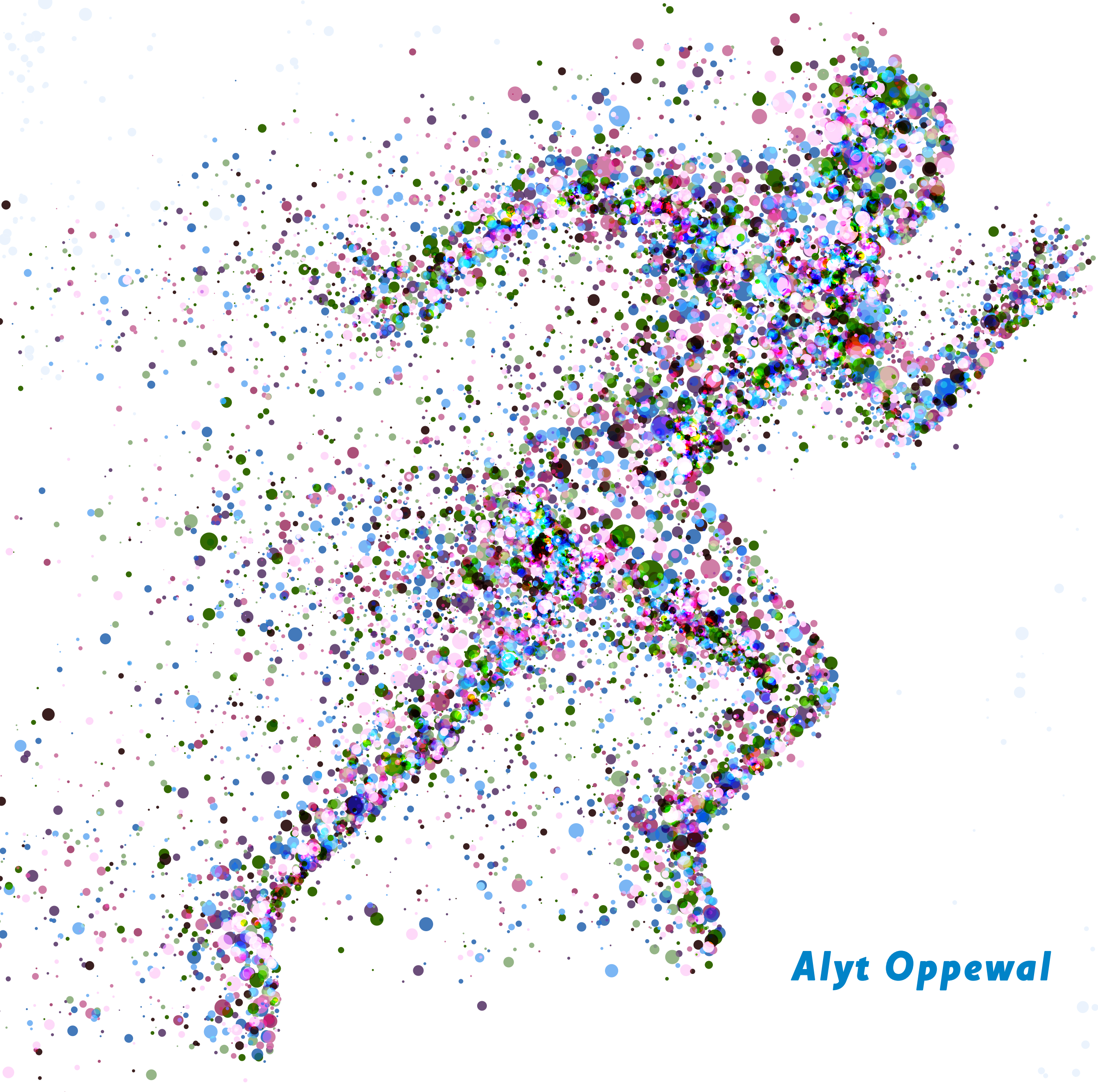
Measuring Physical Fitness in Older Adults with Intellectual Disabilities

Moving towards practice



Alyt Oppewal

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Moving towards practice

Het meten van lichamelijke fitheid bij ouderen met een verstandelijke beperking

Richting de praktijk

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Prof.dr. L.H.V. van der Woude

Copromotor: Dr. T.I.M. Hilgenkamp

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Chapter 1

General introduction

Physical fitness is "a set of attributes or characteristics individuals have or achieve that relates to their ability to perform physical activity" [1]. This definition shows that physical fitness is a multidimensional concept. There are different opinions about the exact components of this concept, but usually the components are divided into health-related components and skill-related components (Table 1) [1]. The health-related components are strongly related to overall health, and a low score on these components is a risk factor for all-cause mortality and developing disabling medical conditions and chronic diseases, such as metabolic syndrome, cardiovascular diseases, and musculoskeletal conditions [1-4]. Skill-related components represent athletic ability and are related with enhanced performance in sports and motor skills [1]. Being physically fit on both aspects is also a prerequisite to perform activities of daily living, for which both health-related and skill-related components are important [5, 6], although not all are equally relevant.

This thesis is about measuring the physical fitness of older adults with intellectual disabilities. Suitable physical fitness tests for this population are needed in clinical practice. Before outlining the contents of this chapter, we will start by providing some background information about why physical fitness should be a focus in the care for this population and why fitness tests are needed.

Table 1. Health-related and skill-related components of physical fitness according to the American College of Sports Medicine.

Health-related components	Skill-related components
Body composition	*Coordination*
Cardiorespiratory fitness	*Balance*
Muscular strength	*Reaction time*
Muscular endurance	Agility
Flexibility	Power
	Speed

People with intellectual disabilities are particularly at risk for low levels of physical fitness, because of a higher prevalence of physical inactivity, sensory and mobility impairments, obesity, and chronic health conditions than in the general population [7-12]. Indeed, low physical fitness levels are seen in people with intellectual disabilities across the lifespan [13-16], and this may lead to unnecessary health problems and functional problems, and higher need for care. This increased need for care leads to increased health care costs, which are already high in this relatively small population. Worldwide, the prevalence of intellectual disabilities has been estimated at 10.4/1000, similar to a prevalence of 1% [17]. In the Netherlands, the number of people with intellectual disabilities receiving any form of support or care has also been estimated to be around 1% of the total Dutch population [18, 19]. Although this is only a small percentage of the

Dutch population, the health care and residential costs for this group were 6.8 billion euros in 2011, representing 12.5% of the total Dutch health care costs [19]. This is higher than for any other patient group [20].

In addition, as in the general population, the life expectancy of people with intellectual disabilities is increasing, due to improved care [21, 22]. However, an increase in the quantity of life does not mean that the quality of life is also increasing. Since physical fitness is strongly related to health and daily functioning in the general population [1-6], the lifelong low physical fitness levels of people with intellectual disabilities may have a detrimental effect on their health and level of dependency at older age, affecting their quality of life and increasing the burden placed on family, caregivers, and health care facilities. Nevertheless, physical fitness has never been a focus in the care of people with intellectual disabilities, though physical fitness may be an important aspect for the optimal care for this population.

However, few longitudinal studies have been performed in people with intellectual disabilities, and it is unknown if low physical fitness levels have the same consequences for health and daily functioning as in the general population [23, 24]. The consequences of low physical fitness levels for people with intellectual disabilities may be confounded by their lifelong cognitive impairments. These cognitive impairments may have negatively influenced their motor development since childhood, which may have negatively influenced their physical fitness throughout life. This line of reasoning is supported by the finding that motor and cognitive functioning are fundamentally interrelated, with similar developmental trajectories and the use of similar brain structures [25]. If the starting point of limitations in physical fitness is already different, this may as well influence the relation between physical fitness and health and daily functioning. Since prevention of diseases and disability has become an important aspect in the policy of the governments worldwide, and also of the Dutch Ministry of Health, Welfare, and Sport [23, 24], knowledge about the exact consequences of low physical fitness in older adults with intellectual disabilities is important to clarify what the value of improving physical fitness could be for the prevention of diseases and disability in this population.

To evaluate the effectiveness of prevention programs and therapeutic interventions in this group, physical fitness tests are needed. This is becoming more and more important, because the Dutch health care system increasingly demands evidence-based care. In addition, regular physical fitness screening could be used to identify people at risk for developing or worsening health conditions and daily functioning, to plan interventions, to educate and inform people with intellectual disabilities, their family, caregivers, and care organizations about the current fitness status, and finally fitness tests may help in motivating people in attaining fitness goals. Unfortunately, physical therapists do not have physical fitness tests to assess the physical fitness levels of people with intellectual disabilities, and physical fitness tests used in the general population may not be suitable, due to the combination of ageing and limited cognitive abilities. It is therefore important that physical fitness tests that are suitable for people with intellectual disabilities become available for use in clinical practice.

The problems that become apparent here clarify the need for suitable physical fitness tests. In the 'Healthy ageing and intellectual disabilities' (HA-ID) study, a first step has been made in investigating physical fitness tests for this population. However, so far, only parts of the information needed for using these physical fitness tests in practice is available.

In this chapter we start by introducing the HA-ID study, and discuss how physical fitness was operationalized and measured in the HA-ID study. Then we will provide criteria for the use of physical fitness tests in practice, identify the gaps in knowledge, and finally present the aims and content of this thesis.

THE HEALTHY AGEING AND INTELLECTUAL DISABILITIES STUDY

The HA-ID study was a large epidemiological study addressing health in older adults with intellectual disabilities from 2008 until 2013. The study was performed in a consort of three Dutch intellectual disability care organizations (Abrona at Huis ter Heide, Amarant at Tilburg, and Ipse de Bruggen at Zoetermeer) and the department of Intellectual Disability Medicine as part of the department of General Practice at the Erasmus Medical Center Rotterdam. Inspired by questions of the care organizations, three themes were chosen for the study: (1) physical activity and fitness, (2) nutrition and nutritional state, and (3) mood and anxiety. For the physical activity and fitness theme there was collaboration with the Center for Human Movement Sciences of the University Medical Center Groningen. At the start of the study in 2008, the care organizations in the consort provided support and care for 2322 clients aged 50 years and over, of which 1050 clients participated in the HA-ID study. This was a near-representative sample with a slight overrepresentation of women, and an underrepresentation of 80 to 84 year-olds and older adults that only receive ambulatory care or only visit a day-activity center. In addition, the HA-ID study sample did not include older adults with intellectual disabilities without any form of registered care or support. Data collection was performed between February 2009 and July 2010. Among others, measurements consisted of an extensive physical fitness assessment, physical activity measurements with pedometers, physical examination, laboratory assessment, swallowing observations, questionnaires regarding daily functioning and falls, and a depression and anxiety screening. To assess the consequences of health and fitness conditions, follow-up data was collected on daily functioning, falls, and medical conditions three years after the baseline data collection. A more detailed description about design, recruitment, and representativeness of the HA-ID study has been described by Hilgenkamp et al. (2011) (Appendix).

OPERATIONALIZATION OF PHYSICAL FITNESS AND SELECTION OF TESTS USED IN THE HA-ID STUDY

In the HA-ID study, physical fitness was operationalized with a combination of health-related components and skill-related components [26]. Those components that are primarily important for daily functioning of older adults were chosen. These components were coordination (manual dexterity), reaction time, balance (dynamic and static balance), muscular strength, muscular endurance, flexibility, and cardiorespiratory fitness (Table 1).

For physical fitness tests to be selected they had to consist of a functional task, have at least one reference of reliability or validity, be provided with an objective scoring system and available normative data of the general population, and not be too difficult in instructions, execution, and administration [26]. After an extensive literature search and expert meetings with physiotherapists and movement experts with experience in working with older adults with intellectual disabilities, eight physical fitness tests were selected [26]. Coordination was measured with the Box and Block test [27], reaction time with an auditive and visual reaction time task [28], balance with the Berg Balance Scale [29] and gait speed [30], muscular strength with grip strength measured with a hand dynamometer [31], muscular endurance with the 30s Chair stand [32], flexibility with an extended version of the modified back saver sit and reach test [26, 33], and cardiorespiratory fitness (maximal oxygen uptake) was measured with the 10-meter incremental shuttle walking test [34].

CRITERIA FOR THE USE OF PHYSICAL FITNESS TESTS IN PRACTICE

To establish which information is still needed before these tests can be implemented in clinical practice, we will start by discussing the criteria which should be met by an ideal physical fitness test [1, 32, 35, 36].

Feasibility

Feasibility of a test encompasses the completion rates, the clarity of instructions, organizational demands of the test (time, space, materials, and training), and quick and objective scoring. Whether the organizational demands and scoring system are acceptable, depends on the setting and aim of the physical fitness assessment. Therefore, a general objective criterion for these two aspects of feasibility is not necessary to establish.

Reliability

Reliability represents the consistency or repeatability of test scores [37]. Test-retest reliability is the consistency of results across different occasions with the same respondents and tester, and inter-rater reliability is a measure of the consistency of results obtained by more than one tester [37].

Validity

Validity is the extent to which a test accurately measures what it is supposed to measure. The three major types of validity are content validity (degree to which a test accurately measures the defined domain or content), construct validity (degree to which a test measures the construct of interest), and criterion validity (degree to which a test correlates with a criterion measure) [37]. Especially the criterion validity of each test has to be assessed in the population of older adults with intellectual disabilities, because the predictive value of a test may be population specific due to specific population characteristics.

Responsiveness to change in physical fitness levels

A physical fitness test should be responsive to detect individual changes in physical fitness levels.

Comparability to normative data

Normative data from large data sets must be available to interpret the test scores. The two types of reference values are norm-referenced and criterion-referenced values [32]. Norm-referenced values from the general population, as well as from the population of older adults with intellectual disabilities should be available, to compare scores of older adults with intellectual disabilities within both these populations. Criterion-referenced values should be population specific, because this is related to the predictive value of a test, which may be different per population.

GAPS IN THE REQUIRED KNOWLEDGE FOR THE USE OF PHYSICAL FITNESS TESTS IN OLDER ADULTS WITH INTELLECTUAL DISABILITIES

To support implementation of the selection of physical fitness tests in clinical practice, all of the above criteria must be met, but so far, research has provided only parts of this information. Table 2 provides an overview of the gaps in knowledge, identified by the empty boxes.

With regard to the feasibility, the clarity of instructions and the level of difficulty of instructions and execution of the eight selected physical fitness tests had to be sufficient

Table 2. Identified gaps in the required knowledge for the use of physical fitness tests in clinical practice for older adults with intellectual disabilities.

Criteria	Coordination *(Box and Block test)*	Reaction time *(Reaction time task)*	Balance *(Berg Balance scale)*	Speed *(Gait speed)*	Strength *(Grip strength)*	Muscular endurance *(30s Chair stand)*	Flexibility *(Back saver sit and reach)*	Cardiorespiratory fitness *(Shuttle walking test)*
Feasibility								
Completion rates [40]	✓	✓	This thesis	✓	✓	✓	✓	This thesis
Clarity of instructions [40]	✓	✓	✓	✓	This thesis	✓	✓	✓
Reliability in older adults with ID								
Test-retest reliability [37]	✓	✓	✓	✓	✓	✓	✓	✓
Inter-rater reliability (unpublished data)	✓	✓	✓	✓	✓	✓	✓	✓
Validity in older adults with ID								
Criterion validity	This thesis	This thesis	This thesis	This thesis	This thesis	This thesis	This thesis	This thesis
Responsiveness to change								
Comparability to normative data								
Norm-referenced values of the general population available [1, 41-45]				✓	✓	✓		✓
Norm-referenced values of older adults with ID available [13]			This thesis	✓	✓	✓		✓
Criterion-referenced values of older adults with ID available								

✓= knowledge available; empty boxes = gaps in knowledge; this thesis = gaps addressed in this thesis; ID = intellectual disabilities.

for the test to be selected [26]. In addition, the completion rates on these tests have previously been investigated by Hilgenkamp et al. (2012), and all tests had moderate to good completion rates, except for participants with severe intellectual disabilities (reaction time test and Berg Balance Scale), profound intellectual disabilities (all tests), and wheelchair users (all tests involving the legs) [38]. Although the 10-meter incremental shuttle walking test had overall good completion rates, 61% of the participants did not reach the required peak heart rate during the test, which was a criterion to estimate maximal oxygen uptake [13, 39]. Maximum oxygen uptake results were therefore not available for the majority of the participants. For the tests that were not feasible (for some subgroups of older adults with ID), more knowledge is needed about the feasibility of possible alterations on tests or alternative tests. With regard to the clarity of instructions of the grip strength test, it is unknown what the effect of handedness is and whether the grip strength of one or both hands should be measured.

Reliability and validity of the selected physical fitness tests have been confirmed in the general population. Hilgenkamp et al. (2012) and de Jonge et al. (2010) have shown that the selected tests also have sufficient test-retest reliability in older adults with intellectual disabilities, with higher values for the same-day interval than for the 2-week interval [38, 40], and good inter-rater reliability (ICC of 0.94 to 0.99; unpublished data). The validity has not yet been investigated in older adults with intellectual disabilities. It is especially important that the predictive validity of each test is investigated specifically for older adults with ID, to assess if the tests can be used to predict future health events.

To assess the responsiveness to change of the tests, longitudinal studies are needed. No studies regarding the responsiveness of these selected tests have been performed in older adults with intellectual disabilities.

Norm-referenced and criterion-referenced values are important for the interpretation of physical fitness tests. For gait speed, grip strength, maximal oxygen uptake, and the 30s Chair stand test, norm-referenced values and criterion-referenced values of the general population are available from large data sets. For these physical fitness tests, the results from the HA-ID study are available and may be used as population specific norm-referenced values for older adults with intellectual disabilities [13], while taking the characteristics of the study sample into account. For the other tests, these values are lacking. In addition, no population specific criterion-referenced values are available for older adults with intellectual disabilities.

CONTENTS OF THIS THESIS

Therefore, the aim of this thesis was to address a number of the above-identified gaps in the knowledge required for implementation of physical fitness tests in clinical practice (Table 2).

First, issues regarding the feasibility will be addressed. Since it appeared that a large number of participants was unable to meet the criteria of peak heart rates for the 10-meter incremental shuttle walking test (cardiorespiratory fitness) [13], a literature review was performed to provide an overview of published information concerning the validity and reliability of cardiorespiratory fitness testing in people with intellectual disabilities, and their cardiorespiratory fitness levels (*Chapter 2*). Then, to address the feasibility issues regarding cardiorespiratory fitness testing, the usefulness of heart rate recovery as an outcome measure in cardiorespiratory fitness testing in older adults with intellectual disabilities was assessed *(Chapter 3*). Personal characteristics associated with heart rate recovery are also described.

To improve the clarity of the instructions for grip strength measurements in older adults with intellectual disabilities, the effect of handedness on grip strength was studied, resulting in recommendations about the best way to measure grip strength and to interpret results (*Chapter 4)*.

To address the feasibility of the Berg Balance Scale, *Chapter 5* describes the reasons for drop-out on the items of the Berg Balance Scale for subgroups with low completion rates, and identifies feasible subtests for subgroups. In addition, to address the lack of population specific norm-referenced values, the scores on the Berg Balance Scale of the HA-ID population are also presented, which may be used as norm-referenced values.

To assess the predictive (criterion) validity of the physical fitness tests, data on falls and daily functioning was collected three years after the baseline data collection. The predictive value of balance, gait speed, muscular strength and muscular endurance for falls is described in *Chapter 6*, and in *Chapter 7* the predictive value of all eight physical fitness tests for daily functioning is described.

Finally, in the general discussion, we reflect on the results presented in this thesis. Implications for clinical practice and further research are discussed (*Chapter* 8).

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Chapter 2

Cardiorespiratory fitness in individuals with intellectual disabilities – A review

Alyt Oppewal, Thessa I.M. Hilgenkamp,
Ruud van Wijck, Heleen M. Evenhuis

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ABSTRACT

Cardiorespiratory fitness is the ability of the circulatory, respiratory, and muscular systems to supply oxygen during sustained physical activity. Low cardiorespiratory fitness levels have been found in individuals with intellectual disabilities (ID), which puts them at higher risk for cardiovascular diseases and all-cause mortality. The aims of this review were to update previous reviews about (a) the cardiorespiratory fitness levels and their determinants in individuals with ID, and (b) the validity and reliability of cardiorespiratory fitness testing in individuals with ID. We searched the databases of Pubmed and Embase for relevant studies, resulting in 31 included articles. These studies mainly included younger participants with mild to moderate ID. Results confirmed previous findings of low cardiorespiratory fitness levels in individuals with ID. Cardiorespiratory fitness levels of children and adolescents with ID are already low, with further decline with increasing age. Furthermore, females have lower cardiorespiratory fitness levels than males. Physical inactivity and chronotropic incompetence are most likely to contribute to low cardiorespiratory fitness levels. Peak cardiorespiratory fitness levels of individuals with ID can be assessed with maximal treadmill protocols, after allowing for familiarization sessions. Although, predicting maximal oxygen uptake from field tests is problematic, field tests have been found valid and reliable as indicators of cardiorespiratory fitness.

INTRODUCTION

Cardiorespiratory fitness is the ability of the circulatory and respiratory systems to supply oxygen to working muscles during sustained physical activity [1]. Poor cardiorespiratory fitness is a major independent risk factor for cardiovascular diseases and all-cause mortality [1-3]. Cardiovascular diseases are the number one cause of disability and premature morbidity and mortality throughout the world [3]. Physical activity levels [4-6] and cardiorespiratory fitness of individuals with intellectual disabilities (ID) are low [7, 8], which puts them at a high risk for cardiovascular diseases.

