

**Cardiorespiratory fitness in individuals with intellectual disabilities – A review**

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Authors: Alyt Oppewal<sup>a</sup>, Thessa I.M. Hilgenkamp<sup>a,b</sup>,

Ruud van Wijck<sup>c</sup>, Heleen M. Evenhuis<sup>a</sup>

<sup>a</sup> Intellectual Disability Medicine, Department of General Practice, Erasmus Medical Center

Rotterdam, P.O. Box 2040, 3000 CA Rotterdam, The Netherlands

<sup>b</sup> Abrona, Amersfoortseweg 56, 3712 BE Huis ter Heide, The Netherlands

<sup>c</sup> Center for Human Movement Sciences, University of Groningen, University Medical Center

Groningen, A. Deusinglaan 1, 9713 AV Groningen, The Netherlands

Corresponding author:

Alyt Oppewal at Erasmus Medical Center, Department of General Practice, Intellectual

Disability Medicine, P.O. Box 2040, 3000 CA Rotterdam, The Netherlands.

Tel.: +31 107043611

E-mail address: a.oppewal@erasmusmc.nl (A. Oppewal).

**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.

**Keywords:** Cardiorespiratory fitness, intellectual disabilities, testing, heart rate, oxygen uptake

## 1. Introduction

Cardiorespiratory fitness is the ability of the circulatory and respiratory systems to supply oxygen to working muscles during sustained physical activity is called cardiorespiratory fitness (U.S. Department of Health and Human Services [DHHS], 2008). Poor cardiorespiratory fitness is a major independent risk factor for cardiovascular diseases and all-cause mortality (DHHS, 2008; Lee, Artero, Sui, & Blair, 2010; World Health Organization [WHO], 2007). Cardiovascular diseases are the number one cause of disability and premature morbidity and mortality throughout the world (WHO, 2007). Physical activity levels (Hilgenkamp, Reis, van Wijck, & Evenhuis, 2012; Peterson, Janz, & Lowe, 2008; Temple, Frey, & Stanish, 2006) and cardiorespiratory fitness of individuals with intellectual disabilities (ID) are low (Fernhall & Pitetti, 2001; Mendonca, Pereira, & Fernhall, 2010), which puts them at a high risk for cardiovascular diseases.

The standard measure to express cardiorespiratory fitness is  $\dot{V}O_2\text{max}$ , 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 (Takken, 2007). It is the product of the maximal cardiac output and arterial-venous oxygen difference. Variations in  $\dot{V}O_2\text{max}$  are primarily due to differences in maximal cardiac output (American College of Sports Medicine [ACSM], 2010), which is the product of heart rate (HR) and stroke volume (SV) (Powers & Howley, 2007). The highest achieved oxygen uptake during a maximal cardiorespiratory exercise test can be considered as  $\dot{V}O_2\text{max}$  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  $\dot{V}O_2\text{max}$  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 ( $HR_{\text{max}}$ ), (b) respiratory exchange ratio (RER) > 1.0, and (c) high levels of lactic acid in the minutes following exercise (Fernhall, Tymeson, Millar, &

Burkett, 1989; Howley, Bassett, & Welch, 1995; Powers & Howley, 2007; Takken, 2007).

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) (Pitetti, Rimmer, & Fernhall, 1993).

Ideally,  $\dot{V}O_2$ max is measured directly with a maximal exercise test on an treadmill or bicycle ergometer with open-circuit spirometry under standardized conditions (ACSM, 2010). 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 (ACSM, 2010). 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 (Draheim, Laurie, McCubbin, & Perkins, 1999; Fernhall et al., 2001; Kittredge, Rimmer, & Looney, 1994).

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 (Pitetti et al., 1993; Rintala, McCubbin, & Dunn, 1995). 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 (Rintala et al., 1995).

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 (Fernhall & Pitetti, 2001). 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, Pereira, & Fernhall (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 (Mendonca et al., 2010). 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 (RFT) 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 (Pitetti et al., 1993). 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.

## 2. Methods

### 2.1 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'].

### 2.2 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.

[Insert figure 1 here]

### **2.3 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 (Faas & Helder, 1990).

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 analysis 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.

