoren hebben voor het krijgen van hart- en vaatziekten. Bijna drie jaar is Stijn volledig bezig met het onderzoek en is hij vrijgesteld van zijn werk als fysiotherapeut. Eind 2018 is hij weer aan de slag gegaan als fysiotherapeut en in 2019 is hij daarnaast ook gestart als implementatiecoach voor GOUD bij Abrona. Sinds 2017 is hij lid van de congrescommissie van het jaarlijkse congres van de NVFVG Stijn is getrouwd met Sarah Beeks. Zij wonen in Utrecht en hebben twee kinderen.

PHD PORTFOLIO

PHD PORTFOLIO

Erasmus MC department: General practice, Intellectual Disability Medicine
PHD period: 01-03-2016 – 01-01-2019
Promotor: Prof. Dr. S.M.A. Bierma-Zeinstra
Copromotoren: Dr. T.I.M. Hilgenkamp
Dr. A. Oppewal

1. **PHD training**	**Year**	**Workload (ECTS)**
General courses		
- BROK (Good Clinical Practice)	2017	**1.5**
- Scientific Integrity	2018	**0.3**
- Scientific writing	2018	**0.5**
Presentations		
- Department oral presentation	2016-2018	**2.0**
- Study oral presentation (managers, physicians, caregivers, physiotherapists and physical activity instructors)	2016-2018	**3.0**
- Amarant academy (oral presentation)	2016	**1.0**
National conferences		
- NVFVG congress 2016 (oral presentation)	2016	**2.0**
- Symposium NPG "Focus op Onderzoek" (2 oral presentations)	2018	**2.0**
- NVFVG congress 2021 (oral presentation)	2021	**2.0**
International conferences		
- 5th Rehab Move Congress, Groningen, the Netherlands (attendance)	2014	**0.3**
- Health SIRG conference, IASSIDD, Belfast, Northern Ireland (poster presentation)	2017	**2.0**
- 18th IASSIDD European congress, Athens, Greece (2 oral presentations)	2018	**3.0**
- 6th Rehab Move congress, Groningen, the Netherlands (poster presentation)	2018	**2.0**
- World congress IASSIDD 19, Glasgow, Scotland (oral presentation)	2019	**2.0**
2. **Teaching**	**Year**	**Workload (ECTS)**
Lecturing		
- Guest lectures "Gait and Gait analysis for adults with intellectual disabilities", Intellectual Disability physicians training, Erasmus MC	2017-2021	**4.0**
- Guest lectures "Physiotherapy with adults with intellectual disabilities", Physiotherapy study, Hogeschool Utrecht	2020-2021	**2.0**
Supervising Masters theses		
- 2 medical science masters students	2017-2018	**8.0**
- Supervising student session "How to judge a paper"	2018	**1.0**
3. **Other**	**Year**	**Workload (ECTS)**
- Member of NVFVG congress committee	2017-2021	**4.0**
Total		**42.6**

LIST OF PUBLICATIONS

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PEER-REVIEWED INTERNATIONAL PUBLICATIONS

Weterings S, Oppewal A, Bierma-Zeinstra SMA, Hilgenkamp TIM. (2022). The responsiveness of muscle strength tests in adults with intellectual disabilities. *Journal of Intellectual Disability Research*. doi: 10.1111/jir.12935.

Weterings, S., Oppewal, A., & Hilgenkamp, T. I. M. (2020a). The feasibility of vigorous resistance exercise training in adults with intellectual disabilities with cardiovascular disease risk factors. *Journal of Applied Research in Intellectual Disabilities, 33(3), 488-495.*

Weterings, S., Oppewal, A., van Eeden, F. M. M., & Hilgenkamp, T. I. M. (2018). A resistance exercise set for a total body workout for adults with intellectual disabilities, a pilot study. *Journal of Applied Research in Intellectual Disabilities. 32(3):730-736. doi: 10.1111/jar.12552*

Weterings, S., Takken, T., Hilgenkamp, T.I.M. (2017). The feasibility of supramaximal verification of peak oxygen uptake of a graded maximal treadmill test in adults with intellectual disabilities. *Cardiopulmonary Physical Therapy Journal* 28, 4, 117-127 doi: 0.1097/CPT.0000000000000057

NATIONAL ARTICLES

Weterings, S. (2018). Krachttraining bij mensen met een verstandelijke beperking. Gezond(er) OUD. *Fysiopraxis, mei 2018:40*

Weterings, S. (2017). Kan krachttraining de gezondheid van volwassenen met een verstandelijke beperking verbeteren? *TAVG, december 2017:160-164*

CONFERENCE PROCEEDINGS

Weterings S., Oppewal A. & Hilgenkamp T.I.M. (2019) Responsiveness of muscle strength tests in adults with intellectual disabilities. Journal of Intellectual Disability Research, Volume 63, Issue 7, 807-808.