The standard measure to express cardiorespiratory fitness is $\dot{V}O_2$max, which is defined as the maximal oxygen uptake, achieved during a maximal cardiorespiratory exercise test, conducted at sea level, in which a sufficiently large muscle mass is used [9]. It is the product of the maximal cardiac output and arterial-venous oxygen difference. Variations in $\dot{V}O_2$max are primarily due to differences in maximal cardiac output [10], which is the product of heart rate (HR) and stroke volume (SV) [11]. The highest achieved oxygen uptake during a maximal cardiorespiratory exercise test can be considered as $\dot{V}O_2$max when a plateau in oxygen uptake with an increase in work rate is reached. This is the primary criterion. However, when a plateau is not reached, there are secondary criteria to check whether $\dot{V}O_2$max is reached (at least two criteria out of three): (a) a plateau in HR with an increase in work rate or within 10 beats of the estimated maximal HR, (b) respiratory exchange ratio (RER) > 1.0, and (c) high levels of lactic acid in the minutes following exercise [9, 11-13]. When these criteria are not met, the highest achieved oxygen uptake during the test is called $\dot{V}O_2$peak. In individuals with ID, test results are often called $\dot{V}O_2$peak, which means that they have reached volitional exhaustion (the point at which the participant feels he/she can no longer continue) [14].

Ideally, $\dot{V}O_2$max is measured directly with a maximal exercise test on a treadmill or bicycle ergometer with open-circuit spirometry under standardized conditions [10]. However, dependent of the population being tested and the availability of personnel and equipment, a submaximal exercise test might be the first choice. $\dot{V}O_2$max is then estimated, based on the relationship between HR and work rate. These estimations are based on the assumptions that (a) a steady state in HR is obtained for each work load and this is consistent each day, (b) the relation between HR and work rate is linear, (c) the maximal achieved work load is indicative for $\dot{V}O_2$max, (d) the maximal HR for a given age is uniform, (e) $\dot{V}O_2$ at a given work rate is the same for everyone, and (f) the participant is not on medication that alters HR [10]. Equations developed to estimate $\dot{V}O_2$max or HRmax are mostly based on data from the general population, but it has been shown that these equations overestimated $\dot{V}O_2$max and HRmax in individuals with ID [15-17].

For valid and reliable assessment of the cardiorespiratory fitness of individuals with ID, it is important that the participant is able to understand and execute the required test procedures. Therefore, a familiarization protocol prior to actual testing should be performed, both for maximal and submaximal testing [14, 18]. Motivational problems, unfamiliar signs of exertion, difficulties with pacing, limited coordination, unfamiliar task and actions, and an unfamiliar test environment and tester are all issues that can and should be addressed in the familiarization sessions. The number of required familiarization sessions should be individually determined based on the participants' performance with the guideline of a minimum of one session for field tests and a minimum of two sessions for laboratory tests [18].

An overview of studies regarding cardiorespiratory fitness in individuals with mild to moderate ID and their possible determinants was provided by Fernhall & Pitetti (2001). They showed that individuals with ID have lower levels of cardiorespiratory fitness compared to individuals without ID, with even lower levels for individuals with Down syndrome (DS). Furthermore, females have lower cardiorespiratory fitness levels than males. Possible determinants for this low cardiorespiratory fitness discussed were (a) poor motivation and task understanding, (b) lack of physical activity and sedentary lifestyle, (c) poor leg strength, and (d) chronotropic incompetence, the inability of the heart to increase its rate proportionally to the demand [7]. None of the studies reviewed by Fernhall & Pitetti (2001) included adults older than 35 years. Since then, more research has been performed and an update is necessary to provide an overview, and pinpoint the gaps in the current knowledge.

Mendonca et al. (2010) provided an update specifically for individuals with DS. The cardiorespiratory fitness of individuals with DS is low and was found lower than in individuals with ID by other causes. Chronotropic incompetence was found to be an important determinant, out of several possible explanations, of low cardiorespiratory fitness. The exact mechanism is not yet completely understood, but it is most likely a combination of reduced catecholamine (noradrenalin, adrenalin) responsiveness and blunted parasympathetic withdrawal during exercise [8]. No update for individuals with ID by other causes is available yet. Therefore, our first aim was to provide an overview of the recent literature about the cardiorespiratory fitness levels and their possible determinants in individuals with ID, excluding DS.

An overview of cardiorespiratory fitness tests applicable to individuals with ID was provided by Pitetti et al. (1993). At that point in time, five field tests and two laboratory tests had been validated for individuals with mild to moderate ID. The 1.5-mile (2.4 km) walk-run and the 1-mile (1.6 km) Rockport fitness walking test (RFWT) had been found valid in men. The protocol for both walk/run tests involved one tester per participant; another tester-to-participant ratio had not been validated yet. Bicycle ergometer testing with the Schwinn 'Air-Dyne' was found valid only if the participants could maintain constant work levels. The modified Leger and Lambert shuttle run test and the modified Canadian step test had also been found valid as field tests. However, maintaining a proper pace, agility, and muscle coordination may affect the validity of these tests by limiting a participant's performance, independent of their cardiorespiratory fitness. The two validated laboratory tests were a maximal treadmill protocol and Schwinn 'Air-Dyne' protocol. The maximal treadmill protocol, in which speed was held constant and grade was gradually increased until exhaustion, was valid and reliable for adolescents and adults with mild to moderate ID. The Schwinn 'Air-Dyne' protocol showed similar results to the maximal treadmill protocol in adults with mild ID [14]. Since the review of Pitetti et al. (1993), more research regarding validity and reliability of cardiorespiratory fitness testing in individuals with ID has been performed. An update is necessary to provide an overview of the current knowledge. This was the second aim of our review.

Therefore, the aims of this review were to provide an overview of the recent literature about (a) the cardiorespiratory fitness levels and their determinants in individuals with ID, excluding DS, and (b) the validity and reliability of cardiorespiratory fitness testing in individuals with ID, including DS.

METHODS

Literature search

A literature search in the databases of Pubmed and Embase was performed in the period July 2012 to March 2013. The following search strategy and keywords were used: **[**'Intellectual disability (MeSH Terms)' OR 'intellectual disabilit*' OR 'mental*' and retard*' OR 'learning disabilit*' OR 'developmental disabilit*' OR 'cognitive disabilit*' OR 'mental* AND handicap*' OR 'Down syndrome'**]** AND **[**'maximal heart rate' OR 'heart rate' OR 'maximal exercise' OR 'exercise response' OR 'cardiorespiratory' OR 'cardiovascular' OR 'cardiorespiratory fitness' OR 'cardiovascular fitness' OR 'cardiorespiratory endurance' OR 'cardiovascular endurance' OR 'endurance' OR 'cardiorespiratory capacity' OR 'cardiovascular capacity' OR 'exercise capacity' OR 'cardiorespiratory' OR 'oxygen consumption' OR 'exercise test*'**]** NOT **[**'animal' OR 'mouse' OR 'rat'**]**.

Selection of articles

To be included in this review, articles had to cover at least one of the following topics in individuals with ID:

1) Cardiorespiratory fitness levels.
2) Determinants for cardiorespiratory fitness levels.
3) Reliability and/or validity of cardiorespiratory fitness testing.

Articles covering topic 1 and 2 were included from the year 2000, as a continuation of the review of Fernhall & Pitetti (2001). Articles about cardiorespiratory fitness testing were included from the year 1993, as a continuation of the review of Pitetti et al. (1993).

Articles were excluded if they were not written in English. For criteria 1 and 2, articles were excluded if they did not include a group of individuals with ID by other causes than DS. When we talk about individuals with ID in the current review, we mean individuals with ID by other causes than DS, if otherwise DS will be explicitly mentioned.

After the electronic database search, articles were screened using the selection criteria in three rounds. In the first round, selection was based on the title, followed by the abstract, and finally the full text. Reference lists of the included articles after the three rounds were checked for relevant studies. Figure 1 is the flow diagram of this process. The selection process was carried out by the first author. However, doubts about inclusion were discussed with the second author until consensus was reached.

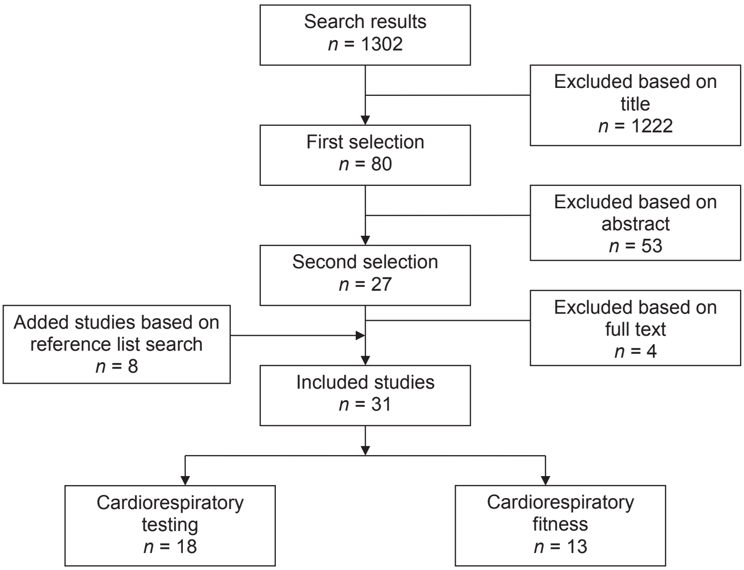

Figure 1. Flow diagram of the data selection.

Data extraction and management

Data was extracted from the selected articles by the first author. Articles were systematically and critically read according to a structured form [19].

The following data was extracted:

1) Objectives of the study.

2) Study design.

3) Study population, number of participants, and participant characteristics (age, gender, and level of ID).
4) Drop-outs, selection bias.
5) Statistical analysis.
6) Methods of cardiorespiratory fitness measurements.
7) Reliability and/or validity of cardiorespiratory fitness testing.
8) Cardiorespiratory fitness levels ($\dot{V}O_2$ and HR).

The results of the numbers 2, 3, 6, 7, and 8 are for each included article presented in the tables. The results of the numbers 1, 4, and 5 are not presented in the tables, but were used for the interpretation of the study results with respect to representativeness of the study population and possible bias, and the suitability of the statistical analyses to answer the objectives of the study.

To include as many articles as possible, we did not apply any kind of formal weighing or selective procedures in appreciating the results. To help readers interpret the quality of the results, information about the study population and sample sizes are presented with the description of individual studies.

Articles were categorized into two groups. One group with the articles covering topic 1 and 2 (Table 1), and one with articles covering topic 3 (Table 2). Some studies that were included for the review of cardiorespiratory fitness testing (topic 3) are also discussed in the section regarding the cardiorespiratory fitness levels (topic 1 and 2), because they add new information to the included studies in that section.

RESULTS

Thirteen studies were included that reported on cardiorespiratory fitness of individuals with ID (Table 1). One study was a cohort study [20] and one a case-study [21]. All other studies had a cross-sectional design. Two studies used a submaximal bicycle protocol [22, 23], and three used field tests [24-26]. Five studies used a maximal treadmill protocol [17, 21, 27-29]. The other three studies reported on possible determinants for cardiorespiratory fitness without performing an aerobic test [20, 30, 31]. Most studies included adolescents and (young) adults with mild to moderate ID. But three studies included children [20, 27, 31] and three studies included adults over the age of 50 [24, 26, 30]. Three studies also included participants with severe and/or profound ID [23, 24, 30]. These studies are discussed below in the topics 'cardiorespiratory fitness levels' and 'determinants for cardiorespiratory fitness levels'.

Table 1. Cardiorespiratory fitness results, reported for individuals with ID (without DS) as means with standard deviations.

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak ($ml \cdot kg^{-1} \cdot min^{1}$)	HRpeak (bpm)	Outcomes
Hilgenkamp et al. (2012b)	Cross-sectional	**ID:** *n*=81, 51 males, 11 DS Comparison with data of the general population	59±7	Borderline-profound	10m incremental shuttle walking test No familiarization sessions	-	-	None of the participants had an estimated $\dot{V}O_2$max above the lower limit of the average range in the general population.
Vis et al. (2012)	Cross-sectional, repeated measures	**ID no DS:** *n*=25, 15 males **DS:** *n*=96, 46 males **No ID:** *n*=33, 18 males	50±11 42±11 40±11	Mild-severe Mild-profound -	10 knee bends	-	-	Adequate cardiac response to exercise in individuals with ID.
Acampa et al. (2008)	Cohort	**Rett:** *n*=32 females	12±6	NR	None (in rest)	-	-	Sympathetic overactivity in females with high plasma leptin values and parasympathetic overactivity in females with low plasma leptin values.
Baynard et al. (2008)	Cross-sectional	**ID no DS:** *n*=180 **DS:** *n*=133 **No ID:** *n*=322 Each group divided in 4 age categories	9-15 16-21 22-29 30-45	NR	Individualized maximal treadmill protocol: 1) 3.2-5.6 kmh for 4 min at 0% grade 2) 2.5% grade increase every 4 min up to 7.5% 3) 2.5% grade increase every 2 min up to 12.5% 4) 0.8 kmh speed increase every min until exhaustion 1-3 familiarization sessions Objective criteria: $\dot{V}O_2$ plateau, HR plateau, RER>1.0	39.8 40.9 34.4 30.8 (50th percentile scores)	189 183 178 176 (50th percentile scores)	Similar $\dot{V}O_2$peak, except for age group 9-15 years (lower), and age-related changes between individuals with and without ID. Lower HR peak (overall 8 bpm) for individuals with ID.
Bricout et al. (2008)	Case study	**Fragile-X:** 1 male	24	NR	Maximal treadmill protocol: 1) slow speed for 1 min at 0% grade 2) grade and speed alternately increased until exhaustion No familiarization sessions Objective criteria: HR within estimated maximal range, RER>1.0, lactic acid levels	49.5	NR	Abnormal high secretion of cortisol and catecholamines during exercise.

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak ($ml \cdot kg^{-1} \cdot min^{-1}$)	HRpeak (bpm)	Outcomes
Patel et al. (2007)	Cross-sectional	**PWS:** *n*=9, 8 males **No ID:** *n*=9, 8 males	28±3 28±3	NR	Intermittent maximal treadmill protocol: 1) 1 MET workload increase every 2 min. Alternated periods of walking and rest No familiarization sessions Objective criteria: none	-	-	Five individuals with PWS had diminished exercise capacity (achieved 4 METs or less).
Van de Vliet et al. (2006)	Cross-sectional	**Athletes with ID:** *n*=313, 231 males (136 cardiovascular endurance test) Comparison with data of the general population	23±5	NR	20m shuttle run test No familiarization sessions	-	NR	Significantly lower cardiorespiratory endurance (completed laps) for athletes with ID. HRpeak was equal to Canadian controls, but significantly lower than Japanese controls.
Ohwada et al. (2005)	Cross-sectional	**ID males:** *n*=23 **No ID males:** *n*=23	36±9 36±8	Mild-severe	Submaximal bicycle protocol at 40, 50 and 60% of estimated $\dot{V}O_2$ max No familiarization sessions	34.0±9.0	NR	Estimated $\dot{V}O_2$max was significantly lower for individuals with ID. Energy expenditure was higher in individuals with ID, possibly as a result from excessive body movements.
Guideri et al. (2004)	Cross-sectional	**Rett:** *n*=28 females **No ID:** *n*=60	7±3 7±2	NR	None (in rest)	-	-	Low plasma serotonin levels are correlated to sympathetic overactivity.
Baynard et al. (2004)	Cross-sectional	**ID no DS:** *n*=15, 8 males **DS:** *n*=16, 10 males	20±2 21±1	Mild	Individualized maximal treadmill protocol: 1) 3.2-5.6 kmh for 4 min at 0% grade 2) 2.5% grade increase every 4 min up to 7.5% 3) 2.5% grade increase every 2 min up to 12.5% 4) 0.8 kmh speed increase every min until exhaustion 1-2 familiarization sessions Objective criteria: RER>1.0	34.3±2.8	179±4	Increase in HR from rest to submaximal exercise levels was primarily through parasympathetic withdrawal. Appropriate cardiac response.

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak ($ml \cdot kg^{-1} \cdot min^{-1}$)	HRpeak (bpm)	Outcomes
Fernhall et al. (2001)	Cross-sectional	**ID:** *n*=276, 97 DS **No ID:** *n*=296	22±8 31±9	Mild	Individualized maximal treadmill protocol: 1) 3.2-5.6 kmh for 1-3 min at 0% grade 2) 2.5-4.0% grade increase every 1-3 min until exhaustion Familiarization sessions Objective criteria: RER>1.0	33.8±10.6	177±15	The formula 210-0.56(age)-15.5(DS) can estimate HRmax for the ages 8-46 years with similar accuracy as in the general population. Lower $\dot{V}O_2$pweak, HRmax, RERmax, $\dot{V}$Emax for individuals with ID.
Graham & Reid (2000)	Cross-sectional	**ID:** *n*=32, 4 DS, 18 males Comparison with data of the general population	41±10	Mild-moderate	Canadian home fitness step test 1 familiarization session	♂: 33.7±3.8 ♀: 26.3±3.7	NR	Individuals with ID had lower cardiorespiratory fitness. Women significantly lower estimated $\dot{V}O_2$max than men and a greater decline than females in the general population. For men the decline was not as large as in the general population.
Wade et al. (2000)	Cross-sectional	**PWS:** *n*=26, 11 males **No ID:** *n*=26, 11 males	21±10 22±12	NR	2 min submaximal bicycle protocol at HRs of 100-120 bpm No familiarization sessions	-	108±20	Individuals with PWS had lower HR, SBP and DBP, however, they had normal autonomic modulation of HR, during exercise.

n = number of participants; ID = intellectual disability; DS = Down syndrome; Rett = Rett syndrome; Fragile-X = Fragile-X syndrome; PWS = Prader-Willi syndrome; $\dot{V}O_2$ = oxygen uptake; HR = heart rate; RER = respiratory exchange ratio; $\dot{V}$E = ventilation; bpm = beats per minute; NR = not reported; SBP = systolic blood pressure; DBP = diastolic blood pressure.

Cardiorespiratory fitness levels

Fernhall et al. (2001) reported a significantly lower $\dot{V}O_2$peak (33.8 ± 10.6 $ml \cdot kg^{-1} \cdot min^{-1}$) and HRpeak (177 ± 15 bpm) in young adults with ID (n = 276) than in older controls without ID (n = 296, $\dot{V}O_2$peak 35.6 ± 10.9 $ml \cdot kg^{-1} \cdot min^{-1}$, and HRpeak 185 ± 12 bpm), measured with a maximal treadmill protocol.

Ohwada et al. (2005) showed that the estimated $\dot{V}O_2$max, based on results of a submaximal bicycle protocol, was also lower in adult males with ID (n = 23, $\dot{V}O_2$max 34.0 ± 9.0 $ml \cdot kg^{-1} \cdot min^{-1}$) than in controls without ID (n = 23, $\dot{V}O_2$max 41.3 ± 10.0 $ml \cdot kg^{-1} \cdot min^{-1}$). Field tests, i.e. the Canadian home fitness step test and the incremental shuttle walking test, also showed poor levels of cardiorespiratory fitness in (older) adults with ID in comparison to already existing $\dot{V}O_2$max results of the general population [24, 26]. These studies used equations to estimate $\dot{V}O_2$max developed for the general population. However, these equations may not be appropriate for individuals with ID, because they tend to overestimate $\dot{V}O_2$max [16, 32].

Even adult athletes with ID (n = 136) (participants in the INAS-FID 2004 Global Games) showed lower cardiorespiratory fitness, as measured with the number of completed laps on the 20m Shuttle run test, than controls without ID [25]. Peak heart rates were equal to those of Canadian controls, but significantly lower than Japanese controls. Data of these control groups without ID had been derived from population data [25]. When it is not possible to directly compare results to the general population, population data provide the opportunity of comparison. However, this study shows that cultural and regional differences can influence the results.

On the other hand, Baynard, Pitetti, Guerra, Unnithan & Fernhall (2008) concluded that individuals with ID did not have low levels of cardiorespiratory fitness. Only children's (9 to 15 years, n = 59) $\dot{V}O_2$peak (39.8 $ml \cdot kg^{-1} \cdot min^{-1}$) and HRpeak (189 bpm) values were significantly lower than in controls without ID (45.7 $ml \cdot kg^{-1} \cdot min^{-1}$ and 196 bpm). In the older age groups (16 to 21, n = 37; 22 to 29, n = 50; and 30 to 45 years, n = 34) $\dot{V}O_2$peak values (40.9, 34.4, and 30.8 $ml \cdot kg^{-1} \cdot min^{-1}$ respectively) were not significantly different from controls without ID (41.1, 36.8, and 29.9 $ml \cdot kg^{-1} \cdot min^{-1}$). However, the reported $\dot{V}O_2$-peak of the control group in this study was lower than normative values from other studies [10, 33]. Comparing $\dot{V}O_2$peak of the individuals with ID in the study of Baynard et al. (2008) to the ACSM normative values, one would classify individuals with ID as having lower $\dot{V}O_2$peak than the general population, across all the age groups.