### **3. Results**

Thirteen studies were included that reported on cardiorespiratory fitness of individuals with ID (table 1). One study was a cohort study (Acampa et al., 2008) and one a case-study (Bricout et al., 2008). All other studies had a cross-sectional design. Two studies used a submaximal bicycle protocol (Ohwada, Nakayama, Suzuki, Yokoyama, & Ishimaru, 2005; Wade, De Meersman, Angulo, Lieberman, & Downey, 2000), and three used field tests (Graham & Reid, 2000; Hilgenkamp, van Wijck, & Evenhuis, 2012b; van de Vliet et al., 2006). Five studies used a maximal treadmill protocol (Baynard, Pitetti, Guerra, & Fernhall, 2004; Baynard, Pitetti, Guerra, Unnithan, & Fernhall, 2008; Bricout et al., 2008; Fernhall et al., 2001; Patel et al., 2007). The other three studies reported on possible determinants for cardiorespiratory fitness without performing an aerobic test (Acampa et al., 2008; Guideri et al., 2004; Vis et al., 2012). Most studies included adolescents and (young) adults with mild to

moderate ID. But three studies included children (Acampa et al., 2008; Baynard et al., 2008; Guideri et al., 2004) and three studies included adults over the age of 50 (Graham & Reid, 2000; Hilgenkamp, van Wijck, et al., 2012b; Vis et al., 2012). Three studies also included participants with severe and/or profound ID (Hilgenkamp, van Wijck, et al., 2012b; Ohwada et al., 2005; Vis et al., 2012). These studies are discussed below in the topics ‘cardiorespiratory fitness levels’ and ‘determinants for cardiorespiratory fitness levels’.

### 3.1 Cardiorespiratory fitness levels

Fernhall et al. (2001) reported a significantly lower  $\dot{V}O_{2\text{peak}}$  ( $33.8 \pm 10.6 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ ) and HR<sub>peak</sub> ( $177 \pm 15 \text{ bpm}$ ) in young adults with ID ( $n = 276$ ) than in older controls without ID ( $n = 296$ ,  $\dot{V}O_{2\text{peak}}$   $35.6 \pm 10.9 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ , and HR<sub>peak</sub>  $185 \pm 12 \text{ bpm}$ ), measured with a maximal treadmill protocol.

Ohwada et al. (2005) showed that the estimated  $\dot{V}O_{2\text{max}}$ , based on results of a submaximal bicycle protocol, was also lower in adult males with ID ( $n = 23$ ,  $\dot{V}O_{2\text{max}}$   $34.0 \pm 9.0 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ ) than in controls without ID ( $n = 23$ ,  $\dot{V}O_{2\text{max}}$   $41.3 \pm 10.0 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ ). 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  $\dot{V}O_{2\text{max}}$  results of the general population (Graham & Reid, 2000; Hilgenkamp, van Wijck, et al., 2012b). These studies used equations to estimate  $\dot{V}O_{2\text{max}}$  developed for the general population. However, these equations may not be appropriate for individuals with ID, because they tend to overestimate  $\dot{V}O_{2\text{max}}$  (Climstein, Pitetti, Barrett, & Campbell, 1993; Kittredge et al., 1994).

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 (van de Vliet et al., 2006). 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 (van de Vliet et al., 2006). 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,  $n = 59$ )  $\dot{V}O_{2\text{peak}}$  ( $39.8 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ ) and HR<sub>peak</sub> (189 bpm) values were significantly lower than in controls without ID ( $45.7 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$  and 196 bpm). In the older age groups (16 to 21,  $n = 37$ ; 22 to 29,  $n = 50$ ; and 30 to 45 years,  $n = 34$ )  $\dot{V}O_{2\text{peak}}$  values ( $40.9$ ,  $34.4$ , and  $30.8 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$  respectively) were not significantly different from controls without ID ( $41.1$ ,  $36.8$ , and  $29.9 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$ ). However, the reported  $\dot{V}O_{2\text{peak}}$  of the control group in this study was lower than normative values from other studies (ACSM, 2010; Cureton, Sloniger, O'Bannon, Black, & McCormack, 1995). Comparing  $\dot{V}O_{2\text{peak}}$  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  $\dot{V}O_{2\text{peak}}$  than the general population, across all the age groups.

