Effect of continuous aerobic vs interval training on selected functional fitness parameters of adults with intellectual disability and Down syndrome

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DEDICATION

The work presented in this thesis is dedicated to my brother, Rory. I am truly blessed to have you in my life. Thank you for all the joy, altruism and unconditional love that you bring into the lives of those around you.

Lamentations 3-25: The Lord is good unto them that wait for Him, to the soul that seeketh Him. Die Here is goed vir wie op Hom bly hoop, vir die mens wat na Sy wil vra.

"You look at me with pity, concern or indifference, for I am a disabled person.

But you see only the outside of me. If I could express myself, I would tell you what I am inside. I am very much like you.

Think of me first as a person, who hurts and loves and feels joy.

And know I am a person to encourage and direct.
Smile and say hello… even that is enough".

Author unknown

PREFACE

I would like to take this opportunity to honour many people who have made this incredible journey possible.

I humbly thank God, our Creator and Saviour who granted me with this opportunity to enhance my field of expertise with such a special group of people. Without your grace, none of this would have been possible.

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AUTHOR'S CONTRIBUTIONS

This study was made possible through the collective work of a group researchers and academia. The contribution of other academia was kept within reasonable limits as to enable the candidate to submit this thesis for examination purposes. This thesis, therefore serves as fulfilment of the requirements for the PhD degree in Human Movement Science within the Research Focus Area, Physical activity, Sport and Recreation, Faculty of Health Sciences at the North-West University, Potchefstroom Campus.

The table below indicate the contributing work of each co-author.

Responsible person	Role in the study
Mr Boer PhD student	Responsible for the execution of the total thesis Main author of the thesis (including articles 1 to 4) Responsible for ethical application Responsible for testing (descriptive and experimental)(article 1,2 & 4) Responsible for data collection and management (article 1,2 & 4) Statistical analyses (article 1-4)
Prof Moss Promoter	Project coordinator and co-author of articles 1,2 & 4 Significant contribution to the writing of the thesis and manuscripts. Critical review of the content Significant assistance and responsible person for ethics
Prof Calders and his team of researchers (Ghent University, Belgium)	Co-authors of the third article. Responsible for data collection and management of the third article

I declare that I have approved the above mentioned articles and that my role in the study as indicated above is representative of my actual contribution and that I hereby give my consent that it may be published as part of the PhD thesis of P. Boer

Prof S.J. Moss

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SUMMARY

Most individuals with intellectual disability (ID), including those with Down syndrome (DS), are overweight or obese and live a sedentary lifestyle. Non-communicable diseases such as hypertension, diabetes, high total cholesterol and metabolic syndrome have a high prevalence in this group. Individuals with an ID report poor aerobic capacity, with persons with DS reporting even poorer aerobic capacity. A poor aerobic capacity is related to poor cardiovascular fitness, a risk factor for cardiovascular disease. Persons with ID and DS are also reported to have low functional fitness capacities. Functional fitness batteries exist in the general, elderly and ID and physical disabled populations, but standardised tests involving adults with DS do not exist. Individuals with DS are therefore often pooled with ID individuals in spite of discernable functional fitness capacities. It is therefore important that a fitness battery specific for DS is developed.

Structured exercise training presented limited potential to significantly ameliorate anthropometrical and cardiovascular variables in adults with ID and DS. Recently, interval training (IT) has been applied to improve health outcomes of populations at risk for chronic diseases. In this regard the significant physical, anthropometrical and metabolic benefits associated with interval training have been demonstrated in a variety of populations.

The purpose of this study was to: 1) determine the test-retest reliability for a battery of test items in an adult population of DS individuals. 2) Determine the validity of two commonly used field tests (PACER and 6MWD) with the gold standard of cardiorespiratory fitness (direct VO_2 max). 3) Determine the effect of structured IT versus continuous aerobic training (CAT) on various anthropometrical, health, functional and physical benefits in adults with ID and 4) DS.

In the first study, forty-three adults with DS (24 men and 19 women) aged 18–50 years (from Potchefstroom, Pretoria and Brits) completed a battery of tests twice in a two-week period. The battery of tests consisted of two balance items, two flexibility items, five muscular strength and endurance items, two aerobic items, and one functional task. The test-retest relative reliability for all repeated tests was assessed with intraclass correlation coefficient performing one-way analysis of variance. The test-retest absolute variability was measured by using the standard error of measurement (SEM) and reliability data was visualised with a Bland-Altman plot. In the second study, the same forty-three adults with DS randomly performed three aerobic tests on non-consecutive days during a one-week period. To assess validity, peak oxygen uptake was measured directly on a motorised treadmill. The two field tests included the 16-metre PACER and the 6MWD test. Pearson-product moment correlations were performed. A linear regression analysis was conducted to determine criterion-related validity between the field tests and the VO_2 peak test. The third study included 46 persons with ID (30 men and 16 women) from Brugge, Belgium. They were matched on age, gender and IQ and distributed between IT

(n=17), CAT (n=15) and control (n=14). The training groups exercised for 15 weeks, twice weekly (40 minutes). IT was performed in two blocks of 10 minutes (block 1 and 3). Participants performed 10 sprints of 15 seconds (45 seconds of relative rest) on a cycle ergometer. During block 2 continuous training was performed. The CAT group performed three blocks of 10 minutes of continuous training (cycling, walking, stepping). After eight weeks the intensity of training was increased. To evaluate pre-post differences between groups, a repeated ANCOVA with post-hoc Bonferroni test was performed. Lastly, the fourth study included 42 adults with DS (25 men and 17 women) and a mean age 33.8 ± 8.6 years. Participants were randomly allocated to one of three groups (IT, CAT, control). Training was performed for 12 weeks (three times a week). The IT group performed ten-30 second all-out sprints with 90 seconds (1:3 work:rest ratio) of low cadence, low intensity cycling or walking. The CAT group performed continuous cycling and walking at an intensity of 70%–80% of VO_2 peak. To evaluate pre–post differences between groups, a repeated ANCOVA with post-hoc Bonferroni test was performed.

Results showed that all functional fitness tests were feasible and demonstrated excellent test-retest reliability (ICCs>0.9) and acceptable measurement precision (SEM<SD/2). The analyses indicated that there was no major systematic bias in the plots and the scatter around the Bland-Altman was distributed randomly. Linear regression revealed that the 16-metre PACER (R^2=0.86) and the 6MWD (R^2=0.75) were significantly related to directly measured VO_2 peak ($p<0.05$). Both the 16-metre PACER and the 6MWD significantly correlated with VO_2 peak for adults with DS. The relationship was stronger for the 16-metre PACER (r=0.87) than the 6MWD (r=0.78). IT showed a significant positive change for waist circumference (–4.3 cm), body fat (–3.8%), systolic blood pressure (–11 mmHg), lipid profile of total cholesterol (–15 mg/dL), HDL-chol (+4.5 mg/dL) and LDL-chol (–9.4 mg/dL), homeostasis model assessment of insulin resistance (–0.6, peak VO_2 (+0.2 L/min)), peak Watt (+23.8 W), ventilatory threshold (+21 W, +0.2 L/min), 6MWD (+67.7 m) and muscle fatigue resistance (+6.3 s) when compared with no training ($p<0.05$) in adults with ID. Moreover IT for the group with ID demonstrated significant improvements for body fat percentage, systolic blood pressure, low-density lipoprotein, fasting insulin, peak VO_2, and peak power and ventilatory threshold when compared to CAT ($p<0.05$). Results of the fourth study, IT in persons with DS, showed that after 12 weeks of training, body mass (–2 kg) and BMI (–0.8 kg/m^2) decreased significantly more with IT compared to CAT ($p<0.05$) in adults with DS. No significant changes were observed for other anthropometrical and health variables between IT and CAT. VO_2 peak and time to exhaustion ameliorated significantly in both the IT and CAT compared to control ($p<0.05$). However, relative VO_2 peak improved significantly more than with CAT ($p<0.05$). Participants in the IT group increased their VO_2 peak from 31.9 ± 8 ml/min/kg to 37.3 ± 8 ml/min/kg. Significant ameliorations in functional parameters and leg strength were shown for CAT compared to control ($p<0.05$). Participants in the CAT group improved their performance in the 6MWD (499 ± 78 m to 563 ± 75 m), 8-foot up and go (5.9 ± 1 to 4.8 ± 1 s) and leg strength (13.1 ± 2 to 15.2 ± 2 number of sit-to-stands).

The conclusion that can be drawn from this study is that all 12 functional fitness tests demonstrated excellent test-retest reliability. Both cardiorespiratory field tests indicated sound validity with the gold standard aerobic fitness test. Lastly, both IT and CAT provided significant improvements to physical, metabolic, functional and anthropometric profiles in persons with ID and DS. The influence of IT was more significant on anthropometry and aerobic capacity compared to CAT in adults with ID and DS. However, the impact of CAT was superior on functional ability and lower limb strength in adults with DS. The outcomes of the IT compared to CAT can now shed light on the volume of training (intensity, duration and frequency) that would be ideal for optimal health and functional capacity development in ID and DS populations. Moreover, these changes in the DS population can be monitored with their own unique battery of test items so that exercise or lifestyle changes can be tailored individually to specific strengths and weaknesses of the participants.

Keywords: Intellectual disability, Down syndrome, functional fitness, aerobic capacity, interval training, continuous aerobic training, anthropometry, reliability, validity

OPSOMMING

Die meerderheid individue met intellektuele gestremdheid (IG), insluitend dié met Down-sindroom (DS), is oorgewig of vetsugtig en leef 'n sedentêre lewe. Nie-aansteeklike siektes soos hipertensie, diabetes, abnormale cholesterol konsentrasies en metaboliese sindroom kom algemeen voor. Individue met IG het swak aërobiese vermoë, en persone met DS se aërobiese vermoë is nog swakker. Swak aërobiese kapasiteit hou verband met swak kardiovaskulêre fiksheid, 'n risikofaktor vir kardiovaskulêre siekte. Persone met IG en DS se funksionele fiksheidskapasiteit is ook laag. Batterye van funksionele fiksheidstoetse bestaan vir die algemene, bejaarde, IG en liggaamlik gestremde populasies, maar gestandaardiseerde toetse vir volwassenes met DS bestaan nie. Dit lei daartoe dat individue met DS saam met individue met IG groepeer word, ten spyte van onderskeibare funksionele fiksheidskapasiteite. Daarom is dit belangrik dat 'n battery toetse spesifiek vir die DS-populasie ontwikkel word.

Daar is gevind dat gestruktureerde oefenprogramme beperkte potensiaal het om die antropometriese en kardiovaskulêre veranderlikes in volwassenes met IG en DS betekenisvol te verbeter. Intervaloefening (IO) is onlangs toegepas op populasies wat die risiko loop om chroniese siektes op te doen om hulle gesondheidsuitkomstes te verbeter. Betekenisvolle fisieke, antropometriese en metaboliese voordele met intervaloefening, is in 'n verskeidenheid van populasies bewys.

Die doel van hierdie studie was om: 1) die toets-hertoetsbetroubaarheid van 'n battery toetse in 'n volwasse populasie van DS-individue te bewys, 2) die geldigheid van twee algemeen gebruikte veldtoetse (16-meter PACER & 6MWD) met die goue standaard van kardiorespiratoriese fiksheidsbepaling (direkte VO_2 maks) vas te stel. 3) Die uitwerking van gestruktureerde IO teenoor ononderbroke aërobiese oefening (OAO) op verskeie antropometriese-, gesondheids-, funksionele- en fisieke voordele in volwassenes met IG en 4) DS vas te stel.

In die eerste studie het 43 volwassenes (24 mans en 19 vroue) van 18 tot 50 jaar oud, met Down-sindroom, 'n battery toetse twee keer in 'n tweeweekperiode afgelê. Die battery toetse het uit twee balansitems, twee soepelheiditems, vyf spierkrag en uithouvermoë-items, twee aërobiese items en een funksionele taak bestaan. Die toets-hertoets-relatiewe betroubaarheid vir al die herhaalde toetse is met intraklaskorrelasiekoëffisient bepaal deur eenrigtingvariansieanalise te doen. Die toets-hertoets-absolute veranderlikheid is gemeet deur die standaardfout van meting (SFM) en betroubaarheidsdata is visueel voorgestel deur 'n Bland-Altman-stipping (grafiek). In die tweede studie het dieselfde 43 volwassenes met DS ewekansig drie aërobiese toetse op nie-opeenvolgende dae gedurende 'n eenweekperiode uitgevoer. Om geldigheid te bepaal, is pieksuurstofopname direk op 'n gemotoriseerde trapmeule gemeet. Die twee veldtoetse het die 16 meter-PACER-toets (progressiewe aërobiese kardiovaskulêre uithoutoets) en die sesminuut-stapafstandtoets (6MWD) ingesluit. Pearson-

produkmomentkorrelasies is bereken. 'n Lineêre regressie analise is uitgevoer om kriteriaverwante geldigheid tussen die veldtoetse en die VO_2-piektoets vas te stel. Die derde toets het 46 persone met IG (30 mans en 16 vroue) van Brugge in België ingesluit. Hulle is afgepaar volgens ouderdom, geslag en IK en versprei tussen IO (n=17), OAO (n=15) en 'n kontrolegroep (n=14). Die oefengroepe het vir 15 weke, twee keer per week, vir 40 minute geoefen. IO is in twee blokke van 10 minute gedoen (blok 1 en 3). Deelnemers het 10 naellope van 15 sekondes (45 sekondes van relatiewe rus) op 'n fietsergometer gedoen. Gedurende blok 2 is ononderbroke oefening gedoen. Die OAO-groep het drie blokke van 10 minute se ononderbroke oefening gedoen (fietsry, stap en trappiesklim). Na agt weke is die intensiteit van die oefening verhoog. Om voor- en naverskille tussen die groepe te evalueer, is 'n herhaalde kovariansie analise met post hoc-Bonferroni-toets gedoen. Die vierde en laaste studie het 42 volwassenes met DS ingesluit (25 mans en 17 vroue) met 'n gemiddelde ouderdom van 33.8 ± 8.6 jaar. Deelnemers is ewekansig toegeken aan een van drie groepe (IO, OAO, kontrole). Die oefenprogram was 12 weke lank (drie keer per week). Die IO-groep het tien keer vir 30 sekondes op hulle vinnigste gehardloop en vir 90 sekondes gestap of fietsgery teen lae ritme en lae intensiteit (1:3 verhouding van werk tot rus). Die OAO-groep het ononderbroke fietsgery of gestap teen 'n intensiteit van 70%–80% VO_2-piek. Om voor- en naverskille tussen die groepe te evalueer, is 'n herhaalde kovariansieanalise met post hoc-Bonferroni-toets gedoen.