Baynard et al. (2008) also showed that $\dot{V}O_2$peak of individuals with ID declined from the youngest to the oldest age group, as in the general population. However, the decrease was not as steep as in controls without ID. Graham & Reid (2000) also studied the decline of cardiorespiratory fitness during adulthood and distinguished between males and females. They found a significant decline in estimated $\dot{V}O_2$max, with results from the Canadian home fitness step test, of middle-aged adults with mild to moderate ID

Table 2. Reliability and validity of cardiorespiratory fitness testing in individuals with ID and test results as means with standard deviations.

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak ($ml \cdot kg^{-1} \cdot min^{1}$)	HRpeak (bpm)	Outcomes
Casey et al. (2012)	Cohort, repeated measures	**DS:** *n*=55, 27 males	11-26	Mild-severe	6MWT No familiarization sessions	-	-	Reliable in individuals with DS with mild (*r* =0.95), moderate (*r* =0.96) and severe (*r* =0.98) ID. A learning effect for the first two tests was found.
Hilgenkamp et al. (2012a)	Cohort, repeated measures	**ID males:** *n*=12 **ID females:** *n*=24	69±10 64±10	Borderline-profound	10m incremental shuttle walking test No familiarization sessions	-	-	Good feasibility (61% participation) and good to high reliability (*r* =0.76-0.90).
Nasuti et al. (2012)	Cohort, repeated measures	**ID:** *n*=13, 4 DS, 7 males	30±8	NR	6MWT & individualized maximal treadmill protocol: 1) 3.2-5.6 kmh for 2 min at 0% grade 2) 2.5% grade increase every 2 min up to 12.5% 3) 0.8 kmh speed increase every min until exhaustion Familiarization sessions Objective criteria: $\dot{V}O_2$ plateau, HR within 85% of estimated HRmax, ventilatory/lactate threshold	32.9±9.8	NR	Reliable test (*r* =0.98) and the maximum walked distance on the 6MWT accounted for 67% of the variance in VO_2peak.
Agiovlasitis et al. (2011)	Cohort	**DS:** *n*=53, 28 males	15±3	Mild-moderate	20m shuttle-run test & individualized maximal treadmill protocol: 1) 2.4-4.0 kmh at 0% grade 2) 2% grade increase every 2 min up to 12% 3) 0.8 kmh speed increase every minute until exhaustion Familiarization sessions *Objective criteria: no*	27.0±5.4	175±11	20m shuttle-run performance did not appear to provide accurate estimation of $\dot{V}O_2$peak (mean group $\dot{V}O_2$peak was accurately estimated, but inaccurate individual prediction).

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak (ml·kg^{-1}·min^{1})	HRpeak (bpm)	Outcomes
Waninge et al. (2011)	Cohort, repeated measures	**ID males:** *n*=29 **ID females**: *n*=18	38±11 44±10	Severe-profound	Modified 10m shuttle run test & 6MWT 2 familiarization sessions	-	Shuttle: 126±20 6MWT: 119±16	6MWT & Shuttle run were feasible (100% participation) and reliable (*r* =0.82-0.96). Shuttle run was not reliable in participants with more severe gross motor impairments.
Vis et al. (2009)	Cross-sectional, repeated measures	**DS with mild or no cardiac disease:** *n*=52, 38 males **DS with severe cardiac disease:** *n*=29, 15 males	41±11 36±10	Mild-profound	6MWT No familiarization sessions	-	-	The 6MWT was not a valid indicator of cardiorespiratory fitness and cardiac restriction in adults with DS. Test-retest reliability was adequate (coefficient of variation was 11%.).
Guerra et al. (2003)	Cohort, cross validation	**DS:** *n*=26, 15 males	15±3	Mild	20m shuttle run test & individualized maximal treadmill protocol: 1) 2.4-4.0 kmh at 0% grade 2) 4% grade increase every 2 min up to 12% 3) 0.8 kmh speed increase every min until exhaustion Familarization sessions Objective criteria: RER>1.0	♂: 27.6±5.1 ♀: 22.7±3.8	♂: 171±12 ♀: 174±10	Estimated $\dot{V}O_2$peak, with a formula developed for children and adolescents with ID without DS, significantly overestimated measured $\dot{V}O_2$peak.
Fernhall et al. (2000)	Cohort, cross validation	**ID:** *n*=17,6 DS, 9 males	14±3	Mild-moderate	20m shuttle run test & individualized maximal treadmill protocol: 1) 2.4-4.0 kmh at 0% grade 2) 4% grade increase every 2 min up to 12% 3) 0.8 kmh speed increase every min until exhaustion Familiarization sessions Objective criteria: RER>1.0	39.4±11.6	182±16	No significant difference between measured and estimated $\dot{V}O_2$peak (*r* =0.86), however, level of agreement was low.

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak (ml·kg^{-1}·min^{1})	HRpeak (bpm)	Outcomes
Pastore et al. (2000)	Cross-sectional	**DS:** *n*=42, 25 males Comparison to data of the general population	10±4	Borderline-severe (2 normal IQ)	Bruce maximal treadmill protocol: 1) 2.7 kmh at 10% grade 2) 0.8-1.4 kmh speed increase and 2% grade increase every 3 min until exhaustion No familiarization sessions Objective criteria: no	-	176±10	84% reasonable or good test compliance. Collaboration during testing depended more on the individuals' mechanical ability to do the technical action required than on their IQ.
Pitetti et al. (2000)	Cross-sectional,repeated measures	**ID males:** *n*=12 **ID females:** *n*=11 **No ID:** *n*=23, 12 males	13±3 14±2 12±3	Mild	Discontinuous maximal treadmill protocol: 1) Four 5 min walking stages at constant speed of 3.2, 4.0, 4.8, and 5.6 kmh alternated with 5 min rest 2) 2.5% grade increase every min up to 12.5% 3) 0.8 kmh speed increase until exhaustion 1-4 familiarization sessions Objective criteria: RER>1.0	♂: 45.7±9.7 ♀: 31.5±6.2	♂: 187±11 ♀: 182±13	Reliability coefficients (*r* =0.85-0.99) of peak physiological parameters similar to those observed in individuals without ID. $\dot{V}O_2$peak and HRpeak of females with ID was significant lower than of females without ID. Only $\dot{V}$Epeak of males with ID was significantly lower than of males without ID.
Draheim et al. (1999)	Cohort, repeated measures	**ID males:** *n*=10 **ID females:** *n*=10	21±3 22±3	Mild-moderate	RFWT & individualized maximal treadmill protocol: 1) individual top safe speed for 2 min at 0% grade 2) 1 min at 2.5% grade 3) 2.5% grade increase every min up to 22% 4) (unknown) speed increase every min until exhaustion Familiarization sessions Objective criteria: $\dot{V}O_2$ plateau, HR plateau, HR within 85% of estimated HRmax, RER>1.0	♂: 41.2±11.2 ♀: 30.8±7.7	♂: 166±38 ♀: 170±23	Reliable results for completion time and end HRs of the RFWT (*r* =0.94-0.91). High correlation between RFWT completion time and $\dot{V}O_2$peak (*r* =-0.73 to -0.75). Low correlation between end HR and measured $\dot{V}O_2$peak (*r* =0.16-0.33). Overestimation of $\dot{V}O_2$peak when estimated from RFWT results.
Mac Donncha et al. (1999)	Cross-sectional,repeated measures	**ID:** *n*=63 males **No ID:** *n*=22 males	16±1 16±1	Mild	20m shuttle run test Familiarization sessions	-	-	Reliable results for completed (*r* =0.94). However, large percentage error of the mean (36.5).

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak (ml·kg^{-1}·min^{1})	HRpeak (bpm)	Outcomes
Pitetti et al. (1999)	Cohort, repeated measures	**ID:** *n*=18, 16 males	15±3	Mild-severe	Individualized maximal treadmill protocol: 1) 4.0-5.6 kmh at 0% grade 2) 4% grade increase every 2 min until exhaustion 1-4 familiarization session Objective criteria: RER>1.0	29.5±4.2	167±20	Reliable results for HRpeak (*r* =0.90), $\dot{V}$E peak (*r* =0.90), RERpeak (*r* =0.88), treadmill time (*r* =0.87). For $\dot{V}O_2$peak test-retest reliability was not as high (*r* =0.77).
Teo-Koh & McCubbin (1999)	Cohort, repeated measures	**ID males:** *n*= 40, 4 DS (24 performed both tests)	14±1	Mild-moderate	RFWT & individualized maximal treadmill protocol: 1) 4.0-5.6 kmh for 2 min at 0% grade 2) 2 min at 2.5% grade 3) 2.5% grade increase every min up to 20% 4) 0.3 kmh speed increase every minute until exhaustion Familiarization sessions Objective criteria: $\dot{V}O_2$ plateau, HR within 90% of estimated HRmax, RER>1.0	41.3±6.4	190±13	Reliable results for the RFWT (*r* =0.87-0.98) and treadmill tests (*r* =0.91-0.87). Significant correlation between $\dot{V}O_2$peak and RFWT completion time (*r* =-.076). Estimating $\dot{V}O_2$peak from RFWT completion time and weight explained 67% of the variance in $\dot{V}O_2$peak.
Fernhall et al. (1998)	Cohort, repeated measures	**ID:** *n*=34, 8 DS, 22 males	14±2	Mild-moderate	600 yard run-walk, 20m and 16m shuttle run test & individualized maximal treadmill protocol: 1) 4.4-7.2 kmh for 2min at 0% grade 2) 4% grade increase every 2 min up to 12% 3) 0.8 kmh speed increase until exhaustion 2-4 familiarization sessions Objective criteria: RER>1.0	36.6±9.1	186±10	Reliable results for the field tests (*r* =0.96-0.98). Field test results were significantly related to $\dot{V}O_2$peak (*r* =-0.80, 0.74 & 0.77). Equations estimating $\dot{V}O_2$peak from field test results explained 74-79% of the variance.

Study	Design	Participants *n*	Age	Level of ID	Exercise test	$\dot{V}O_2$peak (ml·kg^{-1}·min^{1})	HRpeak (bpm)	Outcomes
Rintala et al. (1997)	Cohort, cross validation	**ID males:** n=19	27±8	Borderline-moderate	RFWT & individualized maximal treadmill protocol: 1) 4.0-5.6 kmh for 2 min at 0% grade 2) 2 min at 2.5% or 5.0% grade 3) 2.5% grade increase every min until exhaustion Familiarization sessions Objective criteria: RER>1.0, HR within 85% of estimated HRmax	38.1±8.3	177±13	Reliable results for RFWT completion time (r=0.96) and end HR (r=0.87). Estimating $\dot{V}O_2$peak from RFWT significantly underestimated $\dot{V}O_2$peak in 74-79% of the participants.
Kittredge et al. (1994)	Cohort, repeated measures	**ID males:** n=12 **ID females:** n=13	33±7 34±8	Mild-moderate	RFWT & individualized maximal treadmill protocol: 1) 3.2 kmh for 5 min 0% grade 2) 4.0-4.8 kmh at 0% grade 3) 2.5% grade increase every 2 min until exhaustion 4) Cool-down at 3.2 kmh at 0% grade Familiarization sessions Objective criteria: $\dot{V}O_2$ plateau, HR plateau, RER>1.0	♂: 31.5±7.2 ♀: 27.6±7.0	♂: 175±13 ♀: 177±13	Reliable results for the RFWT completion time (r=0.97) and for end HR (r=0.80).
Climstein et al. (1993)	Cross-sectional	**ID no DS:** n=17, 12 males **DS:** n=15, 12 males	24±3 26±4	Mild	Individualized maximal treadmill protocol: 1) 4.0-4.8 kmh for 2 min at 0% grade 2) 2.5% grade increase every 3 min until exhaustion 2 familiarization sessions *Objective criteria: no* Accuracy of ACSM gender and activity specific regression equations	31.0	175	ACSM equations overestimated $\dot{V}O_2$peak by an average of 129%. Measured HRpeak was on average 21 bpm lower than estimated maximal HR.

n = number of participants; ID = intellectual disability; DS = Down syndrome; $\dot{V}O_2$ = oxygen uptake; HR = heart rate; RER = respiratory exchange ratio; $\dot{V}E$ = ventilation; bpm = beats per minute; NR = not reported; RWFT = Rockport fitness walking test; 6MWT = six-minute walk test; ACSM = American College of Sports Medicine.

over a 13-year period (n = 32). Estimated $\dot{V}O_2$max was higher in males than in females and the decline in males was not as steep as in females. The decline in males was also not as steep as in males in the general population, supporting the finding of Baynard et al. (2008). However, the decline in females with ID was greater than in females in the general population [26]. This study suggests that females have both lower cardiorespiratory fitness levels and a larger decline during adulthood than males. Draheim et al. (1999) (Table 2) supported the finding of gender differences in cardiorespiratory fitness with results from a maximal treadmill test (n = 20). $\dot{V}O_2$peak was higher in males (41.2 ± 11.2 ml·kg^{-1}·min^{-1}) than in females (30.8 ± 7.7 ml·kg^{-1}·min^{-1}). HRpeak was not significantly different: 166 ± 38 bpm in males and 170 ± 23 bpm in females [15].

Differences in cardiorespiratory fitness in ID may be syndrome-specific, as can be seen in individuals with DS [8]. Looking at the few studies of individuals with other genetic syndromes: individuals with Prader-Willi syndrome (PWS) (n = 26) had significantly lower HR during submaximal exercise than controls without ID [22]. Patel et al. (2007) found a subnormal cardiorespiratory fitness in five of nine participants with PWS. However, a case report showed a high $\dot{V}O_2$peak (49.5 ml·kg^{-1}·min^{-1}), above the 75th percentile of $\dot{V}O_2$peak values in the general population, of a 24-year-old male with Fragile-X syndrome [10, 21]. Studies regarding the cardiorespiratory fitness levels of individuals with genetic syndromes, except DS, are scarce and of limited size. Information about specific cardiorespiratory fitness problems in genetic syndromes is therefore limited.

We conclude that studies overall show that individuals with ID have low cardiorespiratory fitness levels, expressed by low $\dot{V}O_2$peak and HRpeak, and these fitness levels are lower than in controls without ID. Females have lower cardiorespiratory fitness levels than males. $\dot{V}O_2$peak of individuals with ID declines with age. However, the decline may not be as steep as in the general population, especially in males. Cardiorespiratory fitness levels may be partly syndrome specific.

Determinants for cardiorespiratory fitness levels

It is often assumed that individuals with ID are not motivated enough and lack task understanding to perform maximally on a maximal exercise test, resulting in low cardiorespiratory fitness results. However, thirteen out of eighteen included studies performing a maximal exercise test used a familiarization protocol and objective criteria to assure maximal effort (see Table 1 and 2), thereby eliminating low motivation and task understanding as a cause for low cardiorespiratory fitness levels found in individuals with ID.

Physical inactivity may be a determinant for these low cardiorespiratory fitness [1, 3]. However, the fact that trained athletes with ID also have low levels of cardiorespiratory fitness [25] suggests that the lack of physical activity may only partly explain low cardiorespiratory fitness and other determinants may also be of influence.

Altered energy expenditure influences the relationship between physical activity and cardiorespiratory fitness. Ohwada et al. (2005) showed that daily activities such as sitting, standing, and walking at 30 and 50 m/min lead to higher energy expenditure in adults with ID than in controls without ID, while energy expenditure in supine position was not different. This higher energy expenditure may be a result of excessive body movements and disturbed gait kinematics [23]. Higher energy expenditure may lead to individuals with ID being less active, because activities will have higher energy cost and thereby negatively impact cardiorespiratory fitness.

Another possible determinant of the low cardiorespiratory fitness levels are the low peak HRs found in individuals with ID and individuals with DS [17, 27]. Since $\dot{V}O_2$max is the product of maximal cardiac output and arterial-venous oxygen difference, and variations in $\dot{V}O_2$max are primarily the result of differences in maximal cardiac output (heart rate [HR] x stroke volume [SV]) [10, 11], it is understandable that a low maximal HR limits $\dot{V}O_2$max. The inability of the heart to increase its rate proportional with increased activity or demand is called chronotropic incompetence [34]. Studies regarding the cardiac response and autonomic modulation of HR in individuals with ID might help to unravel the underlying mechanism of this chronotropic incompetence. However, such studies show conflicting results (Table 1). Vis et al. (2012) found an adequate cardiac response to anaerobic exercise in adults with ID ($n = 25$), whereas adults with DS ($n = 96$) had a diminished cardiac response. Baynard et al. (2004) showed that the increase in HR from rest to submaximal exercise was primarily through parasympathetic withdrawal, both in young adults with ID ($n = 15$) and young adults with DS ($n = 16$). This is an appropriate response from rest to low levels of submaximal exercise. However, since no control group of individuals without ID was included in this study, it is not known if the magnitude of this response was appropriate. Furthermore, in individuals with PWS ($n = 11$) autonomic HR modulation in response to submaximal exercise was also normal, even though HR was lower than in controls without ID [22]. On the contrary, in females with Rett syndrome an altered HR modulation was found. A sympathetic/parasympathetic imbalance was found during rest, which was related to leptin plasma levels ($n = 32$) [20], or low serotonin plasma levels ($n = 28$) [31].

These results support the idea that chronotropic incompetence may play a role in the low cardiorespiratory fitness of individuals with ID. However, autonomic HR modulation seems to be appropriate in individuals with ID and PWS during submaximal exercise. We did not find any studies regarding HR modulation during maximal exercise.

Finally, leg strength has previously been mentioned as a possible determinant to the low cardiorespiratory fitness levels of individuals with ID [7]. We found one study of the relation between leg strength and $\dot{V}O_2$peak [35] (Table 2). This was non-significant, contradicting previous results [7]. However, this study had a small sample size (n = 13), which does not allow drawing conclusions regarding the contribution of leg strength to the low cardiorespiratory fitness of individuals with ID.

In conclusion, familiarization protocols and objective criteria can control for motivational problems and difficulties with task understanding influencing the results of cardiorespiratory fitness testing in individuals with ID. Physical inactivity may partly explain the low cardiorespiratory fitness levels, but chronotropic incompetence is also likely to contribute. However, the underlying cause of the chronotropic incompetence is not yet completely understood. The results regarding leg strength as a possible determinant to low cardiorespiratory fitness are inconclusive.

Reliability and validity of cardiorespiratory fitness testing

Eighteen studies were included that evaluated cardiorespiratory fitness testing in individuals with ID (Table 2). Five studies had a cross-sectional design [32, 36-39], all others were cohort studies. Most studies were performed with children, adolescents, and (young) adults with mild to moderate ID. Three studies included adults over the age of 50 [36, 40, 41]. Six studies included participants with severe to profound ID [36, 39-43]. However, this usually was a small portion of the total study sample. The studies included in this review provide information about the use of equations to estimate $\dot{V}O_2$max in individuals with ID, a continuous and discontinuous maximal treadmill protocol, the Rockport fitness walking test (RFWT), 600-yard run-walk test, shuttle run test, and the six-minute walk test (6MWT).

In the articles discussed below, reliability results refer to test-retest reliability unless mentioned otherwise; the validity of a test was determined by the relationship with objective parameters of maximal testing, such as $\dot{V}O_2$peak.

American College of Sports Medicine (ACSM) equations

Non-invasive and safe procedures to establish $\dot{V}O_2$max would be very useful in the population of individuals with ID. Climstein et al. (1993) showed that ACSM equations, using only information on gender, age, and physical activity level, significantly overestimated $\dot{V}O_2$max by 129% in young adults with ID (n = 17) and 184% in young adults with DS (n = 15) [32]. Therefore, these equations cannot be used to estimate $\dot{V}O_2$max of individuals with ID.

Continuous maximal treadmill protocol

Two studies addressed the use of a continuous maximal treadmill protocol in children and adolescents with ID. Pitetti et al. (1999) showed that cardiorespiratory fitness testing with an individualized continuous maximal protocol is valid and reliable (r = 0.77 to

0.90) in children and adolescents with multiple disabilities (n = 18). Participants, who did not complete the protocol, due to difficulties with walking on the treadmill, breathing through the mouthpiece, and aggressive behavior, had more severe ID and disabilities [43].

Pastore et al. (2000) studied the compliance of children with DS (n = 42) to a continuous maximal treadmill test according to the Bruce protocol (Table 2), and found it to depend more on the individuals' mechanical ability to do the required actions than on their IQ. However, only three participants had severe ID and eight participants had moderate ID, which may have influenced the finding that mechanical ability is more important than IQ. Furthermore, participants did not have the chance to practice the test, which may have led to higher drop-out due to difficulties performing the required actions [39]. Reliability and validity of the Bruce protocol in individuals with ID has not been assessed yet.

These studies show that a continuous maximal treadmill protocol is appropriate for cardiorespiratory fitness testing in children and adolescents with mild to moderate ID. Difficulties performing the actions required during a maximal treadmill test limit the feasibility of treadmill protocols and support the need for familiarization sessions.