Baynard et al. (2008) also showed that  $\dot{V}O_{2\text{peak}}$  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  $\dot{V}O_{2\text{max}}$ , with results from the Canadian home fitness step test, of middle-aged adults with mild to moderate ID over a 13 year period ( $n = 32$ ). Estimated  $\dot{V}O_{2\text{max}}$  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 (Graham & Reid, 2000). 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 ( $41.2 \pm 11.2 \text{ ml} \cdot \text{kg}^{-1} \cdot \text{min}^{-1}$ ) than in females ( $30.8 \pm 7.7 \text{ ml} \cdot \text{kg}^{-1} \cdot \text{min}^{-1}$ ). HRpeak was not significantly different:  $166 \pm 38$  bpm in males and  $170 \pm 23$  bpm in females (Draheim et al., 1999).

Differences in cardiorespiratory fitness in ID may be syndrome-specific, as can be seen in individuals with DS (Mendonca et al., 2010). 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 (Wade et al., 2000). 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 ( $49.5 \text{ ml} \cdot \text{kg}^{-1} \cdot \text{min}^{-1}$ ), above the 75<sup>th</sup> percentile of  $\dot{V}O_2$ peak values in the general population, of a 24 year old male with Fragile-X syndrome (ACSM, 2010; Bricout et al., 2008). 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.

### **3.2 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 (DHHS, 2008; WHO, 2007). However, the fact that trained athletes with ID also have low levels of cardiorespiratory fitness (van de Vliet et al., 2006) 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. Owhada 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 (Ohwada et al., 2005). 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 (Baynard et al., 2008;

Fernhall et al., 2001). 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]) (ACSM, 2010; Powers & Howley, 2007), 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 (Brubaker & Kitzman, 2011). 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 (Wade et al., 2000). 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 ( $n = 32$ ) (Acampa et al., 2008), or low serotonin plasma levels ( $n = 28$ ) (Guideri et al., 2004).

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 (Fernhall & Pitetti, 2001). We found one study of the relation between leg strength and  $\dot{V}O_2$ peak (Nasuti, Stuart-Hill, & Temple, 2012) (table 2). This was non-significant, contradicting previous results (Fernhall & Pitetti, 2001). However, this study had a small sample size ( $n = 13$ ), which does not allow to draw 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.

### **3.3 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 (Climstein et al., 1993; Mac Donncha, Watson, McSweeney, & O'Donovan, 1999; Pastore et al., 2000; Pitetti, Millar, & Fernhall, 2000; Vis et al., 2009), 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 (Hilgenkamp, van Wijck, & Evenhuis, 2012a; Vis et al., 2009; Waninge, Evenhuis, van Wijck, & van der Schans, 2011). Six studies included participants with severe to profound ID (Casey, Wang, & Osterling, 2012; Hilgenkamp, van

Wijck, et al., 2012a; Pastore et al., 2000; Pitetti, Jongmans, & Fernhall, 1999; Vis et al., 2009; Waninge et al., 2011). 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  $\dot{V}O_2\text{max}$  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  $\dot{V}O_2\text{peak}$ .

### *3.3.1 American College of Sports Medicine (ACSM) equations*

Non-invasive and safe procedures to establish  $\dot{V}O_2\text{max}$  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  $\dot{V}O_2\text{max}$  by 129% in young adults with ID ( $n = 17$ ) and 184% in young adults with DS ( $n = 15$ ) (Climstein et al., 1993). Therefore, these equations can not be used to estimate  $\dot{V}O_2\text{max}$  of individuals with ID.

### *3.3.2 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 (Pitetti et al., 1999).



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 (Pastore et al., 2000). 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.

### *3.3.3 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 ( $n = 23$ ) (Pitetti et al., 2000). 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  $\dot{V}O_2\text{peak}$ ,  $HR\text{peak}$ ,  $\dot{V}E\text{peak}$ , and  $RER\text{peak}$  ( $r = 0.85$  to  $0.99$ ), similar or higher to reliability results in controls without ID ( $r = 0.51$  to  $0.99$ ) (Pitetti et al., 2000).

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 (Pitetti et al., 2000).

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.

#### *3.3.4 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 (Draheim et al., 1999; Kittredge et al., 1994; Rintala, McCubbin, Downs, & Fox, 1997; Teo-Koh & McCubbin, 1999).

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  $0.91$  (Draheim et al., 1999; Rintala, Dunn, McCubbin, & Quinn, 1992).