Weterings S., Oppewal A. & Hilgenkamp T.I.M. (2018) Attendance of a resistance-training program in adults with an intellectual disability. Journal of Applied Research in Intellectual Disabilities, Volume 31, Issue 7, 542-543

Weterings S., Oppewal A., ; Razzaq I. & Hilgenkamp T.I.M. (2018) The reliability of measuring muscle strength in adults with an intellectual disability. Journal of Applied Research in Intellectual Disabilities, Volume 31, Issue 7, 543

LETTER TO THE EDITOR

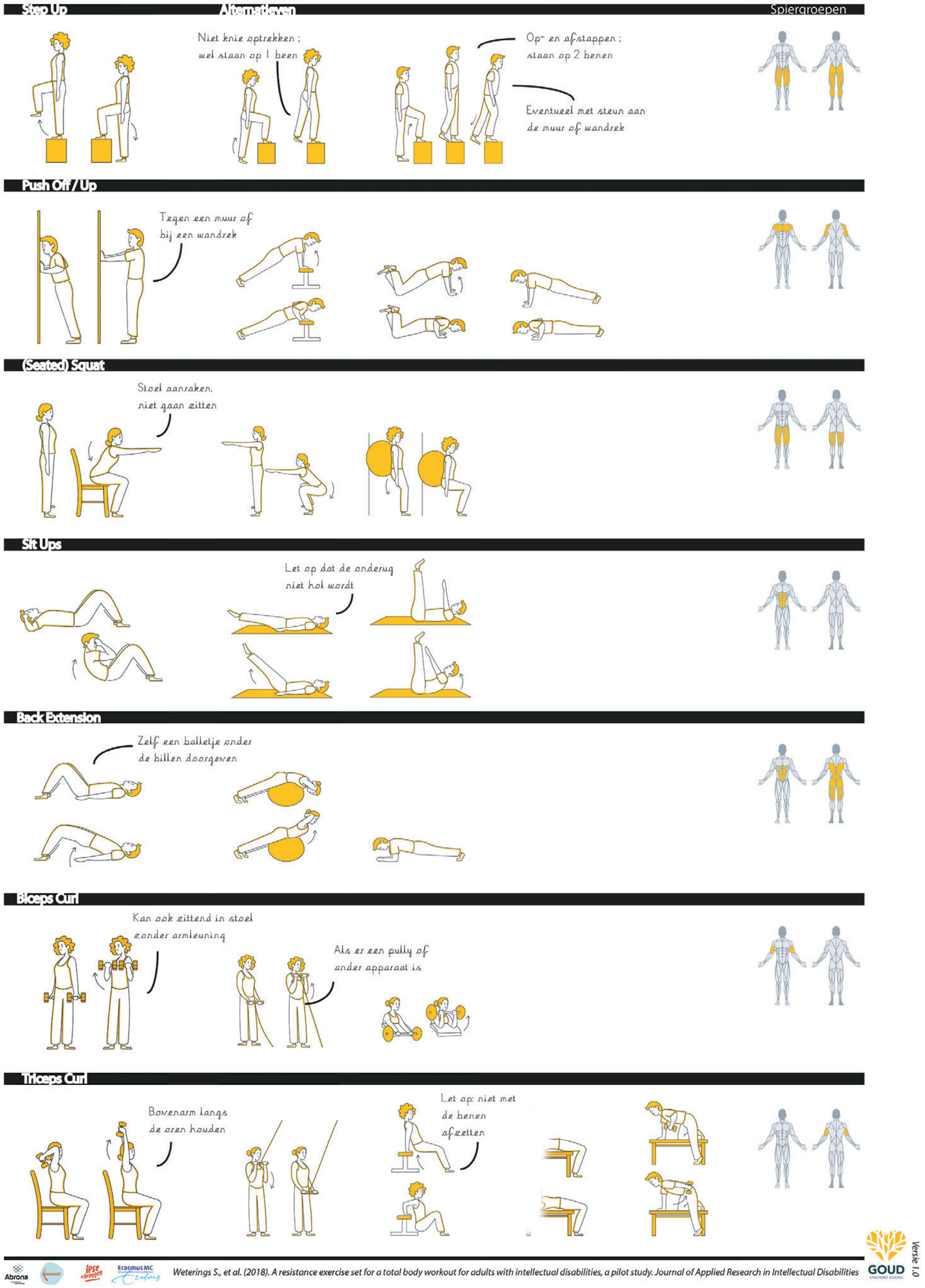
de Boks GG, Reurs MR, Runhaar J, Weterings S. (2019). Comment on 'Effects of a vestibular physiotherapy protocol on adults with intellectual disability in the prevention of falls: A multi-centre clinical trial' by Cortés-Amador et al.. *Journal of Applied Research in Intellectual Disabilities*. 32(4):1002-1003. doi: 10.1111/jar.12574