Discontinuous maximal treadmill protocol

One study evaluated the use of a discontinuous maximal treadmill protocol in children and adolescents with mild ID and results were found reliable (n = 23) [37]. The treadmill protocol consisted of four 5-minute-walking periods at constant speeds of 3.2, 4.0, 4.8, and finally 5.6 kmh (2.0, 2.5, 3.0, and 3.5 mph) at a 0% grade. Walking periods were alternated with 5 minutes rest. At the end of the fourth walking period, grade was increased by 2.5% every minute until a grade of 12.5% was reached. Then speed was increased with 0.8 kmh (0.5 mph) until exhaustion. Prior to testing the participants followed familiarization sessions. This protocol yielded reliable results of the parameters absolute and relative $\dot{V}O_2$peak, HRpeak, $\dot{V}$Epeak, and RERpeak (r = 0.85 to 0.99), similar or higher to reliability results in controls without ID (r = 0.51 to 0.99) [37].

A discontinuous protocol may have some advantages over a continuous protocol, especially for this population. It allows a plateau in $\dot{V}O_2$peak to occur more often, the mouthpiece can be removed in the resting period, which can overcome problems of participants with the mouthpiece, and (muscle) fatigue will not occur as fast [37].

This discontinuous treadmill protocol may be a good alternative for the continuous protocol. However, whether a discontinuous protocol provides the same results as a continuous protocol for individuals with ID is unknown.

1-mile Rockport fitness walking test

For the 1-mile Rockport Fitness walking test (RFWT) participants have to walk 1 mile (1.6 km) as fast as possible, with a tester walking slightly ahead and verbally encouraging the participant. The RFWT has been found valid and reliable in adolescents and adults with mild to moderate ID [15, 16, 44, 45].

Draheim et al. (1999) evaluated the reliability of RFWT with one tester for five participants (1:5) in young adults with mild to moderate ID (n = 20), to make the test more efficient. The tester walked on the inside of the track and provided encouragement to all five walkers. The 1:5 RFWT showed good reliability for the time needed to complete the walk and HR at the end of the test, r = 0.94 and 0.91 respectively. Furthermore, the mean completion time and mean end HR were not significantly different between the 1:1 RFWT and the 1:5 RFWT, with correlations ranging from r = 0.84 to 0.91 [15, 46].

The original equation of the RFWT developed by Kline et al. (1987) for adults without ID (n = 174, 30 to 69 years) overestimated $\dot{V}O_2$peak in adults with ID [15, 16, 47]. This equation for the general population is therefore not appropriate for individuals with ID.

The equation developed by Teo-Koh & McCubbin (1999) to estimate $\dot{V}O_2$max from the RWFT results for adolescent males with ID explained 67% of the variance in measured $\dot{V}O_2$peak (n = 40). Rintala et al. (1997) cross-validated a previously developed equation to estimate $\dot{V}O_2$max in adult men with ID (n = 19). This equation underestimated $\dot{V}O_2$peak values in 74% to 79% of the participants [45]. However, this same equation overestimated $\dot{V}O_2$peak in adults with ID in the study of Draheim et al. (1999). These results show that population-specific equations should be used with caution, because of the small and selected samples and cross-validation did not yield good results.

Shuttle run test

For the shuttle run test, participants have to repeatedly run a set distance at increasing pace, controlled by an audio signal. The test ends when a participant falls behind the pace by 5 meters [48]. The original 20m shuttle run test was found reliable for children (r = 0.97) [49] and adolescent males (r = 0.94) with mild to moderate ID [38]. Modified versions, such as the 16m shuttle run test, 10m shuttle run test, and the 10m incremental shuttle walking test were also found reliable in children with mild to moderate ID (r = 0.96) [49], adults with severe to profound ID and visual disabilities (r = 0.96) [41], and older adults with borderline to profound ID (r = 0.76-0.90) [40], respectively.

The 16m and 20m shuttle run tests correlated significantly with $\dot{V}O_2$peak (r = 0.77 and 0.74) and developed equations to estimate $\dot{V}O_2$peak explained 77% to 79% of the variance of measured $\dot{V}O_2$peak. Cross-validation of the equation of the 20m shuttle run test showed poor level of agreement between estimated and measured $\dot{V}O_2$peak, with a possible underestimation of 13.5 $ml \cdot kg^{-1} \cdot min^{-1}$ or overestimation of 10.8 $ml \cdot kg^{-1} \cdot min^{-1}$ of measured $\dot{V}O_2$peak [50]. This same equation significantly overestimated $\dot{V}O_2$peak in a sample of adolescents with DS (n = 26) [51]. This may be so because individuals with DS have lower levels of cardiorespiratory endurance than individuals with ID [7, 17, 27, 29]. In another study of boys and adolescent men with DS (n = 53), the 20m shuttle run test did not provide an accurate estimate of $\dot{V}O_2$max, as shown by a low explained variance (23%) of measured $\dot{V}O_2$peak and large errors in individual estimation of $\dot{V}O_2$max [52]. These results show that estimating $\dot{V}O_2$max from the shuttle run test is problematic.

Waninge et al. (2011) found that two familiarization sessions were needed for adults with severe to profound ID and visual disabilities (n = 47) to learn the test protocol and promote optimal performance on the 10m shuttle run test. In the study of Hilgenkamp van Wijck, et al., (2012a), the 10m shuttle walking test was performed twice and a learning effect between the two sessions was lacking, contradicting the need for familiarization sessions [40].

Six-minute walk test

For the six-minute walk test (6MWT), participants have to walk as far as possible in 6 minutes without running or jogging. The 6MWT is reliable in adolescents and adults with mild to severe ID, however validity is ambiguous [35, 41, 42].

Nasuti et al. (2012) reported that the 6MWT was a reliable (r = 0.98) and valid indicator of cardiorespiratory fitness in adults with ID (n = 13) [35]. The walked distance correlated significantly with $\dot{V}O_2$peak, measured with a maximal treadmill test, and accounted for 67% of the variance in $\dot{V}O_2$peak [35]. Participants reached 87.5% of their estimated maximal HR, estimated with the population specific formula of Fernhall et al. (2001). Waninge et al. (2011) showed that after 2 practice sessions the 6MWT was feasible and reliable (r = 0.92) in (older) adults with severe intellectual and sensory disabilities (n = 47). However, this sample on average reached only 69.2% of their estimated maximal HR, which was much lower than in the study of Nasuti et al. (2012) [41].

Furthermore, the 6MWT was reliable in adolescents and young adults with DS (n = 55, r = 0.95 for mild, 0.96 for moderate and 0.98 for severe ID) [42]. A learning effect was found, implying the need for familiarization sessions [42].

Vis et al. (2009) found that the 6MWT was not a valid indicator of cardiorespiratory fitness and cardiac restriction in adults with DS (n = 81). There were no differences in the 6MWT results between adults with DS with and without cardiac disease [36]. However, the group with severe cardiac disease was significantly younger and had lower body mass index than the group without cardiac disease. Together with the lack of practice trials this may have influenced these results.

600 yard run-walk test

For the 600 yard run-walk test, participants have to walk or run 600 yard (550 meters) as fast as possible. Only one study evaluating this test for individuals with ID was found.

In this study a tester walked slightly ahead of the participant and provided verbal encouragement. Prior to testing, the participants received instructions and several practice sessions. The 600 yard run-walk test was found reliable for adolescents with mild to moderate ID (n = 34, r = 0.98) [49] and outcomes significantly correlate to measured $\dot{V}O_2$peak (r = -0.80). The equation to estimate $\dot{V}O_2$max explained 74% of the variance.

In conclusion, continuous and discontinuous maximal treadmill protocols can be used to measure cardiorespiratory fitness in adolescents and adults with mild to moderate ID without severe disabilities. Furthermore, the RFWT, 600 yard run-walk test, shuttle run test, and 6MWT can be used as an indicator of cardiorespiratory fitness in this population. However, estimating $\dot{V}O_2$max from these (submaximal) field tests is problematic.

CONCLUSIONS AND RECOMMENDATIONS

Individuals with ID have low levels of cardiorespiratory fitness, starting with low levels at a young age with further decline with increasing age. Physical inactivity and chronotropic incompetence are most likely to contribute to this low cardiorespiratory fitness. However, this may differ in different genetic syndromes. Good progress has been made in developing appropriate methods to measure cardiorespiratory fitness in this population, but several questions remain to be answered.

Since cardiorespiratory fitness levels are low across the entire population with ID, it is interesting to further investigate possible determinants to low cardiorespiratory fitness levels in this group, including the influence of syndromes. More research is needed regarding the role and mechanisms of chronotropic incompetence and leg strength. Furthermore, studies including objective information about physical activity levels can provide insight in to the role of physical activity in cardiorespiratory fitness levels of individuals with ID. It would be interesting to identify the role of other possible determinants, for example ventilatory response to exercise, ventilatory threshold, lactate levels, and lactate threshold.

Both maximal and submaximal tests have been found valid and reliable for cardiorespiratory fitness testing in this group. However, conflicting results about the validity of the six-minute walk test requires attention. More research is needed regarding the use of submaximal and field tests to estimate $\dot{V}O_2$max. Larger study samples are needed to develop equations, and equations should be cross-validated before use. The advantages of a discontinuous treadmill protocol over a continuous protocol make it interesting to further explore its use and compare the outcomes directly to those of a continuous treadmill protocol.

Gender differences in cardiorespiratory fitness levels, found in both the general population and individuals with ID show that reporting one result for both genders will provide a distorted representation of cardiorespiratory fitness levels [10, 15, 26]. Also, gender differences were found in the age-related decline of cardiorespiratory fitness [26] and it is recommended for futures studies to make a distinction between genders.

Research on older adults and individuals with more severe ID is scarce, thereby limiting the generalizability of results of this review to these groups. Knowledge about the cardiorespiratory fitness levels of older adults with ID will provide insight in age-related decline. This knowledge will also help to identify the threshold below which cardiorespiratory fitness levels will become dangerously low, since it has been suggested that extremely low levels of cardiorespiratory fitness (below the twentieth percentile of normative values) are associated to an increased risk for all-cause mortality [10, 53]. Knowledge about the cardiorespiratory fitness levels of the entire population of ID, including older adults and individuals with more severe ID, will thereby help to determine the need for prevention and intervention. Cardiorespiratory fitness testing may be more difficult for older adults and individuals with more severe ID because of (more severe) physical and cognitive limitations. Therefore, detailed reports of testing procedures and problems encountered during testing in these groups will be helpful for future research.

In conclusion, this review provides an overview of the recent literature about cardiorespiratory fitness levels and testing in individuals with ID, thereby summarizing the state of the current scientific knowledge and identifying areas for future research.

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Chapter 3

Heart rate recovery after the 10-m incremental shuttle walking test in older adults with intellectual disabilities

Alyt Oppewal, Thessa I.M. Hilgenkamp,
Ruud van Wijck, Heleen M. Evenhuis

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ABSTRACT

Heart rate recovery (HRR) after exercise is an independent predictor for cardiovascular and all-cause mortality. To investigate the usefulness of HRR in cardiorespiratory exercise testing in older adults with intellectual disabilities (ID), the aims of this study were (a) to assess HRR in older adults with ID after the 10-meter incremental shuttle walking test (ISWT) and (b) its association with personal characteristics (gender, age, distance walked on the ISWT, level of ID, genetic syndrome causing ID, autism, behavioral problems, and peak heart rate [HRpeak]). HRR was assessed after the 10-meter incremental shuttle walking test in 300 older adults (> 50 years) with borderline to profound ID. HRR was defined as the change from HRpeak during the ISWT to heart rate measured after 1, 2, 3, 4, and 5 minutes of passive recovery.The largest decrease in heart rate was in the first minute of recovery leveling off toward the fifth minute of recovery. An abnormal HRR (≤ 12 bpm) was seen in 36.1% of the participants with Down syndrome (DS) and in 30.7% of the participants with ID by other causes. After the fifth minute the heart rates of 69.4% of the participants with DS and of 61.4% of the participants with ID by other causes returned to resting levels. HRpeak and distance walked on the ISWT were positively related to all HRR measures. More severe ID was negatively related and having DS positively related to HRR after 3 to 5 minutes of recovery. The other characteristics were not significantly associated to HRR. HRR is a useful outcome measure in cardiorespiratory fitness testing of older adults with ID with a direct, objective, and non-invasive measurement. Further research is needed to identify the relation between HRR and adverse health outcomes in this population.

INTRODUCTION

After exercise, heart rate returns to resting levels due to the combination of parasympathetic reactivation and sympathetic withdrawal, and therefore reflects the activity of the autonomic nervous system [1]. A delayed heart rate recovery (HRR) after exercise is an independent predictor for cardiovascular events and all-cause mortality in healthy adults and those with cardiovascular diseases and systemic disorders, such as diabetes mellitus and hypertension [2-6]. Because increasing age is a risk factor for cardiovascular diseases, assessment of HRR in older adults is an important outcome measure in cardiorespiratory fitness testing of older adults [7-9].

Despite of its clinical importance, HRR is little used in cardiorespiratory fitness evaluations of individuals with intellectual disabilities (ID). The few studies of HRR in adults with Down syndrome (DS) showed that individuals with DS had lower peak heart rates and lower HRR than controls with normal intelligence [10-12]. However, most studies regarding cardiorespiratory fitness of individuals with ID have focused on heart rate response during exercise and maximal oxygen uptake ($\dot{V}O_2$max). These studies show that individuals with ID have poor cardiorespiratory fitness and low peak heart rates (HRpeak), especially individuals with DS [13-15]. This may be because of an altered autonomic control [14, 16-18]. HRR may therefore be an interesting outcome measure in cardiorespiratory fitness testing of individuals with ID.

An additional advantage of HRR could be that it may not be necessary for participants to perform maximal or close to maximal exercise, which is required for measurement of $\dot{V}O_2$max. This may be especially useful for older adults with ID, because in the 'Healthy ageing and intellectual disabilities' (HA-ID) study we found that 61% of the participants did not achieve 85% of their estimated maximal heart rate (HRmax) during the 10-meter incremental shuttle walking test (ISWT) [19] which was a criterion to validly estimate $\dot{V}O_2$max [20]. Another problem with estimating $\dot{V}O_2$max is that the available equations are problematic for use in individuals with ID [15]. Although exercise intensity has been mentioned as a factor influencing HRR [21-23], HRR after 1 minute has been found equal after exercise at 65% and 85% of HRmax [24]. However, HRR after 2 and 3 minutes of recovery did differ [24]. The prognostic value of HRR for cardiovascular events and all-cause mortality has also been found to not depend upon maximal effort [6, 25, 26]. In addition, Mendonca & Pereira (2010) suggested that the attenuated HRR of individuals with DS was independent of their low HRpeak, supporting the idea that HRpeak achieved during the test may be less important for HRR than it is for $\dot{V}O_2$max.

Therefore, to investigate the usefulness of HRR in cardiorespiratory exercise testing in older adults with ID, the aims of this study were to assess (a) HRR in older adults with ID after the 10-meter incremental shuttle walking test and (b) its association with personal characteristics that are known in the general population to influence HRR (age, gender, cardiorespiratory fitness [expressed as distance walked on the ISWT]) [3], specific characteristics of the ID population that may influence HRR because of a possible influence on the autonomic nervous system (level of ID [27], genetic syndrome causing ID [14, 15, 28], autism spectrum disorder [29], behavioral problems [30]), and HRpeak.

METHODS

Study design and participants

This study was part of the large Dutch cross-sectional HA-ID study conducted by a consort consisting of three ID care-provider services in collaboration with two university departments (Intellectual Disability Medicine, Erasmus MC, University Medical Center Rotterdam and the Center for Human Movement Sciences, University of Groningen, University Medical Center Groningen). All 2150 older clients with ID (≥ 50 years) were invited to participate, resulting in a near-representative sample of 1050 clients. Details about design, recruitment, and representativeness of the sample have been presented elsewhere [31]. Of the HA-ID study sample, 654 older adults performed the cardiorespiratory fitness measurements. Older adults (70 – 79 years) and participants with more severe ID and mobility impairments were underrepresented with respect to the total HA-ID sample (n = 1050) [19], limiting the generalizability to these groups.

For the current study, individuals who had medical conditions and/or used medication that may alter heart rates, and/or in whom information about the presence of Down syndrome (DS) was missing –which is necessary to calculate HRmax– were omitted from the analyses.

Ethical approval was provided by the Medical Ethical Committee at Erasmus Medical Center (MEC 2008-234) and by the ethical committees of the participating ID care-provider services. Informed consent was obtained from all participants or their legal representatives; however, unusual resistance was a reason for aborting measurements at all times. This study followed the guidelines of the Declaration of Helsinki [32].

Measurements

Personal characteristics

Gender and age were collected from the administrative systems of the ID care-provider services. Level of ID was categorized by behavioral therapists or psychologists as borderline (IQ = 70 – 84), mild (IQ = 50 – 69), moderate (IQ = 35 – 49), severe (IQ = 20 – 34), or profound (IQ < 20) [33]. The presence of a genetic syndrome, autistic spectrum disorder, and behavioral problems were obtained from medical and behavioral therapists' records.

To describe the study population in detail, the following information was collected. Professional caregivers gave information about mobility (independent, with walking aid, or wheelchair-bound). Height and weight were measured by trained medical assistants during physical examination. Height was measured with a stadiometer (Seca type 214) with the participant wearing no shoes. Weight was measured with a digital floor scale (Seca robusta type 813) with participants wearing light clothes and no shoes. Body mass index (BMI) was calculated by weight divided by squared height [34]. Physical activity was measured with a pedometer (NL-1000 pedometer, New Lifestyles, Missouri, USA) and a minimum of 7500 steps per day was classified as sufficient. Detailed methods have been described elsewhere [35].

Exclusion criteria

To exclude participants with medical conditions and/or using medications that may alter heart rates, information about relevant medical conditions (arrhythmias, pacemaker, thyroid dysfunction), use of medication (beta blocking agents, digitalis glycosides, vasodilators, calcium channel blockers, thyroid therapy drugs, anti-Parkinson drugs, obstructive airway therapy drugs, and antihypertensives) were retrieved from medical files.

Cardiorespiratory fitness

Cardiorespiratory fitness was measured with the 10-meter incremental shuttle walking test (ISWT) [36]. Participants had to repeatedly walk up and down a 10 m course at increasing pace. An instructor accompanied the participant and set the pace. The starting speed was 0.50 m/s. Every minute, the instructor increased walking speed by 0.17 m/s, according to the test protocol. The test ended when either the participant was too breathless to maintain the required pace or failed to complete a 10 m shuttle within the time allowed [36]. The distance covered was the test result. The ISWT results are reproducible ($r \geq 0.98$) in patients with chronic airway construction [36] and in patients attending cardiac rehabilitation (ICC = 0.94) [37]. Validity has been confirmed in patients with chronic airway construction with the relation between the distance walked on the ISWT and $\dot{V}O_2max$ during maximal treadmill exercise ($r = 0.88$) and increasing oxygen consumption was found in response to increasing intensity of the ISWT [20]. The ISWT is feasible in older adults with ID, except for individuals with profound ID and, obviously, wheelchair users [38]. Also, test-retest reliability was good (ICC of 0.90 [same day interval] and 0.76 [2-week interval]) [39].

Because earlier studies suggested the need for a practice session to obtain valid results [36, 37, 40], the ISWT was performed twice. The ISWT test with the best effort, i.e. in which the participant reached the highest heart rate (HRpeak), was defined as the best test; heart-rate data of this test was used in the analyses.

Heart rates and heart rate recovery

Heart rates were monitored with wireless heart-rate monitors (Suunto T6c) directly before the test (HRrest), continuously during the ISWT, and during a 5-minute passive recovery period in which participants sat quietly.

Heart rate recovery (HRR) after exercise was defined as the change from peak heart rate (HRpeak) during the ISWT to that measured after 1, 2, 3, 4 and 5 minutes of recovery; HRR1 = HRpeak – $HR_{1\text{-min recovery}}$ and HRR2 = HRpeak – $HR_{2\text{-min recovery}}$ etc. A decrease in heart rate equal to or less than 12 beats per minute (bpm) during the first minute of recovery was considered abnormal [7, 26, 41].

The exercise intensity of the ISWT was assessed with peak heart rate and the percentage of age-predicted HRmax (%HRmax) achieved during the ISWT. HRmax was calculated with the population-specific formula: HRmax = 210 – (0.56 x age) – (15.5 x DS), with non-DS coded as 1 and DS coded as 2 [42]. The percentage of HRmax achieved was calculated with the following formula: %HRmax = (maximal heart rate during the ISWT / HRmax) x 100%).

Procedure

Data collection was part of an extensive physical fitness assessment that was conducted at locations familiar or close to participants: a large room within their home, a familiar daycare center, or a gym. Assessments were guided by instructors, who all were physiotherapists, occupational therapists, or physical activity instructors with experience with individuals with ID. All received an instruction manual and followed two days of training on conducting the fitness tests. To avoid undesirable influences of consecutive ISWT tests, both ISWTs were conducted with at least one hour in between.

The standardized encouragement provided by test instructions for testing individuals with normal intellectual capabilities is unsuitable for individuals with ID. To keep this motivational aspect as equal as possible, we prescribed 'maximal motivation' to the test instructors for all tests. In some cases, this meant that participants were motivated to engage in the assessments by constant verbal encouragement and verbal rewarding. In other cases the test instructor had to remain very calm and quiet to motivate the participant as much as possible and to prevent stress or anxiety. The specific background, knowledge, and experience of the test instructors were important conditions for ensuring the most suitable 'maximal motivation' for every participant, while regarding safety as well.