Resultate het getoon dat alle funksionele fiksheidstoetse uitvoerbaar was en uitstekende toets-hertoetsbetroubaarheid getoon het (ICC's>0.9) sowel as aanvaarbare metingspresisie (SFM<SA/2). Die analises het getoon dat daar geen groot sistematiese sydigheid in die stipping was nie en die spreiding rondom die Bland-Altman was ewekansig versprei. Lineêre regressie het getoon dat die PACER (R^2=0.86) en die 6MWD (R^2=0.75) betekenisvol verband gehou het met die VO_2-piek wat direk gemeet is ($p<0.05$). Beide die 16 meter-PACER en die 6MWD het betekenisvol gekorreleer met die VO_2-piek vir volwassenes met DS. Die verband was sterker in die 16 meter-PACER-toets (r=0.87) as in die 6MWD (r=0.78). IO het 'n betekenisvolle positiewe verandering in die volgende metings getoon: middelomtrek (-4.3 cm), liggaamsvet (-3.8%), sistoliese bloeddruk (-11 mmHg), lipiedprofiel van totale cholesterol (-15 mg/dL), HDL-chol (+4.5 mg/dL) en LDL-chol (-9.4 mg/dL), homeostasemodelassessering van insulienweerstandigheid (-0.6, piek VO_2 (+0.2 L/min)), piek Watt (+23.8 W) ventilatoriese drumpel (+21 W, +0.2 L/min, 6MWD (+67.7 m) en spieruitputtingsweerstand (+6.3 s) vergeleke met geen oefening ($p<0.05$) in volwassenes met IG nie. Verder het die IO-groep betekenisvolle verbeterings in liggaamsvetpersentasie, sistoliese bloeddruk, laedigtheidlipoproteïen, vastende insulien, piek-VO_2, piekkrag en ventilatoriese drumpel getoon vergeleke met OAO ($p<0.05$). Resultate van die vierde studie het getoon dat na 12 weke van oefening, liggaamsmassa (-2 kg) en liggaamsmassa-indeks (LMI) (-0.8 kg/m^2) betekenisvol meer afgeneem het met IO as met OAO ($p<0.05$) in volwassenes met DS. Geen betekenisvolle veranderinge is in ander antropometriese en gesondheidsveranderlikes tussen IO en OAO waargeneem nie. VO_2-piek en tyd tot uitputting het

betekenisvol verbeter in beide die IO en OAO vergeleke met die kontrolegroep ($p<0.05$). Relatiewe VO_2-piek het egter betekenisvol meer verbeter met IO as met OAO ($p<0.05$). Deelnemers in die IO-groep het hulle VO_2-piek van 31.9 ± 8 ml/min/kg tot 37.3 ± 8 ml/min/kg verbeter. Betekenisvolle verbeterings in funksionele parameters en beensterkte is aangetoon vir OAO vergeleke met die kontrolegroep ($p<0.05$). Deelnemers in die OAO-groep het hulle prestasie in die volgende toetse verbeter: 6MWD (499 ± 78 m tot 563 ± 75 m), 8 voet-staan-en-stap (5.9 ± 1 tot 4.8 ± 1 s), beensterkte (13.1 ± 2 tot 15.2 ± 2 aantal sittend-tot-staande-oefeninge).

Die gevolgtrekking wat uit hierdie studie gemaak kan word, is dat al 12 funksionele fiksheidstoetse uitstekende toets-hertoetsbetroubaarheid getoon het. Beide kardiorespiratoriese veldtoetse het grondige geldigheid getoon met die goue standaard van aërobiese fiksheidstoetse. Laastens het beide IO en OAO betekenisvolle verbeterings aan die fisieke, metaboliese, funksionele en antropometriese profiele van persone met IG en DS teweeggebring. IO het ’n meer betekenisvolle invloed op die antropometrie en aërobiese kapasiteit van volwassenes met IG en DS gehad as OAO. OAO het egter ’n groter invloed op die funksionele vermoë en beensterkte van volwassenes met DS gehad. Die uitkomste van die IO vergeleke met ononderbroke oefening kan nou lig werp op die volume oefening (intensiteit, duur en frekwensie) wat ideaal sal wees vir optimale gesondheid en funksionele kapasiteitsontwikkeling in IG en DS-populasies. Verder kan hierdie veranderinge in die DS-populasie gemonitor word met hulle eie, unieke battery toetsitems sodat oefening en leefstylveranderinge aangepas kan word vir individuele sterk punte en swak punte.

Sleutelwoorde: intellektuele gestremdheid, Down-sindroom, funksionele fiksheid, aërobiese kapasiteit, intervaloefening, ononderbroke aërobiese oefening, antropometrie, herhaalbaarheid, betroubaarheid

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LIST OF ABBREVIATIONS

6MWD:	6 minute walk distance
AAIDD:	American Association on Intellectual and Development Disabilities
AIT:	aerobic interval training
ANCOVA:	one-way analysis of covariance
ANOVA:	one-way analysis of variance
aPARQ:	adapted Physical Activity Readiness Questionnaire
BIA:	bioelectrical impedance analysis
BMI:	body mass index
BOT:	Bruininks-Oseretsky Test
BOTMP:	Bruininks-Oseretsky Test of Motor Proficiency
BPFT:	Brockport Physical Fitness Test Senior Fitness Test
bpm:	beats per minute
Ca:	calcium
CAT:	continuous aerobic training
cm:	centimetre
Con:	control
CVD:	cardiovascular disease
DS:	Down syndrome
DSSA:	Down syndrome South Africa
EPOC:	excess post exercise oxygen consumption
et al:	and others

Fig:	figure
HDL:	high density lipoprotein
HGS:	hand grip strength
HR:	heart rate
ICC:	intraclass correlation coefficient
ICDWG:	World Health Organisation's Classification of diseases working group
ID:	Intellectual disability
IDD:	Intellectual developmental disorder
IQ:	intelligence quotient
IT:	interval training
kg:	kilogram
Kj:	kilojoule
L/min:	liters per minute
LDL:	low density lipoprotein
m:	metre
$MDC_{90:}$	minimal detectable change at the 90% confidence interval
ml:	milliliter
min:	minute
mmHg:	millimeter mercury
MFR:	muscle fatigue resistance
n:	amount/sample size
NDSS:	National Down syndrome Society

NWU: North-West University

p: probability

r: correlation coefficient

R^2: coefficient of determination

r/min: revolutions per minute

RER: respiratory exchange ratio

s: seconds

S&R: sit and reach

SD: standard deviation

SEM: standard error of measurement

SFT: Senior Fitness Test

SIT: sprint interval training

ss: shoulder stretch

SPSS: Statistical Package for Social Sciences

VE: minute ventilation

VCO_2: carbon dioxide production

VO_2: volume of oxygen

VT: ventilatory threshold

W: watts

WOBB: walking on balance beam

CHAPTER 1

INTRODUCTION

1.1 Introduction

Many years ago, individuals with intellectual disability (ID) were believed to be possessed with evil spirits and treated with disrespect. They were often exploited and persecuted and consequently many parents hid their children from society (Pueschel, 1989:87). Nowadays, individuals with ID are integrated into society more than ever before (Wyznikiewicz-Nawracala, 2002:75). Many companies outsource duties such as packaging, threading, woodwork, needlework, decorating and many more to intellectually disabled care centers. Those with higher competencies leave the centers during the day time and work for various employers.

However, individuals with ID still present with higher inactivity levels, obesity, metabolic and functional limitations amongst many others compared to the general population. These limitations can hamper the ability of individuals with ID to effectively integrate into modern society. Also, it can have lasting effects on their overall quality of life and independence. These limitations are not only discussed in detail in the next section of the chapter but also in second chapter of this thesis.

The next section of this chapter will provide the reader with a framework to understand the need for functional fitness testing in this often neglected population. The second chapter will provide the reader a clear overview of studies that have been conducted on persons with ID. Together, the first two chapters alert the reader of many voids or shortcomings in the studied literature and the importance of research in this field. The latter part of chapter 1 introduces the research questions, aims and hypotheses of the current study. Chapter 3 to 6 report on each of the aims individually in article format. Finally, chapter 7 provides the results of the current study in a summarised form, together with conclusions, recommendations and future studies.

1.2 Problem statement

Individuals with an intellectual disability (ID) have a poor aerobic capacity, and persons with Down Syndrome (DS) demonstrate an even poorer aerobic capacity (Mendonca et al., 2011:37; Baynard *et al*., 2008:1986; Fernhall *et al*., 2001:1657). A poor aerobic capacity is related to poor cardiovascular fitness which is a risk factor for cardiovascular disease (CVD). The presence of CVD can result in a shortened lifespan for individuals with ID (Gonzàlez-Agűero *et al*., 2010:716). Moreover, a large percentage of DS and individuals with ID are overweight or obese (Terblanche & Boer, 2013:833; de Winter *et al*., 2012:401; Salaun & Berthouze-Arande, 2012:231). A sedentary lifestyle frequently encountered in these populations further impacts the effects of an unhealthy body composition (Hilgenkamp *et al*., 2012:481;

Shields *et al*., 2009:313). Obesity and a sedentary lifestyle, is also associated with many other health-related problems commonly found in the population with ID. Non-communicable diseases such as hypertension (in elderly), diabetes, high total cholesterol, metabolic syndrome (Rimmer *et al*., 2010:787; Van de Louw *et al*., 2009:78; Carmeli *et al*., 2004:181) trouble these individuals to an even greater extent compared to the general population. Moreover, individuals with DS are born with multiple health-related abnormalities such as muscle hypotonicity, chronotropic incompetence, congenital heart disease, joint hypermobility and ligamentous laxity which further aggravate the problem toward a physical inactive lifestyle (NDSS, 2015; Abbag, 2006:219; Lewis & Fragala-Pinkham, 2005:31; Guerra *et al*., 2003:1604). It is therefore not surprising that individuals with DS and ID suffer from premature ageing (Terblanche & Boer, 2013:834).

Consequently many studies have incorporated physical activity as a possible intervention strategy to minimise or prevent declining physical fitness and adverse body composition (Calders *et al*., 2011:1097; Elmaghoub *et al*., 2011:2274; Elmaghoub et al., 2009:1327; Rimmer *et al*., 2004:165; Varela *et al*., 2001:135). Research focusing on persons with ID observed an improvement in peak VO_2 with continuous training modalities, whilst this has not been the case with persons with DS (Varela *et al*., 2001:135; Millar *et al*., 1993:270). The peak rate of relative peak oxygen consumption (VO_2 peak) has however improved significantly with the use of combined aerobic and resistance training in a DS population due to an increase in leg strength which allow them to exert themselves maximally in exhaustive exercise tests (Rimmer *et al*., 2004:165). It has also been postulated that adaptations needed for a greater relative VO_2 peak improvement in DS and ID groups, due to aerobic training, may require a longer training period and/or a higher training intensity (Gonzàlez-Agűero *et al*., 2010:723). Only one study on DS adolescents have succeeded in reporting small but significant weight or fat loss (Ordonez *et al*., 2006:416) with aerobic training. The absence of weight loss was confirmed in a meta-analysis by Dodd & Shields (2005:2051). Combined aerobic and resistance training programs seem to result in more weight loss in DS adults (Medonca *et al*., 2011:40; Rimmer *et al*., 2004:165). Mixed findings have been reported for individuals with ID without DS, with some studies indicating significant ameliorations of body composition or body mass whilst others have been less successful (Calders *et al*., 2011:1100; Elmahgoub *et al*., 2011:2276; 2009:1329). Calders *et al*. (2011:1100) did not show any improvement in body weight or body composition whereas Elmaghoub *et al*. (2011:2276, 2009:1329) showed significant improvements in both of these variables with the incorporation of combined aerobic and resistance training. In order to determine health-related fitness in persons with DS and ID, standardised test appropriate for the population is needed.

Over the years various standardised tests of physical fitness have become available for use in children and adolescents of which the most well-known is the Fitnessgram (Meredith & Welk, 2004) and the Senior Fitness Manual (Rikli & Jones, 2001) for the elderly. Through these tests healthy or unhealthy zones of physical activity can be categorised *via* norm-referenced tables. The named standardised tests

incorporate a wide range of variables associated with healthy living such as cardiovascular fitness, body composition, flexibility, balance, muscular strength and endurance. Information of this nature is important as Winnick and Short (1999:13) stated that "all individuals should possess, at minimum, levels of aerobic capacity and body composition consistent with positive health, adequate flexibility for functional health, and levels of abdominal, trunk extensor, and upper-body strength and endurance adequate for independent living and participation in physical activities". In addition, it was suggested that adequate levels of abdominal muscular strength and endurance can reduce the risk of developing low back pain and that the development of upper body muscular strength and endurance can improve the ability to perform daily tasks that require lifting, carrying, pulling or pushing objects (Winnick & Short, 1999:13). Moreover, these researchers are of the opinion that adequate levels of aerobic functioning facilitates the continued ability to sustain physical activity for work and play, and may reduce the risk of developing certain diseases and conditions such as high blood pressure, coronary heart disease, obesity, type II diabetes, dyslipidemia and certain types of cancer.