APPENDICES

APPENDIX I

RESID Total Body Workout

Met deze 7 oefeningen kan je zorgen voor sterke en gezonde spieren en botten.



Weterings S., et al. (2018). A resistance exercise set for a total body workout for adults with intellectual disabilities, a pilot study. Journal of Applied Research in Intellectual Disabilities

APPENDIX II

QUESTIONNAIRE ON THE FEASIBILITY OF THE GXTT AND THE SET FOR ADULTS WITH AN INTELLECTUAL DISABILITY [translated from dutch]

Client ID: …………………………………… date: …………………………..

Experience

1. How was it to do the test?

- Very annoying
- Annoying
- Not annoying, not nice
- Nice
- Very nice

2. How was it to practice before the test?

- Very annoying
- Annoying
- Not annoying, not nice
- Nice
- Very nice

3. How was it to wear a breathing mask?

- Very annoying
- Annoying
- Not annoying, not fine
- Fine
- Totally fine

4. How was it to wear a heart rate monitor?

- Very annoying
- Annoying
- Not annoying, not fine
- Fine
- Totally fine

5. What did you think of the starting speed of the treadmill?

- Way too slow
- Too slow
- Good
- Too fast
- Way too fast

6. What did you think of walking with an inclination on the treadmill?

- o Very annoying
- o Annoying
- o Not annoying, not fine
- o Fine
- o Totally fine

7. Would you like to perform the exercise tests another time?

- o No
- o Yes, namely……………………………………………………………………………………

8. Would you advise other persons to perform the exercise tests as well?

- o No
- o Yes, namely……………………………………………………………………………………

Difficulty

9. Did you understand the explanation and practising the test?

- o Yes
- o No

10. Did you understand what you had to do during the test?

- o Yes
- o No, namely……………………………………………………………………………………

11. How hard was it to perform the test?

- o Very hard
- o Hard
- o Fine
- o Easy
- o Too easy

Practicality

12. Were you able to perform the first part of the test?

- o Yes
- o No, namely……………………………………………………………………………………

13. Were you tired after the first part of the test?

- o No
- o Yes, namely……………………………………………………………………………………

14. Could you have walked longer on the treadmill, than you have done during the first part of the test?

- o No
- o Yes, namely…………………………………………………………………………………

15. Were you able to perform the second part of the test?

- o Yes
- o No, namely…………………………………………………………………………………

16. Were you tired after the second part of the test?

- o No
- o Yes, namely…………………………………………………………………………………

17. Could you have walked longer on the treadmill, than you have done during the first part of the test?

- o No
- o Yes, namely…………………………………………………………………………………

18. Did you sustain any injuries during the whole test?

- o No
- o Yes, namely…………………………………………………………………………………

19. If so, did it disappear?

- o Yes, after …… minutes
- o No

Do you have any remarks or comments on the test and/or the familiarization procedure?

……
……
……
……
…………………………………………………………………………………………………

Questions for the researcher

1. Was the familiarization procedure feasible for this participant?

- Yes, eg. problems……………………………………………………………………………………
- No, because…………………………………………………………………………………………..

2. Did you get the impression the participant understood the explanation of the familiarization procedure and did the participant perform the practice run well?

- Yes
- No, because…………………………………………………………………………………………

Which parts?…………………………………………………………………………….

3. Did you get the impression the participant understood the task and did the participant perform the test well?

- Yes
- No, because………………………………………………………………………………………..

4. Did you have to clarify the test procedure on the day of the test?

- No
- Yes, which part?……………………………………………………………………..................

5. Do you have any remarks or comments on the test and/or the familiarization procedure?

………
………
………
………
………
……………………………………………………………………………

How long did it take to perform the test? … min.

How long did it take to administer this questionnaire? … min