To assure safety during testing, the Revised Physical Activity Readiness Questionnaire (rPAR-Q) was administered by professional caregivers in advance of participation [43, 44]. If any of the questions were answered with 'yes' or 'unknown', the medical physician was consulted to determine whether it was safe for the participant to perform the ISWT. If not, the participant was excluded.

Statistical analyses

Differences in personal characteristics between individuals omitted from the study due to the exclusion criteria, and participants remaining in the study were checked with independent *t* tests and Pearson's chi-square tests.

If categories of categorical variables contained ≤ 5% of the study sample, these categories were compared with remaining categories, using independent *t* tests and Pearson's chi-square tests, to determine if they could be grouped together.

Characteristics of the remaining participants were described for participants with DS and participants with ID by other causes separately, because of established differences in cardiorespiratory fitness and exercise response [14, 42]. Descriptive statistics were calculated for distance walked, duration of the ISWT, HRrest, HRpeak, %HRmax, heart rates during the recovery period, and HRR1 to HRR5. The number of participants with an abnormal HRR1 (≤12 bpm) was calculated. Differences between participants with DS and ID by other causes were analyzed with independent *t* tests and Pearson's chi-square tests.

To check whether the heart rates of participants returned to their resting heart rates after exercise, the difference between resting heart rates and heart rates during recovery was analyzed. When differences were smaller than 5 bpm, participants were considered to have returned to resting heart rates.

A multiple linear regression model was used to determine the association of personal characteristics with HRR. Categorical variables with more than two categories were recoded into dummy variables. HRR1 to HRR5 were used as dependent variables. Independent variables known to be relevant were entered in the first block (age [in years], gender [male = 0, female = 1], and distance walked on the ISWT [in meters]). Other independent variables were placed in the second block with the forward method (level of ID [borderline, mild, moderate, severe, and profound], DS [no = 0, yes = 1], autism spectrum disorder [no = 0, yes = 1], behavioral problems [no = 0, yes = 1], and HRpeak [in bpm]), resulting in a model with only variables that contribute significantly to the predictive power of the model. Multicollinearity was checked with the Variance Inflation Factor (VIF), which had to be below 10 for all independent variables [45]. Results are presented as the unstandardized coefficients (*B*), representing the strength of the relation between each independent variable and the outcome in units of the independent variable; its standard error (SE *B*), the standardized beta (β), representing the strength of the relation between each independent variable and the outcome in standardized units; and the explained variance (adjusted R^2).

Statistical significance was set at $p < 0.05$. Bonferroni correction was used to correct for the inflated familywise error rate of multiple testing. Analyses were performed with the Statistical Package for Social Sciences (SPSS) version 20.0 (IBM Corporation, New York).

RESULTS

Participants

Out of 654 older adults with ID who successfully participated in the ISWT measurements (study population is described elsewhere [19]), 354 were omitted from the analysis because of medical conditions (arrhythmias [n = 12], pacemaker [n = 3], and thyroid dysfunction [n = 73], medication use (beta blocking agents [n = 44], vasodilators [n = 5], calcium channel blockers [n = 18], thyroid therapy drugs [n = 71], anti-Parkinson drugs [n = 24], obstructive airway therapy drugs [n = 37], and antihypertensives [n = 1]), and missing or inconsistent information regarding the presence of such medical conditions

Table 1. Characteristics of participants with intellectual disabilities by other causes than Down syndrome and participants with Down syndrome.

Characteristics	*n* (%)	ID	DS
Total		264	36
Age	Years (m ± sd)	60.5 ± 7.3**	56.1 ± 5.4**
Gender	Female	118 (44.7)	11 (30.6)
	Male	146 (55.3)	25 (69.4)
Height	cm (m ± sd)	165.4 ± 10.3**	153.7 ± 9.4**
Weight	kg (m ± sd)	72.4 ± 14.6**	61.2 ± 8.6**
BMI	kg/m² (m ± sd)	26.5 ± 4.9	26.0 ± 3.7
Level of ID	Borderline - mild	70 (26.5)	2 (5.6)
	Moderate	132 (50)	22 (61.1)
	Severe - profound	56 (21.2)	12 (33.3)
	Unknown	6 (2.3)	0
Autism spectrum disorder	Yes	65 (24.6)	3 (8.3)
	No	181 (68.6)	33 (91.7)
	Unknown	18 (6.8)	0
Behavioral problems	Yes	78 (29.5)	5 (13.9)
	No	166 (62.9)	29 (80.6)
	Unknown	20 (7.6)	2 (5.6)
Mobility	Independent	221 (83.7)	34 (94.4)
	Walking aid	37 (14.0)	2 (5.6)
	Unknown	6 (2.3)	0
Physical activity	> 7500 steps/day	40 (15.2)	1 (2.8)
	< 7500 steps/day	61 (23.1)	7 (19.4)
	Unknown	163 (61.7)	28 (77.8)

m = mean; *sd* = standard deviation; *n* = number of participants; ID = intellectual disability by other causes than Down syndrome; DS = Down syndrome; BMI = body mass index.
** $p < 0.01$

(n = 102) or DS (n = 113). Finally, 63 individuals were omitted because of problems with the heart rate registration. The omitted individuals had a higher BMI ($t(573) = 3.90$, $p <$ 0.001) and less often autism spectrum disorder (χ^2 [1, n = 591] = 8.831, p = 0.003) than the 300 participants remaining in the further analyses.

Six participants had Fragile X syndrome, which was less than 5%. This group was not significantly different from the other participants with ID, therefore they were taken together in the analyses. The groups of participants with borderline (n = 12) and profound (n = 15) ID were also small and not significantly different from the groups mild and severe ID, respectively. Therefore, the borderline and mild ID groups and severe and profound ID groups were combined as well.

The characteristics of the remaining 300 participants are shown in Table 1. Participants with DS were significantly younger ($t(54.31) = 4.39$, $p < 0.001$), shorter ($t(293) = 6.22$, p < 0.001), and weighed less ($t(292) = 4.29$, $p < 0.001$) than participants with ID by other causes (Table 1). The other characteristics did not differ significantly.

Heart rates and heart rate recovery

The range of the percentage of HRmax achieved (%HRmax) during the ISWT was wide (Figure 1). Participants exerted themselves from 42.7% to 108.8% of their age-predicted HRmax.

The heart rates and heart rate recovery results are shown in Table 2 and Figure 2. Participants with DS had significantly lower resting heart rates ($t(296) = 3.05$, $p = 0.002$) and

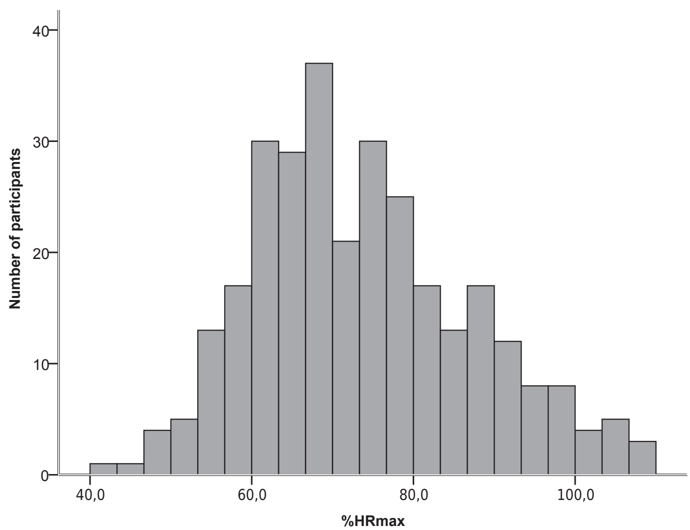

Figure 1. Distribution of the percentage of age-predicted maximal heart rate achieved (%HRmax) during the 10-meter incremental shuttle walking test.

Table 2. Heart rate and heart rate recovery results of participants with intellectual disabilities by other causes than Down syndrome and those with Down syndrome, presented as means with 95% confidence intervals.

	ID	DS	*p*
Distance walked (m)	264.4 [241.6, 287.2]	196.1 [151.4, 240.8]	0.008
Duration ISWT (min:s)	5:16 [4:59, 5:34]	4:27 [3:50, 5:05]	0.046
HRrest (bpm)	82.4 [80.7, 84.2]	74.6 [69.2, 80.0]	0.002*
HRpeak (bpm)	118.5 [115.8, 121.2]	109.6 [102.8, 116.3]	0.022
%HRmax	73.7 [72.1, 75.3]	74.3 [69.6, 79.0]	0.808
HRR1 (bpm)	20.2 [18.4, 21.9]	16.0 [12.1, 19.8]	0.110
HRR2 (bpm)	28.4 [26.3, 30.4]	25.2 [20.8, 29.7]	0.294
HRR3 (bpm)	31.0 [28.8, 33.1]	29.9 [24.7, 35.2]	0.742
HRR4 (bpm)	32.3 [30.1, 34.4]	31.8 [26.9, 36.7]	0.876
HRR5 (bpm)	33.0 [30.7, 35.4]	33.0 [28.2, 37.8]	0.988

ID = intellectual disability by other causes than Down syndrome; DS = Down syndrome; ISWT = 10-meter incremental shuttle walking test; HRrest = heart rate prior to the ISWT; HRpeak = peak heart rate achieved during the ISWT; %HRmax = percentage of age-predicted maximal heart rate reached during the ISWT; HRR1 = heart rate recovery at 1 minute after exercise; HRR2 = heart rate recovery at 2 minutes after exercise; HRR3 = heart rate recovery at 3 minutes after exercise; HRR4 = heart rate recovery at 4 minutes after exercise; HRR5 = heart rate recovery at 5 minutes after exercise; bpm = beats per minute.
* Significant difference between DS and ID, $p < 0.005$ (Bonferroni correction 0.05/10).

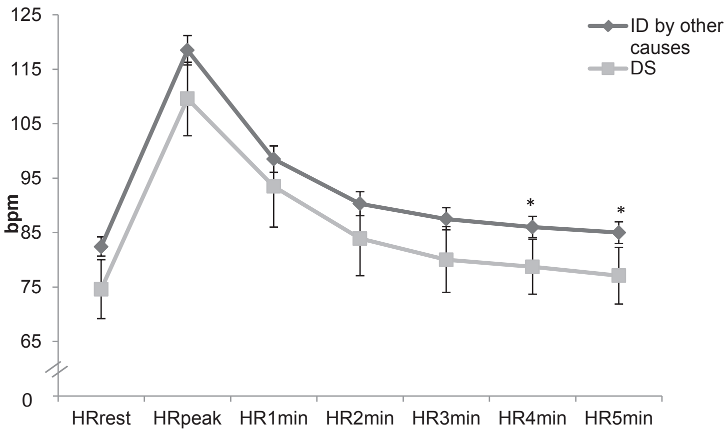

Figure 2. Resting heart rate (HRrest), peak heart rate (HRpeak), and heart rates after 10-meter incremental shuttle walking test (HR1min-HR5min) of participants with intellectual disabilities by other causes than Down syndrome (ID) and participants with Down syndrome (DS), presented as means with 95% confidence intervals.

Table 3. Results of the multiple regression analyses for heart rate recovery after each minute of recovery.

		B	SE *B*	*β*	Adjusted R^2
HRR1 (*n* = 244)					0.281
	Age	0.056	0.113	0.028	
	Female	-0.475	1.600	-0.017	
	Distance walked	0.015	0.005	0.200*	
	HRpeak	0.253	0.042	0.4206*	
HRR2 (*n* = 237)					0.436
	Age	0.074	0.113	0.033	
	Female	1.110	1.642	0.035	
	Distance walked	0.021	0.005	0.255**	
	HRpeak	0.351	0.042	0.497**	
HRR3 (*n* = 241)					0.519
	Age	0.180	0.116	0.075	
	Female	1.198	1.598	0.036	
	Distance walked	0.019	0.005	0.216**	
	HRpeak	0.431	0.042	0.576**	
	More severe ID	-4.553	1.839	-0.114*	
	Down syndrome	5.696	2.298	0.115*	
HRR4 (*n* = 241)					0.596
	Age	0.158	0.105	0.066	
	Female	0.948	1.479	0.028	
	Distance walked	0.022	0.005	0.249**	
	HRpeak	0.459	0.039	0.607**	
	More severe ID	-4.019	1.704	-0.099*	
	Down syndrome	6.126	2.102	0.124**	
HRR5 (*n* = 239)					0.589
	Age	0.198	0.108	0.082	
	Female	2.055	1.534	0.060	
	Distance walked	0.021	0.005	0.235**	
	HRpeak	0.474	0.040	0.615**	
	More severe ID	-4.712	1.745	-0.115**	
	Down syndrome	7.480	2.139	0.151**	

n = number of participants; HRR = heart rate recovery; HRpeak = peak heart rate achieved during the ISWT; ID = intellectual disability; HRR1 = heart rate recovery at 1 minute after exercise; HRR2 = heart rate recovery at 2 minutes after exercise; HRR3 = heart rate recovery at 3 minutes after exercise; HRR4 = heart rate recovery at 4 minutes after exercise; HRR5 = heart rate recovery at 5 minutes after exercise; bpm = beats per minute.

Age (in years), gender (male = 0, female = 1), distance walked (in meters), level of ID (borderline-mild = 0, moderate =1, severe-profound = 1), DS (no = 0, yes = 1), and HRpeak (in bpm).

* $p < 0.05$

** $p < 0.01$

lower heart rates after 4 and 5 minutes of recovery ($t(272) = 2.62$, $p = 0.009$ and $t(270) = 2.81$, $p = 0.005$) than those with ID by other causes. The other heart rate variables did not differ significantly. The largest decrease in heart rate was in the first minute of recovery, leveling off toward the fifth minute of recovery (Table 2 and Figure 2). An abnormal HHR after 1 minute (≤ 12 bpm) was seen in 36.1% of the participants with DS and in 30.7% of the participants with ID by other causes (no significant difference).

After the first minute of recovery, the heart rates of 19.4% of the participants with DS and of 27.3% of the participants with ID by other causes were returned to resting levels. After the second minute this was 48.5% and 38.9%, after the third minute 55.3% and 52.8%, after the fourth minute 57.6% and 58.3%, and after the fifth minute the heart rates of 61.4% of the participants with ID by other causes than DS and 69.4% of the participants with DS were returned to resting levels.

Multiple regression analyses

The results of the multiple regression analysis for HRR1 to HRR5 are presented in Table 3. The explained variance ranges from 28.1% (HRR1) to 59.6% (HRR4).

Participants who walked a larger distance and participants with a higher HRpeak during the ISWT had a higher HRR. For HRR3 to HRR5, participants with a more severe ID had a lower HRR, and participants with DS had a higher HRR.

DISCUSSION

This study was the first to assess heart rate recovery (HRR) in a large sample of older adults with intellectual disabilities (ID) using the 10-meter incremental shuttle walking test (ISWT). At first glance, the trajectory of HRR in older adults with ID does not seem to differ much from the general population, with the largest decrease in heart rate during the first minutes of recovery [3]. One third of the older adults with ID had an abnormal HRR after 1 minute (≤ 12 bpm). This is comparable to the general adult population [4, 25, 26]. However, this result is dependent on the cut-off score used, and different cut-off scores for an abnormal HRR are reported in literature [25, 26, 46, 47]. Together with the lack of normative data and different recovery protocols (passive versus active, sitting, lying) used across studies affecting HRR results [22], this hampers the interpretation of our results.

In addition, cut-off scores for an abnormal HRR have been based on the predictive value of HRR for cardiovascular and all-cause mortality in the general population. The mechanism behind the association between HRR and mortality may be an altered autonomic modulation [48, 49]. There are reasons to suppose that autonomic control is different in individuals with ID, which might imply a different relationship of HRR and health in this group. Blunted parasympathetic withdrawal and reduced sympathetic activation during exercise have been found in individuals with DS [14] and a parasympathetic / sympathetic imbalance during rest has been found in females with Rett syndrome [17, 18]. A diminished vagal withdrawal during isometric handgrip exercise has also been found in individuals with ID without specific genetic syndromes [16]. It seems that an altered autonomic control may be associated with the intellectual disability. This is supported by our finding that the level of ID and the presence of DS were related to HRR, and gender and age were not, in contrary to the general population [3]. Different cut-off score may therefore be needed for the ID population underscoring the necessity of specific studies of predictive validity of HRR.

In this study, the mean HRR of participants with DS after the first minute of recovery (16.0 bpm, 95% CI [12.1, 19.8]) and after the second minute of recovery (25.2 bpm, 95% CI [20.8, 29.7]) were around 9 and 13 bpm lower than those found in previous studies with individuals with DS [10, 11]. This may be caused by differences in age of the study samples and differences in exercise and recovery protocols [10, 11]. Standardization of assessment of HRR in future studies will improve comparison across studies.

Four characteristics were significantly associated with HRR. First, HRpeak had the strongest positive association with HRR, indicating that HRR may be underestimated if participants do not exert themselves fully. Therefore, using HRR as an outcome measure for cardiorespiratory fitness does not solve the problem of low numbers of participation due to too little exertion [19]. Full exertion is difficult for (older) adults with ID, due to little experience with exercise and low physical activity levels [35]. It is therefore important to implement exercise and physical activity in their daily life, allowing them to become familiar with the signs of exercise, hopefully resulting in better fitness and participation in cardiorespiratory fitness testing. Second, the distance walked was positively related to HRR. Since the distance walked during the ISWT is a measure of cardiorespiratory fitness [20], this result provides convergent validity for HRR as an indirect measure of cardiorespiratory fitness of older adults with ID. Third, having DS was positively related to HRR in the third, fourth, and fifth minute of recovery. This was unexpected because it is consistently seen that individuals with DS have poorer cardiorespiratory fitness than individuals with ID by other causes [13-15]. We do not have an explanation for this finding and further research is needed to see if this finding is consistent and what the possible mechanisms might be. Finally, having more severe ID was negatively associated with HRR during the last three minutes of recovery, suggesting that HRR is influenced by the intellectual disability. This is in line with the findings of Keary et al. (2012) that HRR was related to poorer cognitive function. However, the underlying mechanisms are currently unknown, so further research is necessary in this area as well.

A limitation of this study was that we did not have the actual maximal heart rates of the participants and used population-specific age-predicted HRmax to get insight in the intensity of the ISWT. However, the formula we used to calculate HRmax may not be adequate for older adults with ID, because we found actual HRpeak to be higher than calculated age-predicted HRmax in twelve participants. This formula was developed with a study sample of individuals with ID ranging from 9 to 46 years, so no older adults were included in this study and the formula may therefore be not suitable for this group [42]. In addition, the formula predicted only 32.5% of the variance in HR [42]. Unfortunately, a better alternative is lacking. Further research is needed to provide a more adequate formula for older adults with ID.

The participants only differed in BMI and prevalence of autism spectrum disorder in comparison to those omitted from the analyses, and these characteristics did not influence HRR [10, 11]. Therefore, the results of this study are likely to be representative for the sample of 654 participants that performed cardiorespiratory fitness testing in the HA-ID study [19].

Although using HRR as an outcome measure does still require participants to perform maximal or close to maximal exercise, and reference data and standardized protocols are lacking, HRR is a direct, objective measurement that can be assessed during field cardiorespiratory fitness testing. HRR also provides information about autonomic control without invasive measurement. Therefore, HRR is a potentially relevant outcome in cardiorespiratory fitness testing in older adults with ID, but more research is needed to identify the relation between HRR and adverse outcomes and the reliability of HRR measurements.

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Chapter 4

The effect of handedness on grip strength in older adults with intellectual disabilities

Alyt Oppewal, Thessa I.M. Hilgenkamp, Ruud van Wijck, Heleen M. Evenhuis

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ABSTRACT

Grip strength is an important predictor of several health outcomes in the general older population. Grip strength assessment is feasible and reliable in older adults with intellectual disabilities (ID), which makes it a valuable measurement for application in this population. The purpose of this study was to investigate the effect of handedness on grip strength in the older population with ID. Handedness and grip strength were assessed in a sample of 1050 older adults (≥ 50 years) with borderline to profound ID. Results showed that 26.2% of the study sample was left-handed. In right-handed participants the dominant hand (right) was on average 8.7% stronger than the non-dominant hand ($p < 0.001$). For left-handed participants there was no significant difference between the dominant hand (left) and non-dominant hand. However, more detailed analyses revealed that 34.5 % of the participants were stronger with their non-dominant hand, (on average 16.6% stronger for right-handed and 16.3% stronger for left-handed participants). Because of the large strength ratios, distributed in favor of both the dominant as the non-dominant hand, it is recommended to assess both hands to get a valid result of grip strength in older adults with ID.

INTRODUCTION

Grip strength is not only a measurement of hand functioning, but also characterizes overall upper extremity muscle strength [1] and correlates with lower extremity strength and power [2, 3]. It is an important marker in the assessment of sarcopenia [2], nutritional status [4], frailty [5], and muscular strength as a component of physical fitness [6, 7]. Grip strength is a predictor of premature mortality, earlier onset of disability, postoperative complications, increased length of hospital stay [8], fractures, and cognitive decline in older adults [9, 10].

Because of the simple, inexpensive, non-invasive, and quick measurement of grip strength by the use of a hand dynamometer, it is increasingly being used in clinical settings, such as in geriatric practice [2]. Measuring grip strength with a hand dynamometer was found to be feasible and reliable in older adults with intellectual disabilities (ID) [11]. Therefore, it might be valuable to introduce this measurement into this population as a marker for sarcopenia, nutritional status, frailty, and physical fitness.