A number of standardised tests and manuals are available for testing physical and functional performance in youngsters with intellectual and physical disability. The Brockport Physical Fitness Test (Winnick & Short, 1999), FAIT Physical Fitness Test for Mildly and Moderately Mentally Retarded Students (Fait & Dunn, 1984), Ohio State SIGMA (Loovis & Ersing, 1979), Project Active level II (Vodola, 1978), AAHPERD Youth Fitness Test for mildly mentally retarded (AAHPERD, 1978), Special Fitness Test (AAHPER, 1976), and Motor Fitness Test Manual for the Moderately Mentally Retarded (Johnson & Londeree, 1976). These protocols are standardised for persons with a maximum age of 20 years. However, none of these protocols are explicitly standardised for populations with- DS and ID or DS adults. The need for a DS-specific adult functional fitness battery would be of great significance especially due to the fact that they are often ignored or pooled with individuals with ID (without DS). In fact, Winnick and Short (1999:20) acknowledged that the Brockport Physical Fitness Test (BPFT) makes no distinction between persons with and without DS despite evidence that the presence of DS negatively affects physical activity and test performance. A test battery specific to the DS population will provide an economical, easy-to-use assessment tool for measuring DS functional fitness in the clinical or community settings. In a study by Terblanche & Boer (2013:826), 13 tests were performed and selected on a trial and error basis. None of these test were standardised in a population of persons with DS. Therefore research addressing the reliability and validity of the tests in a population with DS, will contribute significantly to future research in this neglected population.

Additional to standardised testing protocols, interventions to improve the functional abilities of this neglected population are scarce with most studies focusing on traditional interventions of aerobic, resistance or combined aerobic and resistance training (Ordonez *et al*., 2012:91; Medonca & Pereira, 2009:33; Rimmer *et al*., 2004:165; Tsimaras *et al*., 2003:1239; Varela *et al*., 2001:135). Recently high intensity interval training, which involves vigorous exercise performed at a high intensity for a brief

period of time interposed with recovery intervals of low-to-moderate intensity or complete rest, has been applied to improve health outcomes of populations at risk for chronic diseases (Ciolac *et al*., 2011:824; Tjonna *et al*., 2008:346; Trapp *et al*., 2008:684). In this regard the significant physical, anthropometrical and metabolic benefits associated with interval training (IT) have been demonstrated in a variety of populations. Some of these populations groups even included individuals with chronic medical conditions such as coronary artery disease or those who suffered a stroke (Globas *et al*., 2012:85; Hesse *et al*., 2011:838; Helgerud *et al*., 2007:665; Ingul *et al*., 2010:852; Babraj *et al*., 2009:1; Gibala & McGee, 2008:58; Praet *et al*., 2008:163). Many studies show that physiological, anthropometrical and functional benefits of IT are superior to traditional continuous aerobic training (Ciolac *et al*., 2011:824; Smart *et al*., 2011:205; Moholdt *et al*., 2009:1031; Tjonna *et al*., 2008:346; Wisløff *et al*., 2007:3086; Nemoto *et al*., 2007:803; Rognmo *et al*., 2004:216) and was confirmed by a meta-analysis (Hwang *et al*., 2011:378). Moreover, IT has been shown to be more fun and less time consuming (Bartlett *et al*., 2012:547). The benefits associated with IT are discussed in detail in Chapter 2.

Despite the enormous wealth of research that provide evidence for the superior effect of IT in the general population on various health, fitness and body composition indices, IT has not been applied in a population of individuals with ID and DS whom is known to age earlier and present with physical and physiological abnormalities. Various traditional exercise training interventions in persons with ID and DS have not been effective to significantly improve aerobic capacity and anthropometric associated variables (Cowley *et al*., 2011:2233; Varela *et al*., 2001:135; Lewis & Fragal-Pinkham, 2005:30; Millar *et al*., 1993:270).

Therefore the research question that arises is: What is the reliability and validity of selected functional tests and the effect of aerobic continuous and IT exercise on the physical, metabolic and anthropometric profiles of persons with ID and DS? The results from this investigation will guide researchers and exercise physiologists to apply reliable and validated testing protocols when testing persons with ID and DS specifically. The outcomes of the IT compared to continuous training will shed light on the volume of training (intensity, duration and frequency) that would be ideal for optimal health and functional capacity development in a DS population. The findings can be used to inform policy makers on the contribution of regular exercise in special populations in particular persons with DS and ID.

1.3 Objectives of the study

The specific objectives of this study are to determine the:

- Test-retest reliability of 12 physical tests in an adult population with DS
- Validity of the 16-metre PACER and six-minute walk test as cardiorespiratory fitness tests in adults with Down syndrome

- Effects of a continuous aerobic exercise and IT programme on the physical, metabolic and anthropometric profiles of persons with ID.
- Effects of a continuous aerobic exercise and IT programme on the physical, metabolic, functional and anthropometric profiles of persons with DS.

1.4 Hypotheses

The study is based on the following hypotheses.

- A good to moderate test-retest reliability will be obtained in the majority of the 12 fitness tests for persons with DS.
- Both field tests will present adequate validity but the 16-metre PACER test will provide a more valid indication of cardiorespiratory fitness.
- That IT intervention will indicate a significantly larger improvement on the physical, metabolic and anthropometric profiles compared to the continuous training intervention of persons with ID.
- That IT intervention will indicate a significantly larger improvement on the physical, metabolic, functional and anthropometric profiles compared to the continuous training intervention of persons with DS

1.5 Structure of the thesis

This thesis is written according to the article format of the North-West University (NWU) as approved by the senate (Potchefstroom campus). The four articles presented herein have been submitted for peer reviewed publication. Chapter 1, 2 and 7 are presented and referenced according the NWU guidelines for the submission of a PhD thesis. Chapters 3 to 6 are presented according to the outline and instructions of each respective journal. The guidelines and instruction to authors are attached as appendices.

The thesis is presented in five main parts, namely an introduction (Chapter 1), a narrative summary (Chapter 2), four research papers (Chapter 3 to 6), and a summary with conclusions, limitations, recommendations and future studies (Chapter 7). The first chapter addresses the introduction, problem statement, objectives and hypotheses of the study. The literature overview chapter considers functional fitness testing in adults with ID and DS. An overview of the use of interval training in the general population is also provided. The first research article (Chapter 3) investigates the test-retest reliability of various functional fitness tests in adults with DS. The research article 3 (Chapter 4) aims to determine whether two field tests are valid indicators of cardiorespiratory fitness in adults with DS. The third and fourth research articles (Chapter 5 and 6) investigate the effects of interval training on body composition,

physical and metabolic fitness in adults with ID and DS respectively. References are listed separately for each chapter. Chapter 7 is followed by a list of appendices.

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CHAPTER 2

LITERATURE REVIEW: FUNCTIONAL ABILITY AND EXERCISE IN PERSONS WITH INTELLECTUAL DISABILITY AND DOWN SYNDROME

2.1 Introduction

Persons with an intellectual disability (ID) have for decades been treated as persons of lesser importance by communities, resulting in a negligence of health and wellness care. By ignoring the health related quality of life needs of persons with ID, functionality and fitness were understudied impacting on physical and metabolic systems in this population. ID impacts the individuals themselves, their families and society as a whole. For instance, more fathers and mothers of children with ID suffer from depression than those who do not have children with ID (Olsson & Hwang, 2001:535). ID is not curable but there are treatment objectives that focus on the normalisation of behaviour and emotional wellbeing (Katz & Lazcano-Ponce, 2008:138). Other psychiatric and somatic disorders are often associated with ID and add to the intricacies of managing this complex and multidimensional disability (Emerson & Hatton, 2007:493). Regrettably, it is often stigmatised and those with ID are discriminated against. Society as a whole should act against stigmatisation and actively promote inclusion of individuals with ID into the work and social sectors. When a diagnosis of ID is made, parents mostly take the news with great difficulty. It is understandable that the acceptance of such information is not easy as it places one on a lifelong journey with extra caretaking, health complications and subsequent financial implications. The joys and love that most parents experience after a period of adaptation with such children are inexplicable.

Against this background, limited studies focussing on the functionality, health and influence of exercise on individuals with ID and Down syndrome (DS) have been reported. This chapter aims to provide the reader with background knowledge regarding research that has addressed these aspects. In this chapter, individuals with ID and those with DS will be discussed separately due to their marked differences in functionality, health and physical parameters such as cardiorespiratory fitness (discussed in Chapter 1). Furthermore, this chapter provides the reader background information for each population group regarding definitions, characteristics, causal factors, prevalence, life expectancy, exercise and standardised functional fitness tests. Lastly, this chapter introduces the concept of interval training and the associated effect on health and physical parameters in the general population.

2.2.1 Definition of intellectual disability

Salvador-Carulla and colleagues (2011:175) presented information pertaining to the name, definition and framework for intellectual disability from the World Health Organisation's International

Classification of Diseases Working Group (ICDWG). Through a combined mixed and qualitative approach (prior knowledge and available evidence), extensive literature reviews and subsequent meetings between the ICDWG, consensus was reached on how to name, define and describe this complex and multidimensional construct. After 15 years of widespread debates between 30 experts from low and high income countries, the name given to this construct is Intellectual Developmental Disorder (IDD). IDD is defined as "a group of developmental conditions characterised by significant impairments of cognitive functions, which are associated with learning, adaptive behaviour and skills" (Salvador-Carulla *et al.*, 2011:177). Main descriptors of IDD are provided in Table 2.1. The term "intellectual" was chosen not only due to its global use and recognition but also for the fact that it is an umbrella term that covers concepts such as adaptive behaviour and cognitive functioning. The word "developmental" was ideal as it denotes a "time frame" during which the brain and nervous system are evolving. Lastly, the term "disorder" was relevant as it showcases a specific set of symptoms or behaviour.

Table 2.1: Main descriptors of Intellectual developmental disorder

1	IDD is characterized by a marked impairment of core cognitive functions necessary for the development of knowledge, reasoning, and symbolic representation of the level expected of one's age peers, cultural and community environment.
2	Persons with IDD have difficulties with verbal comprehension, perceptual reasoning, working memory and processing speed.
3	The cognitive impairment in persons with IDD is associated with difficulties in different domains of learning, including academic and practical knowledge.
4	Persons with IDD typically manifest difficulties in adaptive behaviour; that is, meeting the demands of daily life expected of one's age peers, cultural, and community environment. These difficulties include limitations in relevant conceptual, social, and practical skills.
5	Persons with IDD often have difficulties in managing their behaviour, emotions, and interpersonal relationships, and maintaining motivation in the learning process.
6	IDD is a lifespan condition requiring consideration of developmental stages and life transitions.

Reprinted with permission from Salvador-Carulla *et al*., (2011:177)

By comparison, the American Association on Intellectual and Development Disabilities (AAIDD) defined Intellectual Disability (ID) as "limitations both in intellectual functioning and behaviour" (AAIDD, 2013). Matthews and colleagues (2011:361) operationally defined the term as having an intelligence quotient of less than 70. Cases are further classified as mild (IQ 50–55 to 69), moderate (IQ 35–40 to 40–54) and severe (IQ < 40) (Diagnostic and Statistical Manual of Mental Disorders, 1994:25). Throughout this thesis, the term ID will be used as it is the most recognised and widely used term even though it has the same implicit meaning as IDD and mental retardation.

2.2.2 Causal factors for intellectual disability

Causal factors of ID can be described as genetic, acquired, and environmental (Katz & Lazcano-Ponce, 2008:134). The three causal factors and their subcategories are summarised in Table 2.2.

Table 2.2: Causal factors related to Intellectual Disability

Genetic	**Chromosomal**
	Down syndrome (trisomy 21 or translocation of chromosome 15 & 21)
	Prader-Willi's syndrome, Rett syndrome, tuberous sclerosis, Lesch-Nyhan syndrome, Fragile X syndrome, neurofibromatosis,
	Hereditary
	Tay-Sachs disease, galactosaemia, glycogen storage disease
Acquired	**Congenital**
	Metabolic (neonatal hypothyroidism)
	Toxic (foetal alcohol syndrome, lead poisoning, prenatal exposure to substances)
	Infectious (rubella, syphilis, toxoplasmosis, herpes)
Environmental	Poor prenatal (toxaemia, diabetes) perinatal (asphyxia) and postnatal (encephalitis, meningitis) healthcare
	Adolescent maternity
	Family instability
	Insufficient health professionals
	Low level of stimulation and education

In South Africa, Kromberg *et al*. (2008:89) reported that 21% of the cases of ID were due to a congenital reason, 6% was acquired and 73% was undetermined. The lack of detailed data on the cases for ID prevented further subcategorising. In order to treat and prevent manageable cases of ID, the cause of the condition should be known. Maulik and colleagues (2011:423) also reported in a global meta-analysis of ID prevalence that at least half of the causal factors were unknown. They showed that antenatal, perinatal, and postnatal causes corresponded in number for the remaining half of cases known. Specifically, they showed that genetic causes such as Down's syndrome were a typical antenatal cause. Intra-uterine growth retardation and birth injury were common perinatal causes with infections being associated with postnatal causes (Maulik *et al*., 2011:423).

2.2.3 Prevalence of intellectual disabilities

The prevalence of ID has been estimated to be 10.4 per 1000 individuals. This statistic was attained from a global study that incorporated a meta-analysis of 52 studies spanning from 1980 to 2009 (Maulik *et al*., 2011:419). This meta-analysis included 23 countries from North America, Africa, Asia, Australia and Europe. Discrepancies between studies were explained by the socio-economic status of the country, age group of the study population, and simply study design. The highest percentage of ID individuals documented originated from low to middle income countries (16.4/1000). Prevalence was also higher in men than women and amongst children and adolescents compared to adults (Maulik *et al*., 2011:423). With regards to population type, the highest incidence occurred in urban slums (21.2/1000), followed by rural settings (Maulik *et al*., 2011:423). Prevalence rates for ID in South Africa was reported as 3.6% (Kromberg et al., 2008:89) while a review by Adnams (2010:436) suggests that the prevalence rate are higher in SA than in developed countries. A census conducted in 2001 revealed that 5% of the general population had a disability whereas 0.5% of the population was ID (Statistics South Africa, 2007). However, institutionalised individuals were not included in the census count which excluded a large percentage of the ID population (Adnams, 2010:436). A more recent census conducted in 2011 did not provide information on specific persons with ID due to inconsistencies in the questionnaire used and changes in the terminology and classification of disability. Questions regarding body impairment and activity limitations are provided but none referring directly to ID (Statistics South Africa, 2007). It is unfortunate that low-income countries have fewer resources and qualified personnel to manage persons with ID effectively and to educate the public concerning preventative measures (e.g. no overconsumption of alcohol during pregnancy). Also, lower-income countries are often limited in the resources to deal with some of the clinical causal factors such as brain injury during birth, postnatal infections and perinatal growth retardation.