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 (Draheim et al., 1999;

Kittredge et al., 1994; Kline et al., 1987). 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  $\dot{V}O_{2\text{peak}}$  ( $n = 40$ ). 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  $\dot{V}O_{2\text{peak}}$  values in 74% to 79% of the participants (Rintala et al., 1997). However, this same equation overestimated  $\dot{V}O_{2\text{peak}}$  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.

### *3.3.5 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 (Léger & Lambert, 1982). The original 20m shuttle run test was found reliable for children ( $r = 0.97$ ) (Fernhall et al., 1998) and adolescent males ( $r = 0.94$ ) with mild to moderate ID (Mac Donncha et al., 1999). 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 ( $r = 0.96$ ) (Fernhall et al., 1998), adults with severe to profound ID and visual disabilities ( $r = 0.96$ ) (Waninge et al., 2011), and older adults with borderline to profound ID ( $r = 0.76-0.90$ ) (Hilgenkamp, van Wijck, et al., 2012a), respectively.

The 16m and 20m shuttle run tests correlated significantly with  $\dot{V}O_{2\text{peak}}$  ( $r = 0.77$  and  $0.74$ ) and developed equations to estimate  $\dot{V}O_{2\text{peak}}$  explained 77% to 79% of the variance of measured  $\dot{V}O_{2\text{peak}}$ . Cross-validation of the equation of the 20m shuttle run test showed poor

level of agreement between estimated and measured  $\dot{V}O_{2\text{peak}}$ , with a possible underestimation of  $13.5 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$  or overestimation of  $10.8 \text{ ml}\cdot\text{kg}^{-1}\cdot\text{min}^{-1}$  of measured  $\dot{V}O_{2\text{peak}}$  (Fernhall, Pitetti, Millar, Hensen, & Vukovich, 2000). This same equation significantly overestimated  $\dot{V}O_{2\text{peak}}$  in a sample of adolescents with DS ( $n = 26$ ) (Guerra, Pitetti, & Fernhall, 2003). This may be so because individuals with DS have lower levels of cardiorespiratory endurance than individuals with ID (Baynard et al., 2004; Baynard et al., 2008; Fernhall et al., 2001; Fernhall & Pitetti, 2001). In another study of boys and adolescent men with DS ( $n = 53$ ), the 20m shuttle run test did not provide an accurate estimate of  $\dot{V}O_{2\text{max}}$ , as shown by a low explained variance (23%) of measured  $\dot{V}O_{2\text{peak}}$  and large errors in individual estimation of  $\dot{V}O_{2\text{max}}$  (Agiovlasitis, Pitetti, Guerra, & Fernhall, 2011). These results show that estimating  $\dot{V}O_{2\text{max}}$  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 (Hilgenkamp, van Wijck, et al., 2012a).

### 3.3.6 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 (Casey et al., 2012; Nasuti et al., 2012; Waninge et al., 2011).

Nasuti et al. (2012) reported that the 6MWT was a reliable ( $r = 0.98$ ) and valid indicator of cardiorespiratory fitness in adults with ID ( $n = 13$ ) (Nasuti et al., 2012). The

walked distance correlated significantly with  $\dot{V}O_2\text{peak}$ , measured with a maximal treadmill test, and accounted for 67% of the variance in  $\dot{V}O_2\text{peak}$  (Nasuti et al., 2012). Participants reached 87.5% of their estimated maximal HR, estimated with the population specific formula of Fernhall et al. (2001). Waning 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)(Waning et al., 2011).

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) (Casey et al., 2012). A learning effect was found, implying the need for familiarization sessions (Casey et al., 2012).

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 (Vis et al., 2009). 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.

### *3.3.7 600 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) (Fernhall et al., 1998) and outcomes significantly correlate to measured  $\dot{V}O_{2\text{peak}}$  ( $r = -0.80$ ). The equation to estimate  $\dot{V}O_{2\text{max}}$  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  $\dot{V}O_{2\text{max}}$  from these (submaximal) field tests is problematic.

#### **4. 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  $\dot{V}O_2\text{max}$ . 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 (ACSM, 2010; Draheim et al., 1999; Graham & Reid, 2000). Also, gender differences were found in the age-related decline of cardiorespiratory fitness (Graham & Reid, 2000) 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 (ACSM, 2010; Blair et al., 1995). 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.