In assessing grip strength it is important to consider possible differences between the strength of the dominant and the non-dominant hand. By dividing the strength of the dominant hand by the strength of the non-dominant hand, the resulting strength ratio describes the differences between both hands. The '10% rule' states that the dominant hand is generally 10% stronger than the non-dominant hand [12, 13]. Clerke & Clerke (2001) reviewed the literature regarding handedness (which hand is the dominant hand) and grip strength. They found a wide individual variation in reported strength ratios of 0% to 40%. Also, strength ratios were often larger for right-handed individuals than for left-handed individuals [14, 15]. Thus, the '10% rule' does not seem to hold as a general guideline [14]. Bohannon (2003) reported that right-handed individuals were stronger on the dominant side, but for left-handed individuals results were equivocal [16]. These results suggest that it is important to consider handedness in the assessment of grip strength.

ID has been linked to atypical distribution of right- and left-handedness in comparison to the general population [17-19]. Handedness was less skewed to the right in children [17-20] and adults with ID [21-23]. Furthermore, the incidences of left- and mixed-handedness are about twice those reported for samples with normal intellectual capabilities [17]. The frequency of right-handedness decreased with the severity of ID [23, 24]. No studies concerning the effect of handedness on grip strength measurements in adults with ID have been performed, to the knowledge of the authors.

In clinical practice, it is not standard to consider the strength of both hands to be different. The American Medical Association (AMA) states in its *Guides to the Evaluation of Permanent Impairment* that "little evidence exists that there is a significant difference in grip strength between the dominant and non-dominant hand" [25]. If one of both hands is affected, the AMA recommends comparison of grip strength scores of the affected hand with the uninjured hand; thereby assuming both hands could be equally strong [25]. This is also recommended by The American Society of Hand Therapists (ASHT) [26].

Before introducing grip strength measurement into routine diagnostic work-up of older adults with ID, more information is required about the effect of handedness on grip strength in this population to give recommendations about the best way to measure grip strength and interpret the results. Questions to be answered were whether it is sufficient to measure a single hand, whether the dominant hand needs to be determined, or if it is necessary to always measure both hands. Because of the association of left-handedness with more severe ID [23, 24] it is important to take different levels of ID into account. Therefore, the aims of this study were to investigate (a) the distribution of handedness, (b) the grip strength ratios between the dominant and non-dominant hand, (c) the distribution of the strongest hand with regard to handedness, and (d) the relation between handedness and the severity of ID.

METHODS

Study design and participants

This study was part of the large Dutch cross-sectional study 'Healthy ageing and intellectual disabilities' (HA-ID), executed by a consort consisting of three ID care provider services in the mid, west and south of the Netherlands in collaboration with two university departments (Intellectual Disability Medicine, Erasmus Medical Center at Rotterdam; Center for Human Movement Sciences, University Medical Center at Groningen). All 2150 clients with ID, aged 50 years and over, of the three care provider services were invited to participate, resulting in a near-representative sample of 1050 clients. Details about design, recruitment, and representativeness of the sample have been presented elsewhere [27]. Data collection took place between February 2009 and July 2010.

Ethical approval was provided by the Medical Ethical Committee of the Erasmus Medical Center (MEC 2008-234) and by the ethical committees of the participating ID care provider services. Informed consent was obtained from all participants; however, unusual resistance was a reason for aborting measurements at all times. This study followed the guidelines of the Declaration of Helsinki [28].

Procedure

Data were collected as part of an extensive physical fitness assessment, which was conducted on locations familiar or close to participants: a large room within their home, a familiar daycare center, or a gym. Assessments were guided by test instructors, who all were physiotherapists, occupational therapists, or physical activity instructors with experience with individuals with ID. They all received an instruction manual and followed two days of training for the execution of all assessments.

Standardized encouragement provided by test instructions for testing individuals with normal intellectual capabilities is unsuitable for individuals with ID. To keep this motivational aspect as equal as possible, we prescribed 'maximal motivation' to the test instructors for all tests. In some cases, this meant that participants were motivated to engage in the assessments by constant verbal encouragement and verbal rewarding, in other cases the test instructor had to remain very calm and quiet to motivate the participant as much as possible and to prevent stress or anxiety. The specific background, knowledge, and experience of the test instructors were important conditions to ensure the most suitable 'maximal motivation' for every participant, while regarding safety as well.

Measurements

General information

To describe the study population in detail, the following information was collected.

Gender, age, and residential status (central setting providing intensive care and support, community-based setting, independent living with ambulatory support, or with relatives) were collected from the administration systems of the ID care provider services.

The presence of Down syndrome and spasticity of the arms was collected through the medical files. Professional caregivers provided information about mobility impairments (independent, walking with an aid, or wheelchair-bound). Daily functioning was assessed with the Barthel Index [29] for basic activities of daily living (BADL), scores ranging from 0 (completely dependent) to 20 (completely independent) and the Lawton IADL Scale [30] for Instrumental activities of daily living (IADL), scores ranging from 8 (completely dependent) to 24 (completely independent). The questionnaires were completed by professional caregivers of the participants.

Level of ID was categorized by psychologists or behavioral therapists as: borderline (IQ = 70 – 84), mild (IQ = 50 – 69), moderate (IQ = 35 – 49), severe (IQ = 20 – 34) or, profound (IQ < 20) based on International Classification of Diseases (ICD-10) criteria [31].

Handedness

Handedness is defined as the relative preference for one hand in the execution of unimanual tasks [32, 33]. It was determined by asking the participant, who was sitting down, to pick up a block (2.5 cm^3) which was put on the table in front of him or her. This was part of the Box and Block test, assessed in the extensive physical fitness assessment of the HA-ID study [27].

Grip strength

Grip strength [34] was measured with the Jamar Hand Dynamometer (#5030J1, Sammons Preston Rolyan, USA). Reliability and validity in the general population was good [35, 36]. Test-retest reliability in older adults with ID was good (ICC 0.94 [same day interval] and 0.90 [2-week interval]) [11]. In a previous report from the HA-ID study, selective loss to participation was reported. Older adults with severe or profound ID, Down syndrome or wheelchair users were underrepresented [37].

The handle of the dynamometer was put in the second smallest position according to the instruments' instructions. The middle phalanges had to rest on the handle, if not, the position was adjusted. An example of the test was provided by squeezing a rubber ball by the test instructor. Subsequently, the participant was allowed to squeeze the ball, to assure understanding of the task. The participant squeezed the dynamometer to his or her maximum ability in seated position, according to the recommendations of the ASHT [38]. The best result of three attempts for both the left and right hand (with a one-minute pause between attempts) was recorded, in kilogram (kg). The test instructor had to be convinced that the participant squeezed with maximal effort; otherwise the result was not recorded. In order to check this, test instructors looked at facial expressions, contracting muscles of the arm and hand, turning white of the phalanges, and the consistency of the three attempts.

Statistical analyses

Characteristics of the study sample, completion rates of the tests, and distribution of handedness were described first.

Consequently, differences in strength between the dominant and non-dominant hand were analyzed with paired *t* tests for the total study sample and subgroups according to level of ID. Because eight *t* tests were performed, Bonferroni correction was used to correct for the inflated familywise error rate. *P*-values smaller than 0.00625 (0.05/8) were considered statistically significant.

Grip strength ratios were expressed as a percentage difference between the dominant and non-dominant hand. Individual grip strength ratios were calculated following the formula ((*strength dominant hand / strength non-dominant hand*) - 1) x 100%). Subsequently, the mean grip strength ratios were calculated for the total study sample and the subgroups according to level of ID.

To tell which of both hands was the strongest hand, a categorical variable was made from the grip strength scores, with the categories right, left, and equal strength for both hands. A grip strength difference of 1 kg was used to divide the scores in these categories. Pearson's chi-square test was used to analyze the relationship between handedness and the strongest hand. *P*-values smaller than 5% ($p < 0.05$) were considered statistically significant.

Furthermore, mean grip strength ratios were then calculated for the group of participants with the dominant hand as the strongest hand and for the group of participants with the non-dominant hand as the strongest hand, for right- and left-handed participants separately. Again this was done for the total study group and the subgroups according to level of ID.

All analyses were performed with the Statistical Package for Social Sciences (SPSS) version 17.0 (IBM Corporation, New York).

RESULTS

Participants

Of the 1050 clients that participated in the HA-ID study, 652 (62.1%) had successful measurements of handedness and grip strength for both hands. Reasons for drop-out were mainly attributable to limited understanding (13.6%), physical disability (3.1%), and non-cooperation (2.9%).

The groups of participants with borderline (n = 30) and profound (n = 3) ID were relatively small. Because the borderline and mild ID group did not differ significantly on grip strength scores and gender, and neither did the severe and profound ID group (data not presented), these groups were combined in the analysis.

Characteristics of the study sample are shown in Table 1.

Handedness and grip strength

The study sample consisted of a large number of left-handed participants (n = 171, 26.2%) (Table 2). A trend towards an increase in left-handedness with the severity of the ID was found.

Table 2 shows the mean grip strength scores and strength ratios for right-handed and left-handed participants, respectively. For right-handed participants the right hand was significantly stronger than the left hand, on average 8.7%. In the subgroup analysis for level of ID this significant difference was only present in the moderate ID group. The strength ratio was highest in the severe to profound ID group. For left-handed participants there was no significant difference in strength between the dominant and non-dominant hand, but the strength ratios were large.

Distribution of the strongest hand

However, when analyzing which hand (dominant or non-dominant) was actually the strongest hand, the dominant hand proved to be the strongest only in 54.7% of the right-handed participants and in 46.2% of the left-handed participants (p = 0.001) (Table

Table 1. Participant characteristics of the study sample.

Characteristics		***n* (%)**
Total		652
Age	Years ($m \pm sd$)	61.7 ± 8.0
Gender	Female	324 (49.7)
	Male	328 (50.3)
Type of setting	Central setting	254 (39.0)
	Community-based	345 (52.9)
	Ambulatory support	41 (6.3)
	With relatives	5 (0.8)
Level of ID	Borderline	30 (4.6)
	Mild	184 (28.2)
	Moderate	353 (54.1)
	Severe	63 (9.7)
	Profound	3 (0.5)
	Unknown	19 (2.9)
Down syndrome	Yes	69 (10.6)
	No	440 (67.5)
	Unknown	143 (21.9)
Spasticity	Right hand	25 (3.8)
	Left hand	23 (3.5)
	Unknown	126 (19.3)
Mobility	Independent	493 (75.6)
	Walking aid	103 (15.8)
	Wheelchair	26 (4.0)
	Unknown	30 (4.6)
ADL	BADL ($m \pm sd$)	16.0 ± 4.1
	IADL ($m \pm sd$)	13.3 ± 4.9

m = mean; *sd* = standard deviation; *n* = number of participants; ID = intellectual disability; ADL = activities of daily living; BADL = basic activities of daily living (0 – 20); IADL = instrumental activities of daily living (8 – 24).

3). For 13% of the participants, both hands were equally strong. This means that in 34.5% of the participants ((65 + 160) / 652) the non-dominant hand was the strongest hand.

The strength ratios for the selection of participants with the dominant hand and participants with de non-dominant hand as the strongest hand, for right- and left-handed participants are presented in Table 4.

For right-handed participants with their right hand as the strongest hand, the strength ratio was on average 26%. The highest strength ratio was found in the severe to profound ID group. For the right-handed participants with their left hand as the strongest hand,

Table 2. Handedness, mean grip strength, and strength ratios for the total study sample and ID level subgroups.

	Total	Borderline-mild ID	Moderate ID	Severe-profound ID
Right-handed *n* (%)	**481 (73.8)**	**167 (78.0)**	**255 (72.2)**	**44 (66.7)**
Mean strength right hand (kg) ($m \pm sd$)	24.2 ± 10.1	28.1 ± 10.9	22.7 ± 8.9	19.0 ± 9.1
Mean strength left hand (kg) ($m \pm sd$)	23.2 ± 10.0	27.3 ± 10.6	21.6 ± 8.8	17.5 ± 8.2
t-value	4.53**	1.95	3.67**	2.47
Strength ratio (% difference)	8.7	4.8	10.7	11.9
Left-handed *n* (%)	**171 (26.2)**	**47 (22.0)**	**98 (27.8)**	**22 (33.3)**
Mean strength left hand (kg) ($m \pm sd$)	22.2 ± 9.5	25.5 ± 10.0	21.3 ± 8.7	18.4 ± 8.5
Mean strength right hand (kg) ($m \pm sd$)	21.6 ± 10.6	24.8 ± 11.5	20.1 ± 9.7	20.5 ± 10.8
t-value	1.55	1.06	2.06	- 1.79
Strength ratio (% difference)	27.2	25.6	35.4	- 5.9

m = mean; *sd* = standard deviation; *n* = number of participants; ID = intellectual disability.
** $p < 0.001$

Table 3. Distribution of handedness and the strongest hand for the total study sample ($n = 652$).

		Strongest hand			
	***n* (%)**	**Right**	**Left**	**Equal**	**Total**
Handedness	**Right**	263 (54.7)	160 (33.3)	58 (12.1)	481 (100)
	Left	65 (38.0)	79 (46.2)	27 (15.8)	171 (100)
	Total	328 (50.3)	239 (36.7)	85 (13.0)	652 (100)

n = number of participants.

Table 4. Strength ratios of subgroups of right- and left-handed participants.

	Total	Borderline-mild ID	Moderate ID	Severe-profound ID
Right-handed				
Strength ratio – selection right hand strongest	26.0	18.3	29.8	32.1
Strength ratio – selection left hand strongest	- 16.6	- 16.8	- 16.1	- 17.5
Left-handed				
Strength ratio – selection left hand strongest	72.1	72.3	80.3	16.7
Strength ratio – selection right hand strongest	- 16.3	- 11.7	- 17.2	- 22.5

ID = intellectual disability.

the strength ratio was on average – 16.6 %. Again the highest strength ratio was found in the severe to profound ID group.

For left-handed participants with their left hand as the strongest hand, the strength ratio was on average 72.1%. The highest strength ratio was found in the moderate ID group. For the left-handed participants with their right hand as the strongest hand, the strength ratio was on average – 16.3 %, the highest strength ratio was found in the severe to profound ID group.

DISCUSSION

This first study into the effect of handedness on grip strength in older adults with ID shows that 26.2% was left-handed. When looking at handedness alone, the dominant hand (right hand) of right-handed participants was on average 8.7% stronger ($p < 0.001$). For the left-handed participants there was no significant difference between both hands. However, more detailed analyses revealed a large percentage of participants (34.5%) in which the non-dominant hand was the strongest hand, the non-dominant hand proved to be on average 16.6% stronger for right-handed and 16.3% stronger for left-handed participants. This influenced the strength ratios calculated for the dominant versus non-dominant hand. Therefore, we recommended that both hands are tested in grip strength measurements in this population.

The high percentage of left-handedness found in this study is in agreement with previous research showing high percentages of left-handedness in the population with ID, ranging from 17.3% up to 32.0% as compared to 9.3 % to 16.7% of the nondisabled controls [18, 20, 22-24, 39, 40]. The cause of this increased prevalence of left-handedness is not well understood. Carlier et al. (2011) suggested that the atypical laterality could be related to the general cognitive level; they found a trend for increased left- and mixed-handedness with lower IQ scores [24], as did Lucas et al. (1989). In this study, left-handedness also increased from 22.0% in the group with borderline or mild ID up to 33.3% in the group with severe or profound ID. Another explanation may be that atypical laterality is associated with language, which is often compromised in individuals with ID [23, 40].

Research in the general population has shown that the dominant hand was more often the strongest hand [14, 15], with grip strength ratios ranging from 0% to 11% [12, 15, 41-43]. The results for right-handed participants in this study are in line with these findings; the right hand was significantly stronger than the left hand with a strength ratio of 8.7%. However, for left-handed participants this was not the case. This result is in line with the finding of Bohannon (2003) who reported that right-handed individuals are stronger with their right hand, but for left-handed participants results were equivocal. These results would suggest that it is sufficient to measure only one hand, the dominant hand in right-handed participants, and either hand for left-handed participants.

However, in this study 34.5% of the participants had higher grip strength in their non-dominant hand. In comparison, in adults without ID only 14.1% had higher or equal grip strength with their non-dominant hand [15]. The strength ratios of this selection show that if grip strength is measured for only the dominant hand, an error up to 22.5% can be made. These results stress the need to always measure both hands when assessing the maximal grip strength of an individual with ID. The wide distribution of grip strength ratios (both positively and negatively) for these subgroups, is likely to cause the low grip strength ratios of the dominant versus the non-dominant hand of the first analyses.

Norm-referenced values for grip strength are available for the general population [43-45]. These normative values are either presented for left and right hands [44, 45] or as one value for both hands [43], thus causing difficulties for use in this population. No norm-referenced values for the population of ID are currently available. Until these normative values are developed, the existing values of the general population can be used by comparing the highest grip strength value, regardless from which hand, to the values of Peters et al. (2011) or right-hand values (the strongest hand in these values) of Bohannon et al. (2006) and Bohannon et al. (2007).

A limitation of this study is that handedness was measured by performing just one unimanual task and was categorized in right- or left-handedness with that information, not in mixed-handedness. A more precise way to assess handedness would have been by assessing the preferred hand used in several tasks, for example based on the Annett hand preference questionnaire (AHPQ) [32]. This would provide information about the consistency of the use of one preferred hand, by which a distinction could be made not only between right- and left-handed but also mixed-handed individuals. A thorough assessment of handedness is therefore recommended for future research. The fact that mixed-handedness was not identified could contribute to the large proportion which had the non-dominant hand as the strongest hand.

Older adults with borderline or mild ID without any form of registered professional support or care are not included in the HA-ID population and results are therefore not generalizable to this group. Participants with severe and profound ID were under-represented through selective drop out during grip strength assessment. A higher percentage of left-handedness was found in this group. We conclude that the proportion of left-handedness presented here is likely to be an underestimation rather than an overestimation.

In summary, this study provides guidelines for grip strength assessment in older adults with ID and recommendations how to use existing normative values. Further research is necessary to provide normative values for the older population of ID.

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Chapter 5

Feasibility and outcomes of the Berg Balance Scale in older adults with intellectual disabilities

Alyt Oppewal, Thessa I.M. Hilgenkamp, Ruud van Wijck, Heleen M. Evenhuis

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ABSTRACT

High incidence of falls and increased risk of fall-related injuries are seen in individuals with intellectual disabilities (ID). The Berg Balance Scale (BBS) is a reliable instrument for balance assessment in the population of (older) adults with ID. The aims of this study were to assess the balance capacities of a large group of older adults with ID with the BBS and look for gender and age effects, as well as reasons for drop-out on separate items, and to identify feasible subtests for subgroups in which the complete BBS is not feasible. The balance capacities of 1050 older clients with borderline to profound ID of three Dutch care-provider services (mean age 61.6 [*sd* = 8.0]) were assessed with the BBS. The participants who completed all items of the BBS (*n* = 508) were the functionally more able part of the study sample. Results showed that even this functionally more able part had poor balance capacities, with a mean BBS score of 47.2, 95% CI [46.3, 48.0], similar to adults in the general population aged around 20 years older. Balance capacities decreased with increasing age and females had poorer balance capacities than males. Difficulties understanding the task and physical limitations were most often the reasons for drop-out. Feasible subtests were identified for the subgroups with very low cognitive levels and wheelchair users. Low balance capacities of older adults with ID show the need for regular screening and the urge for fall prevention programs for individuals with ID.

INTRODUCTION

A high incidence of falls and increased risk of fall-related injuries is seen in individuals with intellectual disabilities (ID) [1-4]. The broad age range of participants in these studies indicates that falling is not restricted to older individuals with ID. However, the risk of falling increases with advancing age [1, 5-7], with notable increases in falls found for individuals with ID in their 40's and 50's [1]. Chiba et al. (2009) reported a 2.5 times (odds ratio = 2.46) higher fall risk in those over 50 years of age compared to those younger than 50 years of age [8], and Hsieh et al. (2001) reported a 10-fold risk (odds ratio = 10.63) in those over 70 years of age for falls and related injuries in comparison to those younger than 70 years of age [5]. Furthermore, falling seems to lead more often to injury and hospitalization in individuals with ID than in the general population [4, 9].

Balance assessment instruments are used in the general population to identify fall risk and target and evaluate fall prevention programs. However, not all of these instruments are applicable to individuals with ID, because of their limited cognitive ability and comorbidities [2, 10, 11]. Based on a review by Hilgenkamp et al. (2010), the Berg Balance Scale (BBS) was proposed as the most applicable instrument to assess balance capacities and fall risk in older adults with ID [10]. The BBS is a 14 item performance-based instrument that measures balance capacities [12]. A higher score corresponds to better balance capacities. A score of 45 (of the maximum score of 56) has been proposed as a cut-off to differentiate between those at risk for falls (< 45) and those not at risk for falls (≥ 45) [12]. Residents of a home for the elderly without ID with a score below 45 had a 2.7 times (relative risk = 2.7, 95% CI [1.5, 4.9]) greater risk to fall over the next 12 months, than those with a score above 45 [12]. However, the BBS is better at identifying non-fallers than fallers [13]. The BBS was found valid for balance assessment in residents of a home for the elderly without ID, with significant correlations with other balance scales such as the Timed Up & Go (r = -0.76) and the Tinetti Balance subscale (r = 0.91) and reliable, with high inter-rater reliability (ICC = 0.98), intra-rater reliability (ICC = 0.97), and internal consistency (ICC = 0.83) [14-16].