Basic living conditions such as poverty and insufficient nutrient intake during pregnancy and postnatal periods also play a large causal role and have been shown to be associated with ID, echoing the belief that the prevalence of ID is more prominent in developing countries and poorer areas (Emerson, 2007:107). Considering public health perspectives, it is clinically significant to address manageable issues such as brain injury during childbirth, postnatal infections, and identification of congenital hypothyroidism, immunisation, iodine supplementation, and maternal education amongst others. It is also not surprising why Adnams (2010:436) argued the importance of reliable census data acquisition so that effective intervention strategies can be implemented seeing that most of the causes associated with ID are preventable. The researcher also argued that even though there are policies in place for individuals with ID in South Africa, recognition of and provision for their needs enjoy low priority. There is also insufficient demographic data (prevalence information across different geographical, race, age and gender groups) to pinpoint and plan subsequent supporting services. Adnams (2010:436) specifically argued that there was an extraordinary burden on preventable causes for ID.

2.2.4 Life expectancy of persons with intellectual disabilities

Life expectancy has increased substantially in the ID population in the last two decades (Coppus, 2013:6; Bittles *et al*., 2002:470). It has also been shown that individuals with mild ID live longer compared to those with severe ID (Hutton & Pharoah, 2002:86; Bittles *et al*., 2002:470). However, mortality rates are higher and chronic health conditions are more common in persons with ID, and the average life expectancy is shorter than in the general population (Oeseburg *et al*., 2011:59; Tyrer & McGrother, 2009:900; Janicki *et al*., 1999:284). The average life expectancy for individuals with ID and DS living in America was reported to be 66.1 years and 55.8 years respectively (Janicki *et al*., 1999:284). The most common causes of death are cardiovascular diseases, congenital abnormalities, mental disorders, diseases of the nervous system, respiratory diseases, cancer and neoplasms (Tyrer *et al*., 2009:898; Patja *et al*., 2001:30; Janicki *et al*., 2002:287). However, there are some discrepancies in the cause of death as some records showed that ID was the cause of death (Tyrer *et al*., 2009:902). The leading consequence before death should be recorded, rather than a predisposition that may have an enhanced influence on the cause of death. Another study showed that the causes of death in a population of persons with ID are similar to that of the general elderly population (Janicki *et al*., 1999:284). Thomas and Barnes (2010:201) performed a review that presented the key elements contributing to a reduced life expectancy in individuals with ID. These were listed as swallowing problems, immobility, epilepsy, incontinence and cognitive and emotional impairment. They specifically explained that immobility results in an improved probability to develop osteoporosis. Cardiovascular diseases were less prevalent in persons with ID compared to the general population (Merrick *et al*., 2004:413). However, more recent evidence report that cardiovascular disease is as prevalent and is a common form of death when compared to the general population (Haveman *et al*., 2010:59; Patja *et al*., 2001:32). Variations in prevalence rates were found to be culturally dependent. Regarding support (community centre or institution) or private and family accommodation there is no convincing evidence that a difference in life expectancy exists (Thomas & Barnes, 2010:206)

2.2.5 Health of individuals with intellectual disability

2.2.5.1 Chronic diseases of lifestyle

De Winter *et al*. (2009:427) identified serious chronic conditions in this population in the Netherlands of which diabetes (8.7%), hypertension (36.8%), abdominal weight (70.4%) and hypercholesterolaemia (31.8%) were prevalent. A study by de Winter *et al*. (2011:141) showed that 25% of ID individuals suffer from metabolic syndrome and Einfeld *et al*. (2011:141) reported that 30% to 50% of individuals with ID (from developed and developing countries) suffer from a mental disorder. Jeevanandam (2009:462) reported that a large percentage of Singaporean ID adults and children suffer from obesity, high blood pressure and high total cholesterol. In a more recent study by de Winter and colleagues

(2012a:1726) in a near representative Dutch population it was shown that a prevalence of 53% for hypertension, 13.7% for diabetes, 23.1% for hypercholesterolaemia and 44.7% for metabolic syndrome occurred. These are all risk factors for cardiovascular disease. Likewise the incidence of these chronic conditions was interpreted as similar to the general Dutch population (de Winter *et al*., 2012a:1726). Only hypercholesterolaemia was less prevalent. On the contrary, another study that was conducted more than a decade ago showed that the risk factors for cardiovascular disease (hypertension, hyperlipidaemia and adult onset diabetes) demonstrated lower rates in persons with ID compared to the general population (Janicki *et al*., 2002:287) more than a decade ago. Adults with ID that had a greater tendency for cardiovascular risk factors were women, the elderly, people with obesity and those who lived more independently (de Winter *et al*., 2012a:1726). Cardiovascular diseases and conditions have also been found to increase with advancing age and are more prominent in those with higher functional abilities and body mass index (BMI) (Janicki *et al*., 2002:292). These researchers also reported that cancer was more common in women and that the most noticeable musculoskeletal conditions were osteoporosis and osteoarthritis. The same study reported that dementia manifested in 4% of the population (aged over 70) and that psychological conditions such as depression, anxiety and behavioural problems occurred in 15% to 20% of adults with ID. Many other health conditions such as cerebral palsy (19.8/100), epilepsy (22.0/100), autistic disorder (10.1/100), anxiety disorders (17.1/100) and oppositional defiant disorder (12.4/100) are seen in children with ID (Oeseburg *et al*., 2011:59).

Rimmer *et al*. (2010:787) and Draheim *et al*. (2002b:441) stated that obese individuals with ID were more prone to secondary conditions such as diabetes, hypertension, cholesterol and depression compared to healthy ID individuals. Additionally, Janicki *et al*. (2002:292–293) found that obese adults with ID were more likely to be diagnosed with cardiac, neurological, gastrointestinal and mental health disorders. Unfortunately, health clinics and practices do not perform routine health checks on their patients with ID (Chauhan *et al*., 2010:483). These researchers demonstrated that only 9 (92 patients) out of 27 practices (651 patients) performed health checks on individuals with ID in the United Kingdom. This important remark was reiterated by de Winter *et al*. (2012a:1722) who stipulated that a policy on prevention, recognition and treatment of cardiovascular risk factors is urgently needed. If this is the case in more developed countries such as the Netherlands, one can only assume it to be even more imperative for developing countries. Also, preventative and treatment options and strategies for individuals with ID should be similar to those provided to the general population.

Hypertension occurred in less than 10% of individuals with ID in South Africa (Moss, 2009:738). Even though a low prevalence of hypertension was reported, in many cases where high blood pressure was identified, participants had not previously been diagnosed with the condition (de Winter *et al*., 2012a:1722). This is also the case for metabolic syndrome where 94% of cases had not previously been diagnosed (de Winter *et al*., 2012a:1729). These prevalence rates indicate that a large percentage of individuals with ID are not sure that they have these symptoms or the dangers thereof.

Even though the life expectancy of individuals with ID has increased it has resulted in increased age-associated chronic conditions and functional incapacity (Terblanche & Boer, 2013:835; Hilgenkamp *et al*., 2012:477; Lahtinen *et al*., 2007:132,140). An increase in obesity as these individuals age (Lahtinen *et al*., 2007:133) coupled with a sedentary lifestyle, which is frequently encountered in an ID population, adds to the onset of age-associated diseases and functional incapacity (Hilgenkamp *et al*., 2012:477; Finlayson *et al*., 2011:508; Lotan *et al*., 2006:219). Janicki *et al*. (1999:284) proposed that the implementation of additional medical health centres could have a significant effect to delay or curtail the incidence of life-threatening conditions in adults with ID. These centres could provide norms and standards for health-related ageing. Any deviations from the norm could alert health officials to take appropriate intervention steps. If intervention strategies are implemented at an early stage, age-related diseases and conditions could be prevented. De Winter and colleagues (2012a:1730) also highlighted that future studies should focus on what contributes to chronic conditions and the impact thereof on future morbidity and mortality.

2.2.5.2 Overweight and obesity

A large proportion of individuals with ID are overweight or obese (de Winter *et al*., 2012b:398; Salaun & Berthouze-Aranda 2012:231, 236; Stewart *et al*., 2009:882; Moss, 2009:735; Melville *et al*., 2008:425; Lahtinen *et al*., 2007:131; McGuire *et al*., 2007:500) as measured by BMI, bioelectrical impedance analysis or waist circumference indices. Melville *et al*., (2008:428) specifically reported in a sample of 945 adults with ID that approximately 70% of women and 60% of men are overweight or obese. What is more, those individuals with a higher BMI (>27 kg/m^2) compared to a more optimal BMI (<27 kg/m^2) have more cardiovascular associated diseases and conditions (Janicki *et al*., 2002:292). This increased incidence of obesity may even develop in childhood and adolescence (Salaun & Berthouze-Aranda 2012:231; Stewart *et al*., 2009:882). Salaun and Berthouze-Aranda (2011:333) showed that up to a third of adolescents with ID have excess visceral adipose tissue and close to 47% have excess body fat. This has a snowballing effect as it has been shown that BMI increases as individuals with ID age with up to 70% of individuals in adulthood being overweight (Lahtinen *et al*., 2007:131).

De Winter *et al*. (2012b:398), Melville *et al*. (2008:425) and McGuire *et al*. (2007:500) established that a larger percentage of individuals with ID are obese or overweight compared to the general population. Women are more likely to be obese compared to men and the risk of obesity decreases as the severity of ID increase (Melville *et al*., 2008:425). Mikulovic *et al*. (2010:157) specifically showed that obese children with ID (n=410) engage in less physical activity compared to their non-obese peers. One of the primary reasons for the large incidence of obesity may stem from an inactive lifestyle which is discussed in more detail in the next section. A study by Salaun and Berthouze-Aranda (2012:231) reported that ID individuals with the highest body fat percentages had the lowest physical and functional fitness.

Additionally, obese individuals with ID are more prone to develop secondary functional fitness limitations compared to healthy ID individuals (Rimmer *et al*., 2010:787; Draheim *et al*., 2002b:441).

2.2.5.3 Lifestyle

Lifestyle factors considered to influence long-term health include diet, physical activity, alcohol consumption and smoking. Most individuals with ID live sedentary lifestyles and have poor eating habits (Dixon-Ibarra *et al*., 2013:1; Hilgenkamp *et al*., 2012:477; Finlayson *et al*., 2011:508; Moss, 2009:735; Emerson, 2005:134; McGuire *et al*., 2007:497; Frey, 2004:235; Draheim *et al*., 2002b:436). In a South African population of persons with ID, Moss (2009:735) reported that 85% of the adults with ID living in a care facility were sedentary. International investigations report similar findings. Finlayson *et al*. (2011:508) explained that only 27% of ID adults reach the recommended 10 000 steps per day and that only 15% of individuals meet the required five days a week for 30 minutes moderate physical activity per day. Regarding older individuals with ID, only 6% meet the required 150 minutes of moderate physical activity and only 4% met the required 10 000 steps (Dixon-Ibarra *et al*., 2013:9). Moreover, younger adults with ID had a similar physical activity status compared to older adults in the general population despite a mean age difference of 40 years (Dixon-Ibarra *et al*., 2013:13). Physical inactivity is also seen more prominently in children with ID compared to their non-disabled peers (Hinckson & Curtis, 2013:72). If inactivity stems from childhood it may be more difficult for these individuals to adjust to a habitually active lifestyle. Physical inactivity in this population is a concerning issue that needs to be addressed to avoid the negative health and functional consequences thereof (Lotan *et al*., 2006:219).

An inactive lifestyle may also enhance the risk of falls (Cox *et al*., 2010:1045). Reports indicate that 34% (39/114) of patients with ID (18 years and older) fell during the past 12 months, of which 84% sustained injuries. Even though they did not measure physical activity, results showed that a seizure (in the past five years), fractures and increasing age were significant predictors for future falls.

Some of the barriers that are associated with a physically active lifestyle, as pointed out by Mahy *et al*. (2010:795), are a lack of support from those closest to the individuals, physiological or medical reasons and the fact that individuals with ID just do not want to engage in physical activity. Barr and Shields (2011:1020) identified an additional two barriers to the barriers reported by Mahy *et al*. (2010:795), namely inaccessibility of training programs and competing family responsibilities. It has been shown that the attitudes of physiotherapy students transformed positively from baseline to post-intervention as to whether individuals with ID can successfully complete a physical activity regime or not (Shields *et al*., 2011:360).

Very few studies have actively engaged in research regarding the eating habits of individuals with ID. Robertson *et al*. (2000:476) reported that only 8% of adults with ID met the criteria for a balanced diet. Another study showed that individuals with ID are comparable to the general population regarding sugar and fat intake, but the daily intake of carbohydrates, proteins, dairy, fruits and vegetables is less than ideal (McGuire *et al*. 2007:501). This is important as a study by Draheim *et al*. (2002a:361) proved that more physical activity bouts and less dietary fat intake significantly reduced the odds of hyperinsulinaemia and abdominal obesity. A positive correlation was reported between BMI and the frequency of eating fast foods (George *et al*., 2011:1054) and true for both parents of persons with ID and individuals with ID. Concerning the influence of communal living versus family homes, it was reported that women consumed more fruit and vegetables in communal settings compared to those living with families (Draheim *et al*., 2007:392). Also, a larger proportion of people in the communities consumed precooked meals (Nordstrøm *et al*., 2015). At the same time only 6% of all individuals, irrespective of where they resided, consumed the recommended five portions of fruit and vegetables per day and only 15%–30% consumed the suggested calories from fat intake. De Winter *et al*. (2012a:1729) reported that in their large study (n=980) cardiovascular risk factors might also be influenced by diet which their study did not take into account. It was reiterated by Ewing *et al*. (2004:84) and Heller *et al*. (2011:26) that nutrition programs should go hand-in-hand with intervention strategies if the plan is to reduce obesity. More research is needed to describe the eating habits of individuals with ID in South Africa.