**Conflicts of interest**

None



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

Study	Design	Participants <i>n</i>	Age	Level of ID	Exercise test	$\dot{V}O_{2peak}$ (ml·kg <sup>-1</sup> ·min <sup>-1</sup> )	HR <sub>peak</sub> (bpm)	Outcomes
Hilgenkamp et al. (2012b)	Cross-sectional	ID: <i>n</i> =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 $\dot{V}O_{2max}$ above the lower limit of the average range in the general population.
Vis et al. (2012)	Cross-sectional, repeated measures	ID no DS: <i>n</i> =25, 15 males DS: <i>n</i> =96, 46 males No ID: <i>n</i> =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: <i>n</i> =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: <i>n</i> =180 DS: <i>n</i> =133 No ID: <i>n</i> =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 (50 <sup>th</sup> percentile scores)	189 183 178 176 (50 <sup>th</sup> percentile scores)	Similar $\dot{V}O_{2peak}$ , 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.
Patel et al.	Cross-	PWS: <i>n</i> =9, 8 males	28±3	NR	Intermittent maximal treadmill	-	-	Five individuals with PWS

(2007)	sectional	<b>No ID:</b> $n=9$ , 8 males	$28\pm 3$		protocol: 1) 1 MET workload increase every 2 min. Alternated periods of walking and rest No familiarization sessions Objective criteria: none			had diminished exercise capacity (achieved 4 METs or less).
Van de Vliet et al. (2006)	Cross-sectional	<b>Athletes with ID:</b> $n=313$ , 231 males (136 cardiovascular endurance test) Comparison with data of the general population	$23\pm 5$	NR	20m shuttle run test No familiarization sessions	-	NR	Significantly lower cardiorespiratory endurance (completed laps) for athletes with ID. HR <sub>peak</sub> was equal to Canadian controls, but significantly lower than Japanese controls.
Ohwada et al. (2005)	Cross-sectional	<b>ID males:</b> $n=23$ <b>No ID males:</b> $n=23$	$36\pm 9$ $36\pm 8$	Mild-severe	Submaximal bicycle protocol at 40, 50 and 60% of estimated $\dot{V}O_2$ max No familiarization sessions	$34.0\pm 9.0$	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	<b>Rett:</b> $n=28$ females <b>No ID:</b> $n=60$	$7\pm 3$ $7\pm 2$	NR	None (in rest)	-	-	Low plasma serotonin levels are correlated to sympathetic overactivity.
Baynard et al. (2004)	Cross-sectional	<b>ID no DS:</b> $n=15$ , 8 males <b>DS:</b> $n=16$ , 10 males	$20\pm 2$ $21\pm 1$	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	$34.3\pm 2.8$	$179\pm 4$	Increase in HR from rest to submaximal exercise levels was primarily through parasympathetic withdrawal. Appropriate cardiac response.
Fernhall et al. (2001)	Cross-sectional	<b>ID:</b> $n=276$ , 97 DS <b>No ID:</b> $n=296$	$22\pm 8$ $31\pm 9$	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	$33.8\pm 10.6$	$177\pm 15$	The formula $210-0.56(\text{age})-15.5(\text{DS})$ can estimate HR <sub>max</sub> for the ages 8-46 years with similar accuracy as in the general population. Lower $\dot{V}O_2$ peak, HR <sub>max</sub> , RER <sub>max</sub> , $\dot{V}E$ max for individuals with ID.

Graham & Reid (2000)	Cross-sectional	<b>ID:</b> $n=32$ , 4 DS, 18 males Comparison with data of the general population	$41 \pm 10$	Mild-moderate	Canadian home fitness step test 1 familiarization session	♂: $33.7 \pm 3.8$ ♀: $26.3 \pm 3.7$	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	<b>PWS:</b> $n=26$ , 11 males <b>No ID:</b> $n=26$ , 11 males	$21 \pm 10$ $22 \pm 12$	NR	2 min submaximal bicycle protocol at HRs of 100-120 bpm No familiarization sessions	-	$108 \pm 20$	Individuals with PWS had lower HR, SBP and DBP, however, they had normal autonomic modulation of HR, during exercise.

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$n$  = 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.