In the population with ID, the BBS was found to be a reliable instrument [17, 18] and feasible for older adults with mild to moderate ID who are able to walk for at least 10m and understand simple instructions [19]. Validity has not yet been investigated in this group.

In the 'Healthy ageing and intellectual disabilities' (HA-ID) study, the health of 1050 older adults (50+) with borderline to profound ID was investigated [20]. The BBS was used to assess the balance capacities and was found feasible for this group, except for the subgroups with severe to profound ID and older adults who use a wheelchair inside their homes [21]. Completions rates of these subgroups were lower than 25% [21]. In order to interpret BBS results correctly for the subgroups with a large drop-out, more detailed analysis of the reasons for drop-out of these subgroups is necessary. Furthermore, analysis on item level is important to identify subtests that are feasible in these individuals.

Therefore, the aims of this study were (a) to assess the balance capacities of older adults with ID with the BBS, and look for gender and age effects (b) to assess the reasons for drop-out on item level for subgroups with low completion rates (< 25%), and (c) to identify feasible subtests of the BBS for these subgroups.

METHODS

Study design and participants

This study was part of the large Dutch cross-sectional HA-ID study executed by a consort consisting of three ID care-provider services in collaboration with two university departments (Intellectual Disability Medicine, Erasmus Medical Center at Rotterdam and the Center for Human Movement Sciences, University Medical Center at Groningen). All 2150 clients with ID, aged 50 years and over, of the three care-provider services were invited to participate, resulting in a near-representative sample of 1050 clients. Details about design, recruitment, and representativeness of the sample have been presented elsewhere [20]. Data collection took place between February 2009 and July 2010.

Ethical approval was provided by the Medical Ethical Committee at Erasmus Medical Center (MEC 2008-234) and by the ethical committees of the participating ID care-provider services. Informed consent was obtained from all participants; however, unusual resistance was a reason for aborting measurements at all times. This study followed the guidelines of the Declaration of Helsinki [22].

Procedure

Data were collected as part of an extensive physical fitness assessment, which was conducted at locations familiar or close to participants: a large room within their home, a familiar daycare center, or a gym. Assessments were guided by test instructors, who all were physiotherapists, occupational therapists, or physical activity instructors with experience with individuals with ID. They all received an instruction manual and followed two days of training for the execution of all assessments.

Standardized encouragement provided by test instructions for testing individuals with normal intellectual capabilities is unsuitable for individuals with ID. To keep this motivational aspect as equal as possible, we prescribed 'maximal motivation' to the test instructors for all tests. In some cases, this meant that participants were motivated to engage in the assessments by constant verbal encouragement and verbal rewarding, in other cases the test instructor had to remain very calm and quiet to motivate the participant as much as possible and to prevent stress or anxiety. The specific background, knowledge, and experience of the test instructors were important conditions for ensuring the most suitable 'maximal motivation' for every participant, while regarding safety as well.

Measurements

General information

Gender, age, and residential status (central setting providing intensive care and support, community-based setting providing support of independence, independent living with outreaching support, or with relatives) were collected from the administrative systems of the ID care-provider services. Depending on the residential status, the care and support participants received ranged from complete care and support in basic activities of daily living to only minimal support instrumental activities of daily living, for example managing finances. Additional treatment is only deployed when indicated by a medical diagnosis, just as in the general population. The presence of Down syndrome (DS), spasticity of the legs and arms (unilateral, bilateral), scoliosis, Parkinson's disease, cerebrovascular accident, and visual and hearing problems were collected through the medical files. Professional caregivers provided information about mobility (independent, walking with an aid, or wheelchair-bound). Level of ID was categorized by psychologists or behavioral therapists as borderline (IQ = 70 – 84), mild (IQ = 50 – 69), moderate (IQ = 35 – 49), severe (IQ = 20 – 34), or profound (IQ < 20) based on International Classification of Diseases (ICD-10) criteria [23].

Height and weight was measured by trained medical assistants during physical examination; detailed methods have been described elsewhere [24]. The body mass index (BMI) was calculated by weight divided by squared height [25]. Physical activity was measured with the NL-1000 pedometer (New Lifestyles, Missouri, USA) as part of the extensive physical fitness assessment [26, 27].

Balance

The BBS consists of 14 static and dynamic functional balance tasks varying in difficulty, ranging from unsupported sitting in a chair to tandem stance and standing on one leg (Table 1) [12, 28]. The original test instructions were followed with some aids to enhance understanding of the tasks: two carpet feet and a carpet circle on the floor, to point out where the participant had to stand or turn around on. Walking aids were not allowed. The items were scored on a 5-point scale from 0 (inability to complete the task) to 4 (completion of the task) points. A modified version of the forward reach (item 8) was added, called item 8a, to make the reaching task easier to understand. Participants had to reach forward with their arms stretched holding a ring, and place the ring around a

Table 1. Items of the Berg Balance Scale.

Item	Description
1	Sitting to standing
2	Standing unsupported
3	Standing to sitting
4	Sitting unsupported
5	Transfers
6	Standing unsupported with eyes closed
7	Standing unsupported with feet together
8	Reaching forward with arms stretched while standing
8a	Reaching forward to place a ring around a standing stick
9	Pick up object from floor
10	Look behind while standing with feet fixed
11	Turning 360 degrees
12	Alternating placing foot on step
13	Tandem stance
14	Standing on one leg

standing stick. The stick was placed further away until the participant could no longer place the ring around the stick. The result was calculated by subtracting the arm length (distance of the acromion to the inside of the ring) from the distance between the ankles (malleolus exterior) and the stick.

Reasons for drop-out on each item were recorded by test instructors using the categories: difficulties understanding the task, physical limitations (involving the lower limbs, e.g., spasticity, mobility problems, foot deformations), concentration problems, does not feel like participating, anxiety, and reasons unknown to the investigators.

Statistical analyses

Characteristics of the study sample were described. Based on participation on the original 14 items of the BBS, differences between participants and non-participants of the BBS were analyzed with an independent *t* test and Pearson's chi-square tests. For the group who did not complete the original 14 items of the BBS, the number of items performed was described. The reasons for drop-out were presented as the number of times a specific reason for drop-out was reported for any one of the 14 items of the BBS, divided by the total number of times any reason for drop-out was reported for any one of the 14 items of the BBS.

The scores of the BBS were calculated for the participants who completed all the original 14 items of the BBS. Results are presented as mean scores with 95% confidence intervals for the overall group and for females and males in age categories of 5 years.

The percentage that had a score of 45 or higher was calculated. The differences between the age categories for females and males were analyzed with Kruskal-Wallis tests. When significant, post hoc tests were performed using a Bonferroni correction. The difference in BBS results between men and women was analyzed with an independent *t* test.

Feasibility of the BBS items for the subgroups (who participated on at least one item of the BBS) with severe to profound ID and wheelchair users were assessed. These subgroups had low completion rates on the original BBS (< 25%) [21]. Reasons for drop-out were described. Subtests for these subgroups were composed with the items which were considered feasible. Subtests were considered feasible with completion rates of 25% or higher according to the previously used cut-off score by Hilgenkamp et al. (2013) [21]. We considered individual items feasible if at least 50% of the participants could perform the item. Scores were calculated for these subtests and presented as mean scores with 95% confidence intervals. Feasibility of item 8 compared to item 8a was analyzed with Pearson's chi-square tests for the subgroups with severe to profound ID and wheelchair users.

Both confidence intervals and p-values are reported [29] and *p*-values smaller than 5% ($p < 0.05$) were considered statistically significant. Analyses were performed with the Statistical Package for Social Sciences (SPSS) version 20.0 (IBM Corporation, New York).

RESULTS

Participants

Of the total study population of the HA-ID study ($n = 1050$), 811 participants completed at least one item of the BBS and 508 participants performed all 14 items. The characteristics of these three groups are shown in Table 2. Compared to the total HA-ID study population, participants who completed the BBS ($n = 508$) were taller ($t(868.15) = -6.96$, $p < 0.001$), heavier ($t(897) = -7.60$, $p < 0.001$), had higher BMI ($t(891) = -3.16$, $p = 0.002$), lived more often in the community (χ^2 [4, $n = 1050$] = 206.99, $p < 0.001$), had more often borderline to moderate ID (χ^2 [5, $n = 1050$] = 229.50, $p < 0.001$), less Down syndrome diagnoses (χ^2 [2, $n = 1050$] = 18.28, $p < 0.001$), less spasticity of the legs (χ^2 [3, $n = 1050$] = 14.88, $p = 0.002$) and arms (χ^2 [3, $n = 1050$] = 14.02 $p = 0.003$), less scoliosis (χ^2 [2, $n = 1050$] = 15.19, $p = 0.001$), less visual (χ^2 [3, $n = 1050$] = 39.57, $p < 0.001$) and hearing impairments (χ^2 [4, $n = 1050$] = 42.48, $p < 0.001$), and less mobility impairments (χ^2 [2, $n = 1050$] = 95.16, $p < 0.001$).

Drop-outs

Of the total HA-ID study population ($n = 1050$), 150 participants were absent on the day of the physical fitness assessment. Reasons for absence were illness, lack of coopera-

Table 2. Characteristics of the HA-ID study sample and participants on the Berg Balance Scale.

Characteristics	***n* (%)**	**HA-ID**	**BBS (≥ 1 item)**	**BBS (14 items)**
Total		1050	811	508
Age	Years ($m \pm sd$)	61.6 ± 8.0	61.4 ± 7.8	61.3 ± 7.6
Gender	Female	511 (48.7)	398 (49.1)	248 (48.8)
	Male	539 (51.3)	413 (50.9)	260 (51.2)
Height	cm ($m \pm sd$)	161.6 ± 11.6	162.1 ± 11.5	164.0 ± 10.7
Weight	kg ($m \pm sd$)	71.1 ± 15.7	71.7 ± 15.6	74.6 ± 15.4
BMI	kg/m^2 ($m \pm sd$)	27.2 ± 5.2	27.2 ± 5.1	27.7 ± 5.0
Type of setting	Central setting	557 (53.0)	392 (48.3)	156 (30.7)
	Community-based	432 (41.1)	363 (44.8)	301 (59.3)
	Ambulatory support	43 (4.1)	41 (5.1)	40 (7.3)
	With relatives	7 (0.7)	7 (0.9)	5 (1.0)
	Unknown	11 (1.0)	8 (1.0)	6 (1.2)
Level of ID	Borderline	31(3.0)	30 (3.7)	28 (5.5)
	Mild	223 (21.2)	193 (23.8)	164 (32.3)
	Moderate	506 (48.2)	403 (49.7)	267 (52.6)
	Severe	172 (16.4)	121 (14.9)	31 (6.1)
	Profound	91 (8.7)	43 (5.3)	1 (0.2)
	Unknown	27 (2.6)	21 (2.6)	17 (3.3)
Down syndrome	Yes	149 (14.2)	113 (13.9)	44 (8.7)
	No	724 (69.0)	552 (68.1)	344 (67.7)
	Unknown	177 (16.9)	146 (18.0)	120 (23.6)
Mobility	Independent	731 (69.6)	630 (77.7)	416 (81.9)
	Walking aid	151 (14.4)	119 (14.7)	55 (10.8)
	Wheelchair	107 (10.2)	30 (3.7)	10 (2.0)
	Unknown	61 (5.8)	32 (3.9)	27 (5.3)
Spasticity legs	Unilateral	26 (2.5)	20 (2.5)	12 (2.4)
	Bilateral	69 (6.6)	31 (3.8)	16 (3.1)
	No	799 (76.1)	327 (77.3)	372 (73.2)
	Unknown	156 (14.9)	133 (16.4)	108 (21.3)
Spasticity arms	Unilateral	23 (2.2)	18 (2.2)	10 (2.0)
	Bilateral	53 (5.0)	27 (3.3)	11 (2.2)
	No	818 (77.9)	633 (78.1)	379 (74.6)
	Unknown	156 (14.9)	133 (16.4)	108 (21.3)
Scoliosis	Yes	95 (9.0)	60 (7.4)	25 (4.9)
	No	800 (76.2)	621 (76.6)	376 (74.0)
	Unknown	155 (14.8)	130 (16.0)	107 (21.1)
Parkinson	Yes	8 (0.8)	677 (83.5)	4 (0.8)
	No	891 (84.9)	6 (0.7)	399 (78.5)
	Unknown	151 (14.4)	128 (15.8)	105 (20.7)

Characteristics	*n* (%)	HA-ID	BBS (≥ 1 item)	BBS (14 items)
CVA	Yes	52 (5.0)	646 (79.7)	22 (4.3)
	No	850 (81.0)	38 (4.7)	380 (74.8)
	Unknown	148 (14.1)	127 (15.7)	106 (20.9)
Visual problems	Yes	219 (20.9)	146 (18.0)	61 (12.0)
	No	666 (63.4)	528 (65.1)	335 (65.9)
	Unknown	165 (15.7)	137 (16.9)	112 (22.0)
Hearing problems	Yes	409 (38.9)	307 (37.9)	140 (27.6)
	No	470 (44.8)	366 (45.1)	257 (50.6)
	Unknown	171 (16.3)	138 (17.0)	111 (21.8)
Physical activity	> 7500 steps/day	93 (8.9)	89 (11.0)	80 (15.7)
	< 7500 steps/day	164 (15.6)	161 (19.9)	141 (27.8)
	Unknown	793 (75.5)	561(69.2)	287 (56.5)

m = mean; *sd* = standard deviation; *n* = number of participants; ID = intellectual disability; CVA = cerebrovascular accident.

tion, logistic reasons, or no consent for the physical fitness assessment [26]. Of the participants present, 392 (900 – 508) did not complete all 14 original items of the BBS. The distribution of the number of items of the original BBS performed by this group is shown in Figure 1. The reasons for drop-out on the separate items of the BBS were difficulties understanding the task (38.8%), physical limitations (32.6%), concentration problems (3.0%), did not feel like participating (7.8%), anxiety (3.8%), and reasons unknown to the investigators (11.9%).

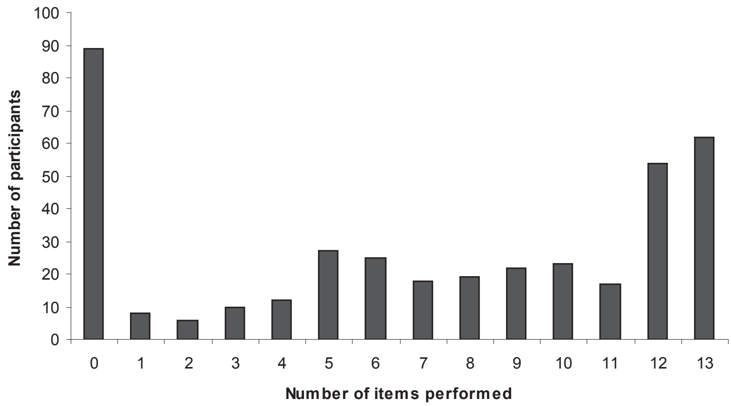

Figure 1. Distribution of the number of items performed by the participants who did not complete the original 14 items of the Berg Balance Scale (*n* = 392).

Results of the BBS

The mean total score of the BBS ($n = 508$) was 47.2, 95% CI [46.3, 48.0]. Of the 508 participants, 78.9% had a score of or above the cut-off value of 45. The results for the 5-year age categories are presented in Table 3. Males had a significantly better mean BBS score than females ($t(506) = 2.76$, $p = 0.006$). The BBS scores deteriorated significantly with increasing age for both females and males, $H(5) = 16.22$, $p = 0.006$ and $H(5) = 28.38$, $p < 0.001$, respectively. However, post hoc tests did not show significant differences between the consecutive age categories.

Table 3. Results of the Berg Balance Scale for age categories of 5 years ($n = 508$).

Age-category (years)	*n*	BBS score mean [95% CI]	Score ≥ 45 (%)
Female	**248**	**46.0 [44.6, 47.3]**	**72.2**
50-54	65	47.7 [44.8, 50.5]	76.9
55-59	66	47.3 [45.1, 49.5]	78.8
60-64	41	45.9 [42.7, 49.0]	73.2
65-69	34	42.6 [37.5, 47.6]	64.7
70-74	25	43.6 [38.7, 48.5]	68.0
75+	17	44.5 [38.7, 50.2]	76.5
Male	**260**	**48.3 [47.4, 49.3]**	**83.5**
50-54	47	49.4 [46.7, 52.2]	93.6
55-59	59	50.6 [49.2, 51.9]	93.2
60-64	70	49.5 [48.3, 50.6]	84.3
65-69	46	46.4 [43.6, 49.1]	73.9
70-74	22	44.6 [39.6, 49.6]	63.6
75+	16	42.8 [36.7, 48.9]	68.8

n = number of participants; BBS = Berg Balance Scale; CI = confidence interval.

Feasibility for subgroups

The feasibility of the BBS items and reasons for drop-out of the subgroups with low completion rates (< 25%) on the original BBS [21] are presented in Table 4. The reasons 'concentration problems', 'does not feel like participating', and 'anxiety' were taken together in the table because they overall represented small proportions (< 5%) of the reasons for drop-out.

Of individuals with severe ID, less than 50% participated on item 8. The main reason for drop-out on this item was difficulties with understanding the task (53.3%) (Table 4).

Of individuals with profound ID, less than 50% participated on the items 6 to 14. The main reasons for drop-out on these items were difficulties understanding the task (25.6% to 76.7%) and physical limitations (9.3% to 14.0%) (Table 4).

Table 4. Feasibility of the BBS items and reasons for drop-out for individuals with severe ($n = 121$) and profound ($n = 43$) ID and wheelchair users ($n = 30$), presented in percentages.

	Severe ID					Profound ID					Wheelchair				
Item	Part	Reason DiffUnd	Reason PhysL	Reason CFA	Unknown	Part	Reason DiffUnd	Reason PhysL	Reason CFA	Unknown	Part	Reason DiffUnd	Reason PhysL	Reason CFA	Unknown
1	97.5	0.8	1.7	-	-	88.4	7.0	2.3	-	2.3	86.7	3.3	10.0	-	-
2	86.0	7.4	1.7	4.2	0.8	53.5	27.9	4.7	11.6	2.3	76.7	6.7	16.7	-	-
3	98.3	-	1.7	-	-	93.0	4.7	-	-	2.3	90.0	-	10.0	-	-
4	94.2	5.0	-	0.8	-	81.4	14.0	-	2.3	2.3	96.7	3.3	-	-	-
5	93.4	5.0	0.8	0.8	-	74.4	18.6	2.3	2.3	2.3	86.7	-	10.0	3.3	-
6	57.9	36.4	2.5	3.3	-	16.3	65.1	9.3	4.6	4.7	43.3	23.3	30.0	3.3	-
7	58.7	28.9	3.3	5.8	3.3	25.6	51.2	11.6	4.7	7.0	50.0	13.3	36.7	-	-
8	37.9	53.3	3.3	3.3	-	7.0	76.7	11.6	2.3	2.3	36.7	16.7	40.0	6.6	-
8a	56.2	33.9	3.3	5.8	0.8	11.6	67.4	14.0	2.3	4.7	40.0	16.7	40.0	3.3	-
9	86.0	8.3	3.3	2.5	-	48.8	25.6	11.6	9.4	4.7	46.7	6.7	43.3	3.3	-
10	51.2	40.5	4.1	4.1	-	9.3	65.1	14.0	7.0	4.7	40.0	16.7	40.0	3.3	-
11	71.1	19.8	4.1	5.0	-	23.3	51.2	14.0	7.0	4.7	43.3	13.3	43.3	-	-
12	54.5	36.4	4.1	4.9	-	7.0	67.4	11.6	9.3	4.7	43.3	16.7	40.0	-	-
13	50.4	36.4	5.0	8.3	-	4.7	67.4	9.3	14.0	4.7	50.0	16.7	33.3	-	-
14	64.5	25.6	4.1	5.8	-	14.0	60.5	11.6	9.3	4.7	50.0	13.3	36.7	-	-

BBS = Berg Balance Scale; Part = Participation; Reason DiffUnd = Difficulties understanding the task; Reason PhysL = Physical limitations; Reason CFA = Concentration/ does not Feel like participating/ Anxiety; Unknown.

In the subgroup of individuals who used a wheelchair inside their home, less than 50% participated on the items 6 and 8 to 12. The main reasons for drop-out were physical limitations (30.0% to 43.3%) and difficulties understanding the task (6.7% to 23.3%) (Table 4).

Item 8 was problematic for all three subgroups. The modified version (item 8a) had a significantly higher participation rate in the subgroup of individuals with severe ID (χ^2 [1, n = 121] = 45.58, $p < 0.001$) and the subgroup of wheelchair users (χ^2 [1, n = 30] = 26.05, $p < 0.001$), mostly explained by less drop-out due to difficulties understanding the task.

Results of the BBS subtests

For individuals with severe ID, a BBS subtest can be composed by omitting item 8. Of the 121 individuals with severe ID, 33 participants (27.3%) completed this subtest. The mean score was 39.7, 95% CI [36.2, 43.2] of the maximum score of 52.