The consumption of alcohol and smoking is consistently reported as very low. Fortunately, only 2.6% and 10.3% of adults with ID are reported to smoke tobacco and consume alcohol regularly (McGuire *et al*., 2007:497). Janicki *et al*. (2002:287) found similar low occurrences. Similar low prevalence was also reported in a South African co-hort (Moss, 2009:738). It is possible that many individuals with ID do not have access or money to buy cigarettes or alcohol.

2.2.6 Physical activity and exercise

Physical activity, improved nutrition and a healthy lifestyle are seen as non-pharmaceutical agents to combat the many chronic health-related problems and risks that individuals with ID are confronted with (Haveman *et al*., 2010:59). Moss (2009:735) proved that two of the most common risks for cardiovascular disease can be combatted successfully with a physically active lifestyle. The initiation of a walking program for adults with ID significantly reduced the levels on inactivity and the percentage of body fat. Even though adherence to the physical activity intervention was low (47%), personal observation revealed that participants continued the walking after the intervention period lapsed. The poor attendance was explained as exercise being a new concept for many of these individuals and their motivation was low.

Physical fitness is a key modifiable factor than can significantly alter the quality of life especially during ageing. It is defined as "having the physical capacity to perform normal everyday activities safely and independently without undue fatigue" (Rikli & Jones, 2013:2). The factors to that are found to be important are muscular strength, aerobic endurance, flexibility, agility, dynamic balance and BMI (Rikli & Jones, 2013:8; Salaun & Berthouze-Aranda, 2012:231). It is known that physical fitness reduces the risk of cardiovascular disease, low bone density, low muscle mass and functional incapacity (Mazzeo &Tanka, 2001:809). Unfortunately, the physical fitness of ID is poor when compared to the general population (Hilgenkamp *et al*., 2012:477; Golubovič *et al*., 2012:610; Chow *et al*., 2005:13). Specifically, Hilgenkamp *et al*. (2012:477) reported that the youngest adults with ID performed in a manner similar to adults in the general population 20–30 years their seniors. It has also been shown that those individuals with a more severe ID performed worse in various physical fitness tasks than those with borderline ID (Golubovič *et al*., 2012:608). Fortuitously, a physical exercise intervention has been shown to improve cardiovascular fitness, functional abilities or daily living activities, balance, flexibility, muscular strength, reaction time, well-being, mental health, body mass, fat mass, lung function, resting systolic blood pressure and cholesterol (total, HDL and LDL) in adolescents and adults with ID (Enkelaar *et al*., 2012:291; Jankowicz-Szymanska *et al*., 2012:675; Yen *et al*., 2012:85; Calders *et al*., 2011:1097; Elmahgoub *et al*., 2011:2274; Hutzler & Korsensky, 2010:767; Yildirim *et al*., 2010:178; Elmahgoub *et al*., 2009:1327; Khalili & Elkins, 2009:171; Moss, 2009:735; Carmeli *et al*., 2008:457; Carmeli *et al*., 2005:299; Chanias *et al*., 1998:119). It should be practice and policy that all individuals with ID engage in a physically active lifestyle to prevent and/or reverse the premature loss of functions and the increased dependence on caregivers (Hilgenkamp *et al*., 2012:477).

2.2.6.1 Facilitators to a habitually active lifestyle

Methods to enhance physical activity and to overcome barriers associated with physical activity have been successful with the use of audio and visual feedback, peer modelling, social support, social interactions and low-cost activity programs in an ID population (Barr *et al*., 2011:1020; Lante *et al*., 2011:197; Hutzler *et al*., 2010:767; Mahy *et al*., 2010:795; Frey *et al*., 2005:241). A lack of education, mindfulness, self-discipline and inspiration to the benefits of exercise contributes to a physically inactive lifestyle (Carmeli *et al*., 2008:463). Fortunately, a long-term, sustainable and integrated community-based program revealed that over time adults with ID spent less time in sedentary behaviour or light activity and more in moderate to vigorous zones (Lante *et al*., 2011:197). They showed that psychosocial benefits such as meeting new people and attaining social respect spurred the individuals to a habitually active lifestyle. This process was gradual but ameliorated common misconceptions that exercise is too boring and difficult (Lante *et al*., 2011:197).

2.2.6.2 Effect of physical activity on aerobic capacity

General physical fitness of individuals with ID are lower than the general population's but more specifically, the aerobic capacity was also shown to be lower across all age categories (Baynard *et al*., 2008:1984). Many years ago it was shown that various physiological variables and VO_2 peak could improve with a well-designed training program (Pommering *et al*., 1994:218). Four years later, a meta-analytic study by Chanias *et al*. (1998:119) also demonstrated significant improvements in cardiovascular endurance after a period of aerobic training. However, a more recent study showed no improvements in aerobic capacity after five months of endurance training (Calders *et al*., 2011:1097). Likewise, the aerobic capacity of adolescents with ID was not improved even when combined aerobic and resistance training were implemented (Elmahgoub *et al*., 2011:2277; Elmahgoub *et al*., 2009:1329). On the contrary, the combined aerobic and strength training group in the study by Calders *et al*. (2011:1101) reported significant improvements in aerobic capacity. It is unclear why there are discrepancies between these studies but it is clear that an in-depth analysis of the ideal training frequency, duration, intensity and type of exercise should be undertaken for optimal results concerning aerobic capacity in persons with ID.

2.2.6.3 Effect of physical activity on body composition

The high prevalence of overweight and obesity has been the focus of the majority of intervention studies in persons with ID. A 20-week intervention study that included three groups with an aerobic, combined aerobic and resistance training and a control group found no significant effects on body mass, fat mass, BMI or waist circumference in adults with ID (Calders *et al*., 2011:1100). Similar results were again obtained by Elmahgoub *et al*. (2011:2276) with a combined aerobic and resistance training program in a 10- and 15-week intervention period respectively. However, the opposite effect was found in another study where body mass, BMI, waist circumference and fat mass all decreased significantly with a combined aerobic and resistance training program over 10 weeks (Elmahgoub *et al*., 2009:1329). In fact, the mean fat mass decreased from 37 kg to 32.9 kg. Differences in results could be attributed to training intensity. However, both the Elmahgoub *et al*. (2009:1329; 2011:2277) studies were performed at a similar training intensity (60–75% of heart rate reserve). Another possible confounding factor could be that two of the studies were conducted on adolescents and the other on adults. Still, both the Elmahgoub *et al*. (2009:1327; 2011:2274) studies were conducted on adolescents. The baseline BMI reported in the studies was similar, therefore the outcome reported by the studies should have been similar.

In another study conducted on 100 men and women with ID (21 to 72 years), a significant reduction in body fat percentage was recorded after 12 weeks of a 3-times weekly walking program (Moss 2009:740). No changes were recorded for body mass and BMI. On the contrary, significant improvements in body

mass and BMI in adults with ID were reported after a six-month aerobic intervention strategy in Taiwan (Wu *et al*., 2010:713). Another aerobic intervention study just in excess of six months not only showed an improvement in BMI but also in the loss of fat mass (Merrick *et al*., 2013:45). A meta-analysis conducted for two decades on individuals with ID reported that no significant results were reported for an improvement in body composition (Chanias *et al*., 1998:119). A more recent review by Hamilton *et al*. (2007:340) reported that some studies reported no loss in body mass, whilst others reported small but significant loss in body mass. None of these studies monitored nutrient intake which could possibly explain differences in the results obtained. Future studies should explore the effect of monitored nutrient intake in association with an exercise intervention period. Hamilton *et al*. (2007:340) explained that there was a need to develop more evidence-based weight management programs for adults with ID.

It is clear from the presented research that there are disparities between findings regarding the influence of exercise on body mass, BMI and body composition. Some of these differences can be explained by the study population tested, intervention strategies, intervention duration, and means of fat mass determination or other methodological and procedural differences. It is worrisome that only a few studies have shown significant improvements in either or most of the anthropometric variables tested. Rimmer and Yamaki (2006:22) pointed out that there is a lack of research on body mass reduction strategies and that this matter should be explored in more detail. Yet again, it is recommended that research should focus on the precise frequency, duration, intensity and type of exercise for optimal and ideal body composition ameliorations.

Whether exercise plays an important role in the significant amelioration of anthropometric variables remains unclear but what we know is that those individuals with the lowest incidence of obesity have the highest physical fitness (Salaun & Berthouze-Aranda, 2012:231). Whether this relationship is causal remains to be proven.

2.2.6.4 Effect of physical activity on functional ability

Optimal functional ability is needed to perform activities of daily living. A review study by Andriolo *et al*. (2010:2) proved that aerobic activities performed in a population of individuals with ID significantly improved performance in the timed up-and-go test. This test requires an individual to get up from a chair, walk a certain distance and return to a seated position. It reflects an everyday life activity such as getting into and out of a car. Improvements in other functional tests such as the sit-to-stand test (number of stands in 30 seconds) and the six-minute walk distance test were also significant in studies that included a three-weekly aerobic or combined aerobic and resistance training program (Calders *et al*., 2011:1101; Elmahgoub *et al*., 2011:2277; Elmahgoub *et al*., 2009:1329). Improvements in physical activity and aerobic fitness is important in ID individuals as it predicts functional ability (Guerra-Balic *et al*., 2015).

2.2.6.5 Effect of physical activity on muscular strength

A combined aerobic and resistance training program improved abdominal strength, hand grip strength, leg strength, 1-repetition upper limb and 1-repetition lower limb strength in adolescents and adults with ID (Golubovič *et al*., 2012:608; Calders *et al*., 2011:1101; Elmahgoub *et al*., 2011:2277; Elmahgoub *et al*., 2009:1329; Wu *et al*., 2010:713). A meta-analysis conducted on individuals with ID reported that moderate effect sizes were established for an improvement in muscular strength (Chanias *et al*., 1998:119). Particularly, they proved that a resistance training program had larger effect sizes on muscular strength compared to combined training programs. Regarding aerobic training only, a 32-week walking program conducted on adults with ID reported no improvement in maximal isometric quadriceps muscle strength (Merrick *et al*., 2013:45). On the other hand, a 6-month treadmill walking program conducted on elderly individuals with ID found significant improvements in isokinetic knee flexion and extension (Carmeli *et al*., 2002[c]:106).

Current data reported indicate that continuous aerobic exercise of between 20 – 30 minutes for at least 12-weeks, as presented by the general physical activity guidelines, in individuals with ID has a limited impact on variables such as anthropometry, and physical fitness especially cardiorespiratory endurance. Perhaps the training type, intensity or volume has been insufficient to result in physiological improvements in variables associated with body composition and aerobic capacity. Improvements in health, functional and physical parameters are important since low levels of functional fitness and poor health status are reported in adults with ID. Alternative exercise interventions should be considered in this special population to ensure weight loss, address lack of motivation and sustainable physical activity over a long-term.

2.3.1 Down syndrome as a form of intellectual disability

Intellectual disability, a general term for low functional and intellectual functioning, may also be due to genetic abnormalities of which DS is one. John Langdon Down was the first person to describe the characteristic traits of individuals with DS in 1866. This English physician is called the father of Down syndrome. Close to a century later Professor Lejeune of France learned that DS had a genetic origin. There were times when individuals with DS were believed to be possessed by evil spirits and treated as if they were a burden on society. They were hidden by their parents, demoralised, abused and even persecuted. They did not have the same rights as ordinary citizens.

DS occurs when there is a full or partial extra copy of the 21st chromosome (Figure 2.1), and is the most prevalent chromosomal cause of ID (National Down Syndrome Society, NDSS, 2014; Barnhart & Connolly, 2007:1399). In medical practice prenatal screening and prenatal diagnoses (with close to

100% accuracy) is performed with the aid of an amniocentesis or chorionic villus sampling can determine DS before 16 weeks of pregnancy.

The extra chromosome results in a disruption of gene expression and influences the structure and function of all physiological systems. DS is not a disease but a genetic abnormality. It is associated with an intellectual disability that is not a sign of future limitations or functional incapacity. Individuals with DS have unique physical characteristics, altered development patterns, and most individuals have moderate to mild intellectual disability (NDSS, 2014). Unlike those with ID without DS, DS is dispersed across all socio-economic classes. In today's society many individuals with DS attend school, work, recreate and participate in sporting events and has recently been included in the Special Olympics. There are many initiatives, such as South Africa's National DS Society, that drive DS inclusion into everyday society. There is also an International DS Conference held every three years promoting fundamental issues for optimal development such as sound education, stimulating environments, adequate health care, and social support. Due to improved healthcare and education, most adults with DS can hold a job or function well in a centre for the intellectually disabled (Smith, 2001:1031). Many individuals with DS function independently in the community with minimal support for most of their adult life (Smith, 2001:1031).

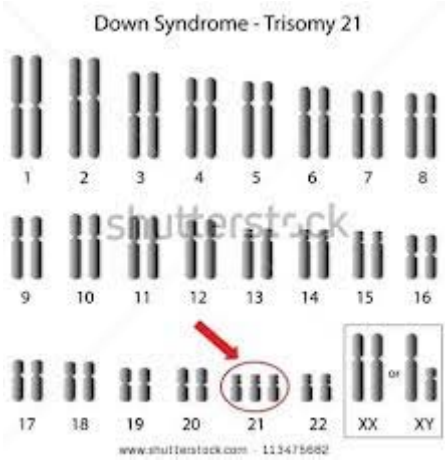

Figure 2.1: Down syndrome karyotype

There are a number of unique physical characteristics associated with DS (Figure 2.2). Some of these include: short stature, small nose with flat bridge, eyes slanted upward and outward, exaggerated folds of the skin around the eyes, short legs and arms in relation to torso, short neck, small low-set ears, small head and face, broad hands and feet and stubby fingers and toes (NDSS, 2014; Winnick & Short 2005:55).