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

Study	Design	Participants <i>n</i>	Age	Level of ID	Exercise test	$\dot{V}O_{2peak}$ ( $ml \cdot kg^{-1} \cdot min^{-1}$ )	HRpeak (bpm)	Outcomes
Casey et al. (2012)	Cohort, repeated measures	<b>DS:</b> <i>n</i> =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	<b>ID males:</b> <i>n</i> =12 <b>ID females:</b> <i>n</i> =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	<b>ID:</b> <i>n</i> =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 $\dot{V}O_{2peak}$ .
Agiovlasitis et al. (2011)	Cohort	<b>DS:</b> <i>n</i> =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 $\dot{V}O_{2peak}$ (mean group $\dot{V}O_{2peak}$ was accurately estimated, but inaccurate individual prediction).
Waninge et al. (2011)	Cohort, repeated measures	<b>ID males:</b> <i>n</i> =29 <b>ID females:</b> <i>n</i> =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

Vis et al. (2009)	Cross-sectional, repeated measures	<b>DS with mild or no cardiac disease:</b> $n=52$ , 38 males <b>DS with severe cardiac disease:</b> $n=29$ , 15 males	41±11 36±10	Mild-profound	6MWT No familiarization sessions	-	-	impairments. 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	<b>DS:</b> $n=26$ , 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 Familiarization 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	<b>ID:</b> $n=17$ , 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.
Pastore et al. (2000)	Cross-sectional	<b>DS:</b> $n=42$ , 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	<b>ID males:</b> $n=12$ <b>ID females:</b> $n=11$ <b>No ID:</b> $n=23$ , 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	♂: 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 $\dot{V}E$ peak of males with ID

Author (Year)	Study Design	Participants	Mean (SD)	Severity	Protocol	Male Mean (SD)	Female Mean (SD)	Notes
Draheim et al. (1999)	Cohort, repeated measures	<b>ID males:</b> $n=10$ <b>ID females:</b> $n=10$	21±3 22±3	Mild-moderate	Objective criteria: RER>1.0 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	was significantly lower than of males without ID. Reliable results for completion time and end HRs of the RFWT ( $r=0.94-0.91$ ). High correlation between RFWT completion time and $\dot{V}O_{2peak}$ ( $r=-0.73$ to $-0.75$ ). Low correlation between end HR and measured $\dot{V}O_{2peak}$ ( $r=0.16-0.33$ ). Overestimation of $\dot{V}O_{2peak}$ when estimated from RFWT results.
Mac Donncha et al. (1999)	Cross-sectional, repeated measures	<b>ID:</b> $n=63$ males <b>No ID:</b> $n=22$ 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).
Pitetti et al. (1999)	Cohort, repeated measures	<b>ID:</b> $n=18$ , 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 $\dot{V}O_{2peak}$ test-retest reliability was not as high ( $r=0.77$ ).
Teo-Koh & McCubbin (1999)	Cohort, repeated measures	<b>ID males:</b> $n=40$ , 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 $\dot{V}O_{2peak}$ and RFWT completion time ( $r=-0.076$ ). Estimating $\dot{V}O_{2peak}$ from RFWT completion time and weight explained 67% of the variance in $\dot{V}O_{2peak}$ .
Fernhall et al. (1998)	Cohort, repeated	<b>ID:</b> $n=34$ , 8 DS, 22 males	14±2	Mild-moderate	600 yard run-walk, 20m and 16m shuttle run test & individualized	36.6±9.1	186±10	Reliable results for the field tests ( $r=0.96-0.98$ ). Field

	measures							
Rintala et al. (1997)	Cohort, cross validation	<b>ID males:</b> $n=19$	27±8	Borderline-moderate	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 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 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 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	38.1±8.3	177±13	test results were significantly related to $\dot{V}O_{2peak}$ ( $r=-0.80, 0.74$ & $0.77$ ). Equations estimating $\dot{V}O_{2peak}$ from field test results explained 74-79% of the variance. Reliable results for RFWT completion time ( $r=0.96$ ) and end HR ( $r=0.87$ ). Estimating $\dot{V}O_{2peak}$ from RFWT significantly underestimated $\dot{V}O_{2peak}$ in 74-79% of the participants.
Kittredge et al. (1994)	Cohort, repeated measures	<b>ID males:</b> $n=12$ <b>ID females:</b> $n=13$	33±7 34±8	Mild-moderate		♂: 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	<b>ID no DS:</b> $n=17, 12$ males <b>DS:</b> $n=15, 12$ males	24±3 26±4	Mild		31.0	175	ACSM equations overestimated $\dot{V}O_{2peak}$ 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; RFWT = Rockport fitness walking test; 6MWT = six-minute walk test; ACSM = American College of Sports Medicine.





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