The BBS subtest for individuals with profound ID consists of the items 1 to 5. Of the 43 individuals with profound ID 16 (37.2%) completed this subtest. The mean score was 14.4, 95% CI [10.9, 18.0] of the maximum score of 20.

The subtest of the BBS for individuals who are in a wheelchair inside their home consists of items 1 to 5, 7, 13, and 14. Of this subgroup 43.3% (13/30) completed this subtest. The mean score was 8.3, 95% CI [3.9, 12.7] of the maximum score of 32.

DISCUSSION

The results of the BBS reported in this study concern a functionally more able part of the total study sample. Half of the participants of the total HA-ID study sample (n = 1050) were not able to complete all 14 items of the BBS, largely due to physical limitations and limited cognitive ability. Since physical limitations and more severe level of ID are associated with poor balance capacities [3], the BBS results are likely to be an overestimation of the balance capacities of the population of older adults with ID as a whole. Feasible subtests of the BBS were identified for subgroups with low completion rates on the original BBS to enable some balance assessment in these individuals.

For this functionally more able part of the study sample (n = 508, mean age 61.3 [*sd* = 7.6]), the mean BBS total score was 47.2, 95% CI [46.3, 48.0]. This result is comparable to the result found by Enkelaar et al. (2013), in a group of 76 older adults with mild to moderate ID (43 males, mean age 63.1 years [*sd* = 7.6]). They found a mean BBS score of 46.8 (*sd* = 6.9), which was significantly lower than the BBS score of 55.8 (*sd* = 0.4) in a control group with normal intelligence (n = 20, 14 male, mean age 62.2 years [*sd* = 5.6]) [19]. As a comparison, 113 residents of a home for the elderly without ID (20 males) with a mean age of 83.5 years (*sd* = 5.3), a mean BBS score of 47.7 (*sd* = 5.5) was also found

[12]. Older adults with ID thus seem to have balance capacities that are comparable to or worse than those in adults without ID who are on average 20 years older and not capable of living independently.

Of the functionally more able part of our study sample, 21% had an increased risk of falling according to the cut-off value of 45. However, we question the use of this cut-off in the population of (older) adults with ID, because it is based on the assumption of a predominantly age-related decline in balance capacities. In the general population, balance capacities decrease with age, along with cognitive decline [30, 31]. Increasing age and cognitive decline are also found as risk factors for falls [32]. This coexistence of decline in balance capacities and cognitive capacities is supported by the finding that cognitive and motor functioning are fundamentally interrelated, with equally long developmental trajectories, and the use of similar brain structures (dorsolateral prefrontal cortex and neocerebellum) [33]. This interrelation, between cognitive and motor functioning, can also be seen in children with developmental disorders such as ADHD, autism, and language disorders, who often have accompanying motor problems, for example problems with motor coordination and timing, balance, and rapidly alternating movements [33]. Individuals with ID have lifelong cognitive impairments which may influence their balance capacities during their entire life, and not just at an older age. The age-related decline in balance capacities and increase in fall risk may therefore be different in individuals with ID, because of this different cognitive component, and leading to possibly different compensation strategies. Therefore, the cut-off at which balance impairments lead to an increased risk of falling may be different in this population, emphasizing the need for validation of the cut-off value of 45 for this population.

This study showed that females have poorer balance capacities than males and that balance capacities decrease with increasing age. Since a poor balance is a risk factor for falling [12], these results are in line with previous studies finding a higher prevalence of falls in females and with increasing age, both in individuals with ID [3] and in the general population [34-36]. Other risk factors for falls in individuals with ID were being female, more severe level of ID, having arthritis, a heart condition, back pain, urinary incontinence, a seizure disorder, polypharmacy, and mobility and strength impairments [3] older age [1, 5], past fractures [1], abnormal gait pattern [2], and ambulatory status [5]. Identification of risk factors helps to target fall prevention programs to those in need of these programs. Guidelines and recommendations for fall prevention have been developed for the general older population [32, 37]. However, to date there are no fall prevention guidelines for the population of adults with ID. The poor balance capacities found in this study underpin the need for these guidelines.

Difficulties understanding the task and physical limitations were most often the reasons for drop-out on the BBS for the subgroups of individuals with severe and profound ID and individuals who use a wheelchair inside their home. Subtests consisting of most feasible items were proposed for these subgroups. The independent items of these subtests all had participation rates over 50%. The participation on the subtests varied from 27.3% to 43.3%. These subtests now meet the feasibility demands proposed by Hilgenkamp et al. (2013) [21]. Furthermore, item 8a, reaching forward to place a ring around a standing stick, seemed to be easier to understand than the original reach forward item, and improves this item's feasibility.

Allowing participants to become familiarized with the test, the tester, and the procedures may help to solve some of the difficulties of participants with understanding the items of the BBS [2]. A modified and shortened version of the BBS was found feasible in individuals with severe intellectual and visual disabilities. After allowing for five practice sessions, 92% of the participants completed all tasks, supporting the need of familiarization sessions for individuals with more severe disabilities [38].

To interpret tests results, norm-referenced and/or criterion-referenced values are needed. Norm-referenced values are summarizing statistics based on large datasets reflecting the performance of a population. These values provide information about the results that can be expected in individuals at different ages and help understanding the rates of change across various age groups, as opposed to criterion-referenced values which are associated with health or performance outcomes [39]. To date, no large scale studies have been performed to provide these values for older adults with ID. On the condition that the characteristics of the study sample will be taken into account (functionally more able part of a fairly sedentary population [Table 2]), BBS results of this study can be used as norm-referenced values for older adults with ID.

The next step is to validate the original BBS, the subtests, and item 8a, to allow use for balance assessment and prediction of fall risk in this population. Cut-off values related to an increased fall risk need to be identified for the original BBS and the subtests. Furthermore, criterion-referenced values, based on associations of BBS results with other health and performance outcomes, are needed to improve interpretation of the BBS results and support decision making for treatment and interventions.

This study shows that even the more functionally able older adults with ID have poor balance capacities similar to adults in the general population who are on average 20 years older. Regular screening can help to identify those individuals with poor balance capacities in need for fall prevention programs to reduce the fall risk and to determine what kind of fall prevention programs are suitable for older adults with ID.

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Chapter 6

The predictive value of physical fitness for falls in older adults with intellectual disabilities

Alyt Oppewal, Thessa I.M. Hilgenkamp, Ruud van Wijck,
Josje D. Schoufour, Heleen M. Evenhuis

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ABSTRACT

A high incidence of falls is seen in people with intellectual disabilities (ID), along with poor balance, strength, muscular endurance, and slow gait speed, which are well-established risk factors for falls in the general population. The aim of this study was to assess the predictive value of these physical fitness components for falls in 724 older adults with borderline to profound ID (≥ 50 years). Physical fitness was assessed at baseline and data on falls was collected at baseline and after three years. Gait speed was lowest in participants who fell three times or more at follow-up. Gait speed was the only physical fitness component that significantly predicted falls, but did not remain significant after correcting for confounders. Falls at baseline and not having Down syndrome were significant predictors for falls. Extremely low physical fitness levels of older adults with ID, possible strategies to compensate for these low levels, and the finding that falls did not increase with age may explain the limited predictive value of physical fitness found in this study.

INTRODUCTION

A high incidence of falls and related injuries is seen in people with intellectual disabilities (ID) [1-5]. Falling is not restricted to the elderly in the population of ID [4], but fall risk does increase with advancing age [1, 6-8].

A number of personal and medical characteristics can lead to an increased fall risk. In people with ID, older age, being female, more severe level of ID, impaired mobility, physically active, back pain, arthritis, fracture history, cerebral palsy, good visuo-motor capacity, good attentional focus, urinary incontinence, heart condition, epilepsy, visual impairments, polypharmacy, and behavioral problems have been mentioned as possible risk factors for falls [1, 3, 5, 7, 9]. Having Down syndrome (DS) was found to reduce the risk for falls and related injury [9].

Next to personal and medical characteristics, physical fitness may be an important aspect for falls in people with ID. Older adults with ID have poor balance, strength, muscular endurance, and slow gait speed [10, 11]. In the general population, these physical fitness components are well-established risk factors for falls [12-18]. However, results from prospective studies performed in the general population may not apply to older adults with ID. The predictive value of physical fitness for falls in the general population is related to an age-related decrease in physical fitness or due to diseases. This relationship may be confounded by the lifelong cognitive impairment of people with ID. This lifelong cognitive impairment may negatively influence their motor development since childhood, which may negatively influence their balance, strength, endurance, and gait throughout their life, and not just at an older age. This line of thinking is supported by the finding that motor and cognitive functioning are fundamentally interrelated, with similar developmental trajectories and the use of similar brain structures [19]. Impairments in physical fitness may not necessarily be related to an increased fall risk in the same amount as in the general population because people with ID may have developed different compensation strategies and utilize them over their entire lifespan. For example, people with DS show more variability in gait than people with normal intelligence, but they use this variability functionally to optimize their movement. This implies the use of different control strategies to compensate for their limitations [20, 21]. Based on this hypothesis, the correlation between a decrease in physical fitness and falls may be less strong.

A recent prospective study investigating risk factors for falling in older adults with mild to moderate ID did not find balance and gait speed to differ between fallers and non-fallers. However, adults who fell indoors, performed worse on balance and gait tests [5]. In contrast, retrospective studies did find strength and gait impairments to be associated with an increased fall risk in people with ID [2, 3, 6]. More knowledge is needed to identify the predictive value of the physical fitness in predicting fall risk. This will help to identify people at risk and thereby the decision-making for treatment.

The aim of this study was to assess the predictive value of balance, gait speed, strength, and muscular endurance for falls, over a 3-year period, in a large sample of older adults with ID.

METHODS

Study design and participants

This study was part of the large Dutch 'Healthy ageing and intellectual disabilities' (HA-ID) study performed in a consort of three ID care organizations in collaboration with two university departments in the Netherlands (Intellectual Disability Medicine, Erasmus MC, University Medical Center Rotterdam and the Center for Human Movement Sciences, University of Groningen, University Medical Center Groningen). For the baseline measurements all 2150 older clients with ID (≥ 50 years) of the care organizations were invited to participate, resulting in a near-representative sample of 1050 clients. Themes in the study were physical activity and fitness, nutrition and nutritional state, and mood and anxiety. Data collection on these themes took place between February 2009 and July 2010. Details about design, recruitment, and representativeness of the sample have been presented elsewhere [22]. Three years after the baseline measurements, follow-up data on falls were collected with a questionnaire.

This study was approved by the Medical Ethical Committee at Erasmus Medical Center (MEC 2008-234 and MEC 2011-309) and by the ethical committees of the participating ID care organizations. Informed consent was obtained from all participants or their legal representatives for both baseline and follow-up measurements; however, unusual resistance was a reason for aborting measurements at all times. This study followed the guidelines of the Declaration of Helsinki [23].

Baseline measurements

Of the risk factors for falls mentioned in the introduction, the following risk factors were collected in the HA-ID study: age, gender, level of ID, Down syndrome (DS), mobility, physical activity levels, fracture history, spasticity of the legs (as an aspect of cerebral palsy), urinary incontinence, heart condition, epilepsy, visual impairments, polypharmacy, and behavioral problems. In this study, these risk factors are used to describe the study sample and as possible confounders in the analyses.

Personal characteristics

Age and gender were collected from administrative systems of the care organizations. Level of ID was categorized by behavioral therapists or psychologists as borderline (IQ = 70 – 84), mild (IQ = 50 – 69), moderate (IQ = 35 – 49), severe (IQ = 20 – 34), or profound (IQ < 20) [24]. The presence of DS was collected through the medical files. Professional caregivers provided information about mobility (independent, walking with an aid, or wheelchair-bound). Physical activity was measured with pedometers (NL-1000 pedometer, New Lifestyles, Missouri, USA). A minimum of 7500 steps per day was classified as sufficient.

Medical information

Professional caregivers provided information about urinary incontinence. Information on the history of fractures and the presence of spasticity of the legs, heart condition (arrhythmias and coronary heart disease), epilepsy, visual impairments, and polypharmacy was retrieved from medical files. Polypharmacy was defined as taking four or more medications. The presence of behavioral problems was obtained from behavioral therapists' records.

Physical fitness

Balance was measured with the Berg Balance Scale (BBS) [25, 26]. The BBS consists of 14 static and dynamic functional balance tasks varying in difficulty, ranging from unsupported sitting in a chair to tandem stance and standing on one leg. The original test instructions were followed with some aids to enhance understanding of the tasks: two carpet feet and a carpet circle on the floor, to point out where the participant had to stand or turn around on. Walking aids were not allowed. The items were scored on a 5-point scale from 0 (inability to complete the task) to 4 (completion of the task) points, with a maximum of 56 points. A score of 45 is used in the general population as a cut-off to differentiate between those at risk for falls (< 45) and those not at risk for falls (≥ 45) [25]. Validity and reliability has been previously demonstrated in the general population [27-29]. In the population with ID, the BBS was also reliable [30, 31] and feasible for older adults with borderline to moderate ID [32, 33].

Comfortable gait speed (GSC) was measured over a distance of 5 meters, after 3 meters for acceleration [34]. Participants walked three times and the average gait speed in m/s was the result of the test. Participants had to walk without someone walking alongside or physically supporting them to avoid influencing their comfortable speed. Validity and reliability in the general population is good [35-39]. In older adults with ID, measuring GSC was feasible and reliable (ICC of 0.96 for same-day interval and 0.93 for 2-week interval) [33, 40].

Muscular endurance was measured with the 30s Chair stand test (30sCS) [41]. Participants had to stand upright and sit down again as often as possible in 30 seconds without using their hands. The total number of complete stances was the result of the test. Validity and reliability has been demonstrated in the general population [42]. In older adults with ID, feasibility and test-retest reliability was moderate to good (ICC of 0.72 for same-day interval and 0.65 for 2-week interval) [33, 40].

Grip strength (GS) was measured with the Jamar Hand Dynanometer (#5030J1, Sammons Preston Rolyan, USA). Participants squeezed the dynamometer with maximum force in seated position, according to the recommendations of The American Society of Hand Therapists [43], three times for both hands with a one-minute pause between attempts. Participants were able to practice by squeezing a rubber ball, to assure understanding. The best result was recorded (in kg). The result was only recorded if the test instructor was convinced the participant had squeezed with maximum effort. Validity and reliability in the general population is good [44, 45]. In older adults with ID, measuring grip strength was feasible and test-retest reliability was good (ICC of 0.94 for same-day interval and 0.90 for 2-week interval) [33, 40].

The physical fitness assessment was conducted at locations familiar or close to participants. Tests were guided by trained test instructors, who all were physiotherapists, physical activity instructors, or occupational therapists with experience with people with ID. To motivate the participants we prescribed 'maximal motivation' to the test instructors for all tests. In some cases, this meant that participants were motivated to engage in the tests by constant verbal encouragement and verbal rewarding, in other cases the test instructor had to remain very calm and quiet to motivate the participant as much as possible and to prevent stress or anxiety. The background, knowledge, and experience of the test instructors were important for ensuring the most suitable 'maximal motivation' for every participant, while regarding safety as well.

Baseline fall assessment

A fall was defined as an unexpected event in which the participant comes to rest on the ground, floor, or lower level [46]. Professional caregivers provided information on how often the participants fell in the last three months (not fallen, 1 – 2 falls, 3 – 5 falls, 6 – 10 falls, 11 falls or more) before the baseline measurements. We chose for a recall period of three months because we expected this period to be long enough to distinguish non-fallers from recurrent fallers, because of the high fall incidence in people with ID [6].

Follow-up fall assessment

Professional caregivers provided information at three years after baseline on how often the participant fell in the last three months (not fallen, 1 – 2 falls, 3 – 5 falls, 6 – 10 falls, 11 falls or more).

Statistical analyses

Falls at baseline and follow-up were recoded into three categories: 'non-fallers', 'one or two time fallers', and 'three times or more fallers'. Baseline personal characteristics, medical information, falls, and physical fitness were described for the three categories of fallers at follow-up. Differences between groups were analyzed with Pearson's chi-square tests and one-way independent analysis of variance (ANOVA), with post hoc tests. Bonferroni correction was used to correct for multiple testing. Spearman's correlation coefficient was calculated to assess the correlation between falls at baseline and follow-up.

Simple and multiple logistic regression analyses were used to assess the predictive value of each physical fitness component for falling at follow-up. Because the aim was to predict falling, falls at baseline and follow up were recoded into categories non-fallers and fallers (≥ 1 fall).

First, the predictive value of physical fitness for falls was assessed with a simple logistic regression, with each physical fitness component as the independent variable and falls at follow-up as the dependent variable.

Second, a multiple logistic regression was performed to adjust for confounders. Spearman's correlation coefficients were calculated between baseline personal characteristics and medical information and falls at follow-up to identify potential confounders. Variables that significantly correlated with falls at follow-up were considered potential confounders. Dummy variables were constructed for independent categorical variables with more than two categories. Age, gender, and the potential confounders were entered in the first block, and each physical fitness component was entered in the second block. Multicollinearity was checked with the Variance Inflation Factor (VIF), which had to be below 10 for all independent variables [47]. Results are presented as unstandardized coefficients (*B*) and their standard errors (*SE)*, representing the strength of the relation between each independent variable and the outcome; odds ratios (provided as *Exp(B)* by SPPS) and its confidence intervals, representing the magnitude of the influence of the predictors and is defined as the change in odds resulting from a unit change of the predictor; the explained variance by the model (Cox & Snell R^2 and Nagelkerke R^2); and the model chi-square statistic, representing the fit of the model [47].

Statistical significance was set at 5% ($p < 0.05$). Analyses were performed with the Statistical Package for Social Sciences (SPSS) version 21 (IBM Corporation, New York).

RESULTS

Baseline characteristics and falls

Of the 1050 participants in the HA-ID study follow-up fall data were available from 724 participants. Of these participants, 25.5% (185 participants) fell at follow-up, of whom

Table 1. Baseline personal characteristics categorized according to follow-up fall data.

	n		Non-fallers (*n* = 539)	1 – 2x fallers (*n* = 142)	≥3x fallers (*n* = 43)
Personal characteristics					
Age	724	Years (*m* ± *sd*)	60.7 ± 7.8	61.5 ± 6.8	63.2 ± 8.0
Gender	724	Female	261 (48.4%)	74 (52.1%)	25 (58.1%)
		Male	278 (51.6%)	68 (47.9%)	18 (41.9%)
Level of ID	707	Borderline	13 (2.4%)	5 (3.5%)	0
		Mild	104 (19.3%)	33 (23.2%)	7 (16.3%)
		Moderate	263 (48.8%)	72 (50.7%)	24 (55.8%)
		Severe	100 (18.6%)	22 (15.5%)	7 (16.3%)
		Profound	47 (8.7%)	5 (3.5%)	5 (11.6%)
Down syndrome	619	Yes	80 (14.8%)	11 (7.7%)	1 (2.3%)
Mobility	694	Independent	407 (75.5%)	105 (73.9%)	24 (55.8%)
		Walking-aid	63 (11.7%)	22 (15.5%)	12 (27.9%)[a]
		Wheelchair	52 (9.6%)	4 (2.8%)[b]	5 (11.6%)
Physical activity	191	Steps/day (*m* ± *sd*)	6830.1 ± 3772.9	8003.5 ± 4115.0	5682.7 ± 2207.8
Medical information					
Fracture history	626		39 (7.2%)	12 (8.5%)	8 (18.6%)
Spasticity legs	632	Unilateral	13 (2.4%)	4 (2.8%)	4 (9.3%)
		Bilateral	39 (7.2%)	5 (3.5%)	2 (4.7%)
Urinary incontinence	694		231 (42.9%)	60 (42.3%)	23 (53.5%)
Heart condition	640		16 (3.0%)	7 (4.9%)	0
Epilepsy	636		85 (15.8%)	29 (20.4%)	18 (41.9%)[a]
Visual impairments	627		121 (22.5%)	20 (14.1%)	8 (18.6%)
Polypharmacy	649		225 (41.7%)	77 (54.2%)	21 (48.8%)
Behavioral problems			148 (27.5%)	39 (27.5)	10 (23.3)
Falls at baseline					
Falls in 3 months before baseline?	689	None	436 (80.9%)	82 (57.7%)	15 (34.9%)
		1 or 2 times	70 (13.0%)	36 (25.4%)	10 (23.3%)
		≥3 times	13 (2.4%)	11 (7.7%)	16 (37.2%)
Physical fitness					
Balance	352		*n* = 271	*n* = 63	*n* = 18
		m ± *sd*	47.4 ± 10.3	47.3 ± 8.7	43.7 ± 11.8
Comfortable gait speed	506		*n* = 386	*n* = 92	*n* = 28
		m/s (*m* ± *sd*)	0.996 ± 0.346	0.935 ± 0.323	0.784 ± 0.362[c]
Grip strength	512		*n* = 389	*n* = 94	*n* = 29
		kg (*m* ± *sd*)	24.4 ± 10.4	24.4 ± 10.3	22.8 ± 8.8
Muscular endurance	383		*n* = 296	*n* = 68	*n* = 19
		m ± *sd*	9.4 ± 3.2	10.1 ± 3.8	7.7 ± 2.9

m = mean; *sd* = standard deviation; *n* = number of participants; ID = intellectual disability.

[a] Observed value significantly higher than expected value.

[b] Observed value significantly lower than expected value.

[c] Significantly different from non-fallers.