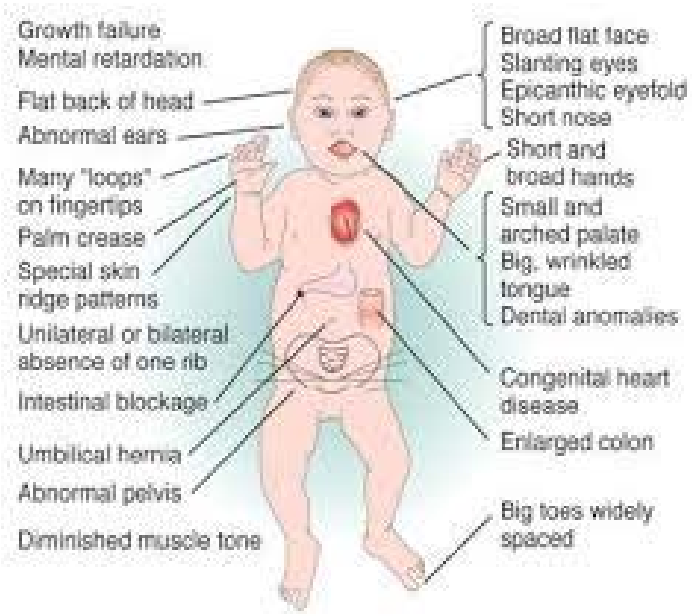

Figure 2.2: Common characteristics of individuals with Down syndrome

2.3.2 Causal factors of Down syndrome

DS results from three likely causes that are all chromosome abnormalities. All three causes are related to a full or partial extra copy of the 21st chromosome. Trisomy 21 is by far the most frequent occurrence (95% of all cases) and results, as the name suggests, in an extra chromosome 21 (Mutton *et al*., 1996:387). These individuals have 47 chromosomes instead of 46 (23 from each parent). A second cause is translocation (4% of all cases) and as in trisomy 21, there is still a third chromosome 21, but one of these grow onto another chromosome, appearing as one, but containing the genetic material of two chromosomes. The most common being Robertsonian where chromosome 14 and 21 are involved. Lastly, a third possible form is mosaic DS (1% of all cases). These individuals have two cell lines, one of which contains 46 chromosomes and the other 47 chromosomes. These are the individuals with DS who are most likely to attend school or university. In Figure 2.3 below, the first picture demonstrates normal typical cell division, the second shows trisomy 21 (non-disjunction) cell division and the third mosaic cell division. Translocation is not shown.

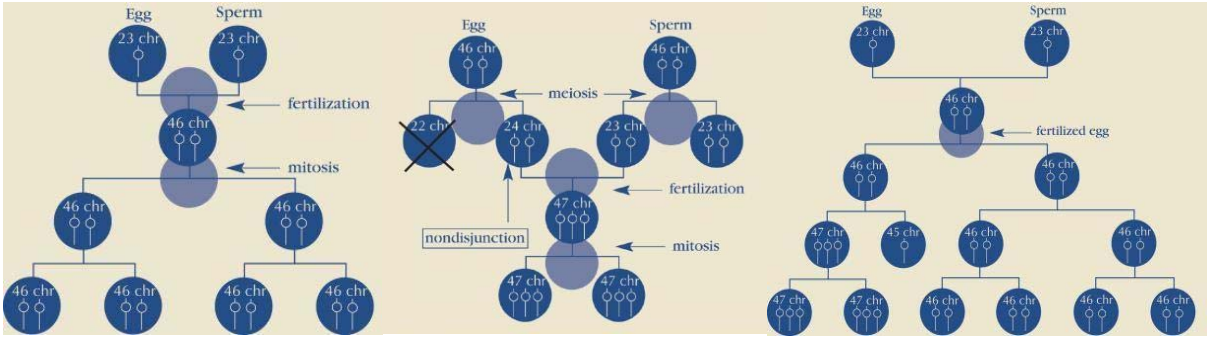

Figure 2.3: Normal cell division, nondisjunction cell division and mosaic cell division.

The maternal age has an influence on trisomy 21 but not on translocation (Mutton *et al*., 1996:387). The odds of having a child with DS increase when the maternal age is more than 35 years (NDSS, 2014). Only the translocation type carries a hereditary gene. Thirty-three percent of translocations are inherited from the parents, six-fold more often from the mother (Mutton *et al*., 1996:387).

There are more men with DS compared to women but when mosaics and twins are considered there are more women with DS in total (Mutton *et al*., 1996:387). Shin *et al*. (2009:1565) also found that there are more men with DS compared to women. They also showed that the prevalence was higher among non-Hispanic white people compared to non-Hispanic black people and other ethnic groups.

2.3.3 Prevalence of Down syndrome

The number of individuals with DS in the USA is approximately 250 700 and the prevalence is currently 8.27/10 000 (Presson *et al*., 2013:1163). This finding is based on proportions of deaths by age from 1968 to 2007. The authors explained that this estimate is 25 to 40 per cent lower than that based exclusively on current birth prevalence. Another study in the United Kingdom showed that 1.72 out of 1000 pregnancies was affected by DS (Irving *et al.,* 2008:1366). The proportion, however, increased over the 20 years (1985-2005) from 1.3 to 2.5 per 1000 total births. A total of 389 pregnancies were terminated and 51 were still-born. Concerning live births, a ratio of 1.08 per 1000 was found. It was found that total live births decreased by 20% over 20 years. The authors concluded that although the number of pregnancies with DS has increased, there has been no change in live birth prevalence. This discrepancy is due to increased maternal age and improved treatment of babies with DS after birth which is counterbalanced by the effects of prenatal diagnosis and subsequent abortions. On the other hand, another study found that the prevalence of DS increased by 31.1% (9.0 to 11.8 per 10 000 live births) from 1979 to 2003 (Shin *et al*., 2009:1565). They attributed the increase to later child-bearing by mothers.

There are no published reports on prevalence of DS in South Africa but Down Syndrome South Africa (DSSA) published findings that 1 in a 1000 live births in developed countries are DS and this statistic increases to 0.5 per 1000 in developing countries (DSSA, 2014). However, such approximations rely on a number of assumptions such as a fixed South African population, similar regularity of death in persons with DS as in the general population and a steady incidence of DS over time. This lack of information reflects the priorities, challenges and perceptions that are carried within the South African context with regards to persons with intellectual disabilities and in particular toward DS. The lack of information contributes to the challenges to introduce change and to monitor change as a result of intervention.

2.3.4 Life expectancy of persons with Down syndrome

The life expectancy of individuals with DS has increased radically over the past three decades with many individuals reaching middle to old age (Torr *et al*., 2010:70; Chicoine & McGuire, 1997:477). Chicoine and McGuire (1997:477) reported that a woman with DS lived up to the age of 83 in America. Specifically, Yang *et al*. (2002:1019) proved that the mean life expectancy has increased from 25 years in 1983 to 49 years in 1997. The mortality in individuals with DS is the highest among those with congenital anomalies (Abbag, 2006:219). If early intervention strategies for congenital abnormalities had been implemented, the one-year survival increased tremendously, reaching almost 100% (Irving *et al*., 2008:1336). Even though life expectancy has increased in this population, it has been shown that individuals with DS have a fundamentally different ageing process (Barnhart & Connolly, 2007:1400–1401).

2.3.5 Health of individuals with Down syndrome

2.3.5.1 Chronic diseases of lifestyle

DS individuals are born with many health-related disorders of which congenital heart disease (61% of all cases) is the most common (Abbag, 2006:219). The most common congenital heart disease reported was ventricular and atrioventricular septal defect (56% of all cases) (Abbag, 2006:219). Early intervention of congenital heart anomalies is crucial in this population (Abbag, 2006:219). These individuals are at greater risk of developing thyroid problems, leukaemia and respiratory complications that will eventually require surgery or treatment (de Asua *et al*., 2015; Pikora *et al*., 2015; NDSS, 2014; Hermon *et al*., 2001:167; Smith, 2001:1031). Yang *et al*. (2002:1019) showed that these conditions were often the cause of mortality. Specifically, the authors showed that the most common causes of death in descending order of frequency were congenital heart disease, dementia, hypothyroidism and leukaemia. Leukaemia was the only form of cancer frequently encountered in this population with all other malignant type conditions having a strikingly low odds ratio. The authors further explained that this could be due to a decreased exposure to environmental factors, tumour-supressing genes on chromosome 21 or a slower rate of replication of cancerous cells.

Adults with DS are also at a greater risk of developing diabetes, Alzheimer's disease, depression, obsessive-compulsive disorder and epilepsy (Hermon *et al*., 2001:167). Torr *et al*. (2010:70) have reiterated that dementia of the Alzheimer's type was highly prevalent in this population especially in the sixth decade of life. In all these conditions, behaviour change or loss of a function may be the only signs of the disease (Smith, 2001:1031). A recent study conducted in Western Australia reported the prevalence of medical conditions in young adults with DS (Pikora *et al*., 2015)(Table 2.3), and indicated

that most of the parents reported that these conditions negatively impact the employment opportunities and leisure activities in this population.

Table 2.3: Prevalence of medical conditions in young adults with Down syndrome (n=197).

Condition	%
Eye and vision problems	73
Ear and hearing	45
Cardiac	25
Respiratory	36
Musculoskeletal	61
Body weight	57
Skin	56
Mental health	32
Menstrual conditions	58

Although the life expectancy of individuals with DS has increased they still age prematurely as explained by increased mortality, and plummeting functional fitness and weight values into old age (Pikora *et al.,* 2015; Terblanche & Boer, 2013:834; Carmeli, Kessel, *et al*., 2004; Rubin *et al*., 1998:178). Torr *et al*. (2010:70) also reported functional decline in this population with early onset of age-related diseases. In addition, most individuals with DS develop Alzheimer's disease in old age (Zigman & Lott, 2007:237; Folin *et al*., 2003:267). By the age of 50 years, most adults with DS have developed (or are in the process of developing) senile plaques and neurofibrillary tangles indicative of Alzheimer's disease (Mann & Esiri, 1989:169). De Asua *et al*. (2015) and Torr *et al*. (2010:70) pointed out that it was imperative to perform regular screening, monitoring and preventative interventions in this population to limit conditions associated with ageing and premature disability. They advised that specialist disability physicians and geriatrics had to play a prominent role in this regard.

2.3.5.2 Overweight and obesity

A large percentage of individuals with DS are overweight or obese (Terblanche & Boer, 2013:830; Pitetti *et al*., 2013:47; Melville *et al*., 2008:425; Carmeli *et al*., 2002[b]:460; Rubin *et al*., 1998:175; Prasher & Filer, 1995:437). Rubin *et al*. (1998:178) demonstrated that 45% of men and 56% of women are overweight whilst Terblanche and Boer (2013:830) found that 79% of men and 95% of women are overweight in a sample of 371 individuals with DS in South Africa as obtained with the BMI. It may not be appropriate to use the BMI as an indication of obesity in this population due to their inherent short stature. However, a study by Baptista *et al*. (2005:382) indicated a higher body fat mass in this population compared to the general population. Furthermore, individuals with DS have an increased risk of obesity compared to individuals with ID without DS (Melville *et al*., 2008:425). Rubin *et al*. (1998:175) reported that individuals with DS living in a private home had a greater tendency to be

overweight compared to those living communally. One of the reasons for this finding may be easier access to food.

Being overweight is a risk factor for cardiovascular disease and could act as a mediator to develop other chronic conditions in this population (Rubin *et al*., 1998:179). Obesity has been associated to hypertension, diabetes, premature ageing and other health-related diseases in the general population (Lacobellis *et al*., 2012:767; Chiang *et al*., 2011:700; Niemann *et al*., 2011:577). It is uncertain if the same association is found in the DS population. Moreover, we are unsure if adults with DS in developed and developing countries are at the same risk for cardiovascular disease.

2.3.5.3 Lifestyle

Most individuals with DS live sedentary lifestyles (Nordstrøm *et al*., 2013:4395; Esposito *et al*., 2012:109; Shields *et al*., 2009:307). Only 42% of children with DS performed at least 60 minutes of moderate to vigorous exercise per day (Shields *et al.,* 2009:307). A study in Norway reported that only 12% of adults with DS, Prader-Willi syndrome and Williams syndrome (18–45 years) met the required physical activity levels (Nordstrøm *et al*., 2013:4395). Furthermore, adults with DS were the least active of these three groups. Another study reported that young children with DS (3–10 years) did not meet the required vigorous activity needed per day when compared to their non-DS peers (Whitt-Glover *et al*., 2006:158). Finally, a review article reiterated that children and adolescents with DS did not meet the required amount of aerobic physical activity (Pitetti *et al*., 2013:47). Moreover, they showed that the amount of physical activity decreased from childhood to adulthood. This may indicate that the root of a habitually sedentary lifestyle is initiated at a very young age. However, Hinckson and Curtis (2013:12) reported that there was a clear deficiency in studies reporting reliability and validity of measuring instruments to analyse physical activity in children with ID. It is clear from the literature presented that in general the required physical activity guidelines are not met in children, adolescents and adults with DS.

Nevertheless it is recommended that future studies focus on practical ways to get this population habitually active from a young age (Kerstiens & Green, 2015:192; Pitetti *et al*., 2013:47). In fact, Rubin and colleagues (1998:175) pointed out that it should be a major public health concern to find ways to get this population more active and to an optimal body composition. Wuang and Su (2012:841) reported that those children with DS who had better cognitive and motor functions engaged in physical activities more often and relished them more. The focus should perhaps be to target those individuals who do not possess these attributes. Also, those individuals with a high BMI do not necessarily have to be targeted as Nordstrøm *et al*. (2013:4395) reported no association between physical activity levels and BMI in adolescents and adults with DS. However, they did not distinguish between moderate and vigorous physical activity as the study by Whitt-Glover *et al*. (2006:158). These authors stipulated that children

with DS should spend more time in vigorous physical activity that may prevent obesity and secure future lifelong health.

2.3.6 Physical activity, exercise and standardised tests

The physical fitness of individuals with DS is poor compared to the general population and to those with ID but without DS (Baynard *et al*., 2008:1984; Baynard *et al*., 2004:1285; Pitetti & Fernhall 2004:219; Guerra *et al*., 2003:1604; Fernhall, McCubbin, *et al*., 2001:1657; Fernhall *et al*., 1996:366**;** Eberhard *et al*., 1989:167). The physical fitness of individuals with DS may be poor because they have a set of health, cognitive, physiological, and psycho-social traits predisposing many of them to limited exercise (Pitetti *et al*., 2013:47). A poor aerobic capacity is considered to be a risk factor for cardiovascular diseases and can result in a reduced lifespan for individuals with DS (Gonlález-Agüero *et al*., 2010:720). Low levels of physical fitness may cause functional deterioration as well as reduce bone mineral density, worsening existing clinical conditions (Gonlález-Agüero *et al*., 2010:717) and increase the fall risk.

In a review article it was explained that the reasons for a poor aerobic capacity in this population is largely unknown (Gonlález-Agüero *et al*., 2010:716). Wee *et al*. (2015:198) reported that the influence of obesity and age does not have an effect on the low aerobic capacities of adults with DS. A recent review study proposed three possible causes for low aerobic capacities namely autonomic dysfunction, reduced ventilatory parameters and metabolic dysfunction (Pitetti *et al*., 2013:49). Chronotropic incompetence could be a possible causal factor due to autonomic dysfunction. Baynard *et al*. (2008:1984) showed that the maximal heart rate for all age groups of individuals with DS is consistently lower compared to the general population. Fernhall *et al*. (2009:724) went one step further and proved that blunted catecholamine response to peak exercise is the primary constraint to chronotropic incompetence. Pitetti *et al*. (2013:49) added that the interaction of obesity and autonomic control may also negatively impact a reduced aerobic capacity. Ventilatory parameters such as peak minute ventilation and the ventilatory equivalents could restrict aerobic capacity due to the large tongues of individuals with DS but Fernhall and Pitetti (2001:176) proved that these were appropriate for a given VO_2 value even though the pulmonary function of adults with DS is poorer compared to age matched controls in the general population (Salgueirinho *et al*., 2015). Lastly, metabolic limitations (such as the ventilatory threshold) are difficult to assess in this population (Pitetti *et al*., 2013:49). Future studies should perform blood lactate testing in conjunction with maximal exercise tests to determine whether a reduced lactate threshold causes physiological limitations to a reduced aerobic capacity. In addition, supramaximal tests such as the Wingate test has not been reliable in providing a complete metabolic profile for this population (Guerra *et al*., 2009:47). There are also other frequently encountered health-related problems that could inadvertently affect the aerobic capacity of individuals with DS such as muscle hypotonicity, ligamentous laxity and joint hypermobility (NDSS, 2014).

As discussed in Chapter 1, adults with DS do not have a standardised battery of instruments to measure functional fitness. Individuals in the general population (children, adults and elderly adults) and those with physical and intellectual disabilities have measuring instruments to ascertain functional fitness parameters such as flexibility, balance, coordination, aerobic capacity, functional capacity and muscular strength and endurance. Adults with DS are consequently pooled with individuals with ID without DS, even though the presence of DS negatively affects test performance. A standardised functional test battery of items is especially important in a population whom we know suffer from many health, functional and physical limitations. Currently health professionals do not have standardised test for functional fitness or norms to categorise functional fitness in persons with DS. Consequently, the health professional cannot identify specific weaknesses or strengths to focus interventions on for the improvement of activities of daily living and self-care. Persons presenting with below normative data are the target group for interventions to pro-actively prevent future conditions or diseases. Furthermore, if a proper identification of strengths and weaknesses are not made, training programs cannot be tailored to the specific needs of the individual and the effectiveness of a particular training intervention cannot be studied. In a previous study (Boer, 2010:105) the feasibility and discriminant reliability of various functional fitness tests was examined over five months in 371 adults with DS across seven provinces in South Africa. Findings from the study indicated that standardisation of functional testing specific to the DS population is needed. Research addressing the reliability and validity of these tests, will contribute significantly to assessment procedures by health practitioners in this neglected population.

2.3.6.1 Facilitators and barriers to a habitual active lifestyle

Mahy *et al*. (2010:795) performed semi-structured interviews with adults diagnosed DS and their immediate families in order to determine facilitators and barriers to physical activity. Facilitators to exercise were identified as support from others, physical activity that was fun and that has a stimulating goal, routine or familiarity. Barriers on the other hand were identified as lack of support, not wanting to engage in physical activity and medical or physiological factors. They added that individuals with DS did not initiate physical activity but that it was enforced by support staff or family. Individuals with DS were also more likely to engage in physical activity that they had mastered and participated in frequently. Heller *et al*. (2003:162) also stipulated that accessibility to exercise facilities had to be improved and the awareness of the importance of exercise as perceived by the carers had to be reinforced. Often the support staff were not active themselves and would be unlikely candidates to assist these individuals with training exercises. Mahy *et al*. (2010:802) echoed this belief and stipulated the importance of educating support staff about the significance of a physically active lifestyle.

Unlike individuals with DS, those with ID did not identify that a support person was needed to enforce physical activity (Mahy *et al*., 2010:803). Financial issues were not seen as a constraint. They explained that the answer for this incongruity might lie in the poorer functionality of persons with DS.

A very successful and intuitive study by Heller *et al*. (2004:175) in older adults with DS proved that a combined exercise and health education program could overcome the many barriers that these individuals face. After 12 weeks of health education and training, the exercise group demonstrated significant changes on their perception of exercise. These included an increased exercise self-efficacy, less cognitive–emotional barriers, more positive anticipated results, improved life satisfaction, and lower depression. Moreover, another study by Shields *et al*. (2011:360) demonstrated that the perceptions and attitudes of those individuals who facilitate training in this population could be positively and significantly altered.

2.3.6.2 Effect of physical activity on aerobic capacity

Again, as studies in the intellectually disabled field have shown, adults, adolescents and children with DS can improve parameters associated with physical fitness (Cowley *et al*., 2011:2229; Mendonca *et al*., 2013:353; Mendonca *et al*., 2011:37; Gupta *et al*., 2011:425; Shields *et al*., 2010:187; Mendonca & Pereira, 2009:33; Lewis & Fragala-Pinkham, 2005:30; Carmeli, Barchad, *et al*., 2004:180; Rimmer *et al*., 2004:165; Tsimaras *et al*., 2003:1239; Carmeli *et al*., 2002[c]:106).

Considering an improvement in aerobic capacity, two studies showed no improvement after an aerobic intervention period of jogging/walking and rowing although there were improvements in peak exercise time and work performance (Varela *et al*., 2001:135; Millar *et al*., 1993:270). On the other hand, two other studies reported significant improvements in aerobic capacity (Mendonca & Pereira 2009:33; Tsimaras *et al*., 2003:1239). Improvements in the study by Mendonca and Pereira (2009:33) could be explained due to the fact that a 28-week intervention period was used compared to the studies by Millar *et al*. (1993:270) and Varela *et al*. (2001:135) where the intervention period lasted less than 16 weeks. Secondly, participants in the study trained at 60% to 85% of VO_2 peak whereas participants in the other two studies trained at lower intensities (55% to 70% peak VO_2 and 65% to 75% of maximum heart rate). Lastly, exercise intensity was not always monitored during the intervention period of these two studies. The study by Mendonca and Pereira (2009:37) reported highly significant improvements in peak aerobic capacity of up to 27.8%. Improvements of this nature are clinically significant as most individuals with DS have VO_2 peak values well below normative values. The study by Tsimaras *et al*. (2003:1243) also lasted less than 16 weeks but its success in significantly improving the aerobic capacity could perhaps be attributed to the nature of training. Training was performed on a treadmill, which is a weight-bearing activity, whereas the other three studies performed non-weight-bearing activities such as cycling or rowing ergometer training.

Studies that used combined aerobic and resistance training as an intervention study also showed significant improvements in aerobic capacity (Mendonca *et al*., 2013:353; Mendonca *et al*., 2011:37;

Lewis *et al*., 2005:30; Rimmer *et al*., 2004:165). Mendonca *et al*. (2011:37) showed that the magnitude of improvements in aerobic capacity were identical when the results of adults with DS were compared to adults without DS after the same intervention protocol. Collectively, it seems that a combination of progressive resistance and aerobic exercise may have a larger impact on cardiovascular fitness than aerobic exercise alone. This is supported by the findings of Pitetti & Boneh (1995:423) who found that there was a strong correlation between VO_2 peak and leg strength in people with DS. Resistance-based exercise, especially of the lower limbs, may thus provide the individual with a better capacity to use his/her legs during running or cycling. It was also shown that leg strength is a strong predictor of performance in everyday functional abilities in a DS population (Cowley *et al*. 2010:388). What is also interesting to note from these studies is that the VO_2 peak improved by 1% to 20%. As such, some individuals with DS only appear to improve their VO_2 peak marginally while others appear to improve more significantly. Similar findings were observed in a study by Bricourt *et al*. (2008:559) with VO_2 peak values ranging from 23–60 ml/kg/min in a physically active group. Perhaps the small change seen in some individuals with DS could be attributed to the many physiological barriers they face such as heart abnormalities, skeletal muscle hypotonia, joint hypermobility and chronotropic incompetence.

Collectively, a meta-analysis in 2005 reported that aerobic or combined aerobic and resistance training were effective in increasing the time to exhaustion, maximum workload achieved, VO_2 peak and peak minute ventilation in individuals with DS. All effect sizes for these four variables were greater than 0.71. Time to exhaustion showed an improvement of up to four minutes. This meta-analysis did not include the more recent studies that also reported significant findings in support of those mentioned (Mendonca *et al*., 2013:353; Mendonca *et al*., 2011:37; Lewis *et al*., 2005:30).

2.3.6.3 Effect of physical activity on submaximal exercise capacity

Mendonca and Pereira (2009:33) proved that 28 weeks of aerobic training significantly improved submaximal exercise capacity as shown by the respiratory exchange ratio. They explained that this was a positive physiological adaptation to exercise and might be explained to a greater recruitment of lipids (Mendonca & Pereira 2009:36). This was also shown in a case study by Lewis *et al*. (2005:30) where heart rate and respiratory exchange ratio values at each submaximal stage of the maximal exercise test decreased. No studies have analysed the effect of exercise training on submaximal lactate values or the lactate threshold. One study attempted to identify the ventilatory threshold (e.g. V-slope method) in this population but explained that in most individuals with DS it is extremely difficult to pinpoint (Baynard *et al*., 2004:1285). The improvement of submaximal exercise capacity is important as most activities of everyday living are submaximal (Mendonca & Pereira 2009:34).

2.3.6.4 Effect of physical activity on body composition

Aerobic training

Ordonez *et al*. (2006:416) was the only study to report a significant decrease in body fat percentage (32% to 26%) in adolescents with DS after water- and land-based aerobic activities lasting 12 weeks. Similarly, there was only one study to show a significant decrease in fat mass after a 28-week aerobic training program in adult men with DS (Mendonca & Pereira, 2009:33). Numerous studies reported no changes in fat mass with aerobic training (Varela *et al*., 2001:135; Tsimaras *et al*., 2003:1243; Millar *et al*., 1993:270). Furthermore, we are not aware of any studies that have reported body mass reduction after an aerobic training regime.

Combined aerobic and resistance training

Rimmer *et al*. (2004:165) reported small but significant weight loss in adults with DS after a combined aerobic- and resistance-based training program lasting 12 weeks. Gonlález-Agüero *et al*. (2011:2383) performed a combined conditioning and plyometric training program for 21 weeks and found that lean mass improved significantly but no change resulted in fat mass. Unlike the study by Rimmer *et al*. (2004:165), the studies by Mendonca *et al*. (2011:37) and Lewis *et al*. (2005:30), which also used a combined aerobic and endurance training protocol for the same duration (12 weeks), showed no changes in either body mass or fat mass in a DS population. This finding was also shown in a more recent study by the same author (Mendonca *et al*., 2013:356). Two recent studies involving individuals with ID without DS, also demonstrated dissimilar results regarding the effect of combined aerobic and resistance exercise on weight and fat loss (Calders *et al*., 2011:1100; Elmahgoub *et al*., 2009:1327). The study by Calders *et al*. (2011:1100) reported no significant loss in body or fat mass nor in waist circumference whereas the study by Elmahgoub *et al*. (2009:1327) determined small but significant improvements in all of these variables. The discrepancies between these findings may be attributed to different training intensity. However, it is difficult to compare as the one study trained at 90% of ventilatory anaerobic threshold (Calders *et al*., 2011:1100) whilst the other trained at 60% of heart rate reserve (Elmahgoub *et al*., 2009:1329). Many other methodological and procedural factors may also have caused the discrepancies between studies.

A meta-analytic study by Dodd and Shields (2005:2051) also revealed no significant body weight ameliorations after a period of aerobic or combined aerobic and endurance training (Cohen's d = 0.09). It is not clear why significant weight loss is absent in the vast majority of intervention studies. Perhaps the intensity at which aerobic exercise takes place is not sufficient to stimulate the necessary metabolic, biochemical or physiological processes to drive weight loss. In most of these studies exercise intensity was based on the percentage of VO_2 peak and the VO_2 peak of the majority of individuals with DS is so

low that the training stimulus may perhaps be insufficient to alter body mass or body composition. Also, most individuals with DS do not reach a VO_2 maximum, hence the term VO_2 peak. If training is based on VO_2 peak the intensity of exercise may be too low. Also, many of the studies performed training on a bicycle or rowing ergometer which are non-weight-bearing and perhaps an inadequate stimulus for fat loss. Lastly, nutrition was not controlled or monitored in any of the studies. Training may promote the individual's appetite and result in excess food intake during the intervention period.

2.3.6.5 Effect of physical activity on functional ability

A low physical fitness limits the ability of individuals with DS to perform functional tasks of daily living (Cowley *et al*., 2010:388). It has been shown that aerobic training, resistance training and combined aerobic and resistance training can improve various functional tasks of daily living such as a timed up-and-go test, grocery shelving task, time to ascend and descend chairs (Cowley *et al*., 2011:2229; Shields & Taylor, 2010:187, Shields *et al.,* 2008:1215; Carmeli, Barchad, *et al*., 2004:180, Carmeli *et al.,* 2002[c]:106).

2.3.6.6 Effect of physical activity on muscular strength

Individuals with DS have reduced muscle strength compared to the general population and reduced leg strength compared to both the general population and individuals with ID without DS (Carmeli *et al*., 2002:316; Croce *et al*., 1996:369; Pitetti *et al*., 1992:847). Muscle strength is an important predictor of functional performance and everyday living activities in this population (Terblanche & Boer, 2013:830 Cowley *et al*., 2010:388).

Shields *et al*. (2008:1215) performed a community-based progressive resistance training program on 20 adults with DS. They determined that resistance training improved muscle strength, muscle endurance and physical function of the upper limbs. Improvement in upper limb strength, endurance and function is of practical value to these individuals who are involved with manual labour tasks. The authors attributed the absence of changes in lower limb improvements to high baseline values with an intervention period that was too short (10 weeks).

On the other hand, Carmeli *et al*. (2002[c]:106) proved that a simple but long-term (25 weeks) walking program improved lower-limb muscle strength, functional ability, walking duration (+150%), walking speed (+86%) and walking distance (+180%) in a geriatric DS population. Isokinetic strength measures (mean peak torque and peak torque) of the quadriceps and hamstring muscles improved significantly. Furthermore, functional ability as assessed with a timed up-and-go also improved. What makes this study unique is that aerobic training only resulted in these improvements in lower-limb muscle strength and function. Shields and Taylor (2010:187); Tsimaras and Fotiadou (2004:343) and Cowley *et al.*

(2011:2229) also showed that lower-limb muscle strength, endurance, balance and functional ability (stair climbing) can be improved (in adults and adolescents with DS) after 10 to 12 weeks with a resistance training protocol. An improvement in lower-limb muscular strength and balance was also shown in children with DS after only six weeks of strength training (Gupta *et al*., 2011:425).

Lastly studies that incorporated combined aerobic and strength training have also reported improvement in trunk, lower and upper limbs (Mendonca *et al*., 2013:353; Mendonca *et al*., 2011:42; Lewis *et al*., 2005:30; Rimmer *et al*., 2004:165). Many of the strength measure improvements were similar or even better when compared to age-matched individuals in the general population (Mendonca *et al*., 2013:356; Mendonca *et al*., 2011:42). Significant improvements were also reported in the ID population without DS with combined aerobic and resistance training (Calders *et al*., 2011:1100; Elmaghoub *et al*., 2011:2274; Elmahgoub *et al*., 2009:1327).

All of these researchers expressed the importance of adequate strength in this population as many DS individuals perform physical- rather than cognitive-related work. Also, improvements in strength helped these individuals with functional tasks of everyday living (Cowley *et al*., 2011:2229; Shields *et al*., 2008:1215; Carmeli *et al*., 2002[c]:106). A further advantage of improved muscle strength (especially of the lower limbs) could lead to an improved aerobic capacity as Pitetti and Boneh (1995:423) reported a very close relationship between these variables for adults with DS. This finding is very important as muscular fatigue may occur before the cardiopulmonary system is maximally stressed in maximal exercise tests. It is therefore possible that the aerobic capacity of individuals with DS is underestimated as maximal exercise may not stress these individuals to true aerobic limits. That is why a combined aerobic and resistance training program may be a better alternative for improvements in aerobic capacity compared to aerobic training alone.

2.3.6.7 Effect of physical activity on balance

Carmeli *et al*. (2002c:106) found that aged adults with DS can significantly improve muscle strength and dynamic balance by adopting suitable programs of treadmill walking. However, a year later, these researchers (Carmeli *et al*., 2003:767) could not repeat these findings using an intervention of treadmill training and ball exercises in adults with ID. The participants only improved their performance in two of five balance tests. They speculated that either the small sample size or the non-compliance of two individuals to the training program may have influenced the results.

Wai-Yiwang and Yun-Hueiju (2002:443) investigated balance and jumping performance in 20 DS children (three to six years). Thirty children without DS acted as the comparison group. Children with DS completed three sessions of jump training per week for six weeks, while the children without DS were the controls. Both balance and jumping skills were significantly better in the DS children and it

was concluded that DS children have the ability to improve balance and jumping skills. These findings were confirmed by Tsimaras & Fotiadou (2004:343) who studied the effects of training on quadriceps femoris and hamstring muscle strength and dynamic balance in a DS population. Dynamic balance was measured using a balance deck and determined by a stabilometer in 30-, 45-, and 60-second intervals. The exercise group performed 15- to 20-minute training periods consisting of dynamic balance activities and plyometric exercises with and without resistance. This group showed a significant improvement in dynamic balance (30 seconds: $p<0.01$; 45 seconds: $p<0.001$; 60 seconds: $p<0.01$).

Lewis *et al*. (2005:30) performed a case study on a child with DS and applied a similar combined aerobic and strength training regime as Rimmer *et al*. (2004:165). The study was performed on a 10.5-year-old girl with DS. Not only were gains in cardiovascular and strength variables observed, but the program successfully improved her balance, coordination, and power in gross motor tasks.

2.4 Exercise intervention for persons with Intellectual disability

Interval training (IT) was shown to have a significant and positive impact on various physiological, anthropometrical and functional aspects in the general population. IT involves vigorous exercise performed at a high intensity for a brief period of time interposed with recovery intervals of low-to-moderate intensity or complete rest. The modes of IT training usually include running/walking on a treadmill or cycling on an ergometer.

The benefits associated with this training modality were demonstrated in a variety of populations. Some of these population groups even included individuals with chronic medical conditions such as coronary artery disease and heart failure and some who had suffered a stroke or heart attack (Globas *et al*., 2012:85; Hesse *et al*., 2011:146; Ingul *et al*., 2010:852; Babraj *et al*., 2009:1; Gibala & McGee, 2008:58; Praet *et al*., 2008:163; Helgerud *et al*., 2007:665). Many studies showed that the physiological, anthropometrical and functional benefits of IT were superior to traditional continuous aerobic training (CAT) (Ciolac *et al*., 2011:824; Smart & Steele, 2011:205; Moholdt *et al*., 2009:1031; Tjønna *et al*., 2008:346; Wisløff *et al*., 2007:3086; Nemoto *et al*., 2007:803; Rognmo *et al*., 2004:216) and this was confirmed by a meta-analysis (Hwang *et al*., 2011:378). The tolerance and compliance of high-intensity exercise in this meta-analysis was reported to be excellent. Moreover, IT was shown to be more fun and less time-consuming than traditional CAT (Bartlett *et al*., 2011:547). We are not aware of an IT intervention that has been performed on individuals with ID or DS.

2.4.1 Types of interval training

IT can be categorised into two distinct types namely sprint interval training (SIT) and aerobic interval training (AIT). SIT is characterized by 4–6 cycles of 30-second 'all out sprints' followed by 4–4.5

minutes of recovery. AIT is performed at a slightly lower intensity (80% to 95% of peak heart rate) than SIT but for longer periods of time (4 minutes per exercise bout). However, many of the protocols that were used are highly variable with respect to work ratio, rest ratio, work-to-rest ratio, type of rest, exercise intensity (based on % of HR max or VO_2 max), minutes per exercise session, duration of study and type of physical activity (running, cycling) (Table 2.4).

Table 2.4: Different types of interval training protocols in general and clinical populations

Study	Con	Mode	Type of IT	Rest	Intensity	Duration
Boudou *et al*., 2003	T2D	Cycling	8-second sprint for 20 minutes	12-second recovery	80–90% of peak heart rate	8 weeks
Burgomaster *et al*., 2008	GP	Cycling	4–6 Wingates	4.5 minutes	All out	6 weeks
Gibala *et al*., 2012	GP	Cycling	30-second sprint (4–6 times)	4.5 minutes rest	All out	12 weeks
Helgerud *et al*., 2007	CAD	Running	15 seconds	15 seconds at 70% max HR	90% of max HR	8 weeks
Ingul *et al*., 2010	Obese	Running	4 x 4 minutes	3 minutes of active rec	90% of max HR	13 weeks
Moholdt *et al*., 2009	Bypass surgery	Running	4 x 4 minutes	3 minutes at 70% of max HR	90% of max HR	4 weeks
Nemoto *et al*., 2007	HBP	Walking	5 x 3 minutes	3 minutes at 40% of VO_2 peak	70% of VO_2 peak	21 weeks
Rognmo *et al*., 2004	CAD	Running	4 x 4 minutes	3 minutes at 55% VO2 peak	80–90% of max HR	10 weeks
Smart *et al*., 2011	HF	Cycling	30 x 60 seconds	60 seconds of passive rest	70% of VO_2 peak	16 weeks
Talanian *et al*., 2007	GP	Cycling	10 x 4 minutes	2 minutes passive rest	90% of VO_2 peak	2 weeks
Tjonna *et al*., 2009	Obese	Running/ Walking	4 x 4 minutes	3 minutes at 70% of max HR	90% of max HR	12 weeks
Tjonna *et al*., 2008	MS	Running	4 x 4 minutes	3 minutes at 70% max HR	90% of max HR	16 weeks
Warburton *et al*., 2005	CAD	Running	7 x 2 minutes	2 minutes	90% of max HR	16 weeks
Wisloff *et al*., 2007	HF	Running	4 x 4 minutes	3 minutes at 70% of max HR	90–95% of max HR	12 weeks

CAD: coronary artery disease; Con: condition; GP: general population; HBP: high blood pressure; HF: heart failure; MS: metabolic syndrome; T2D: type 2 diabetes

2.4.2 Effect of interval training on aerobic, anaerobic and submaximal exercise capacity

It was demonstrated in many studies that the maximal and submaximal exercise capacity can be significantly improved (range: +6% to +46%) after various IT training protocols (Table 2.5) (Gist *et al*., 2014:269; Rustad *et al*., 2014:181; Ciolac *et al*., 2011:824; Whyte *et al*., 2010:1421; Tjonna *et al*., 2009:317; Burgomaster *et al*., 2008:155; Tjonna *et al*., 2008:346; Trapp *et al*., 2008:684; Schjerve *et al*., 2008:282; Helgerud *et al*., 2007:665; Talanian *et al*., 2007:1439; Wisloff *et al*., 2007:3086; Warburton *et al*., 2005:1080; Rognmo *et al*., 2004:216). IT was shown to increase aerobic capacity after only two weeks of training (Whyte *et al*., 2010:1421; Talanian *et al*., 2007:1439). It is also important to note that aerobic capacity continued to improve for another three months after the 3-month intervention study with unsupervised home-based training (Moholdt, 2009:1035). IT was also proven to hold superior benefits compared to traditional CAT (Table 2.5) (Pattyn *et al*., 2014:687; Smart *et al*., 2011:205; Ciolac *et al*., 2011:824; Hwang *et al*., 2011:378; Tjonna *et al*., 2009:317; Tjonna *et al*., 2008:346; Wisloff *et al*., 2007:3086; Nemoto *et al*., 2007:803; Rognmo *et al*., 2004:216) or similar improvements but with reduced total training time (Gist *et al*., 2014:269; Burgomaster *et al*., 2008:151) or when total work during the intervention period was equated (Smart *et al*., 2011:205; Schjerve *et al*., 2008:283; Rognmo *et al*., 2004:219).

Anecdotal reports stated that the mechanisms responsible for an improvement in aerobic capacity with IT is that the heart is more maximally stressed (even if for short periods) compared to CAT allowing for more specific physiological adaptations. A few scientists reported evidence-based research proving that both central and peripheral adaptions were enhanced (Table 2.5)(Hwang *et al*., 2011:378; Tjønna *et al*., 2008:346; Wisløff *et al*., 2007:3086; Rognmo *et al*., 2004:219). The study by Wisløff and colleagues (2007:3086) showed that left ventricular remodelling (improved left ventricular ejection fraction and end diastolic and systolic volumes), stroke volume, endothelial functioning and quality of life were more improved in the IT program compared to a CAT program in their cohort of 75-year-old adults with heart failure. Their study showed that the aerobic capacity of those that followed IT improved with 46% whereas the CAT group improved by a mere 14%. Central adaptions to IT through an improved stroke volume were also supported by another study (Helgerud *et al*., 2007:665). With respect to peripheral adaptations, Tjønna *et al*. (2009:317) and Tjønna *et al*. (2008:346) stipulated that endothelial function increased more significantly in the IT group. Schjerve *et al*. (2008:282) postulated that the improvement in IT was enhanced by improved endothelial function and Ca^{2+} transport in the skeletal muscle. This finding was confirmed in a meta-analysis by Hwang *et al*. (2011:383). Tjønna *et al*. (2008:346) also found insulin signalling in fat and skeletal muscle, skeletal muscle biogenesis, excitation-contraction coupling and reduced blood glucose in adipose tissue more enhanced with IT compared to CAT. A review article by Boutcher (2010:176) stated that skeletal muscle adaptations resulted in enhanced

skeletal muscle fat oxidation and improved glucose tolerance. Talanian *et al*. (2007:1439) also found that fat oxidation increased by 36% with IT.

Table 2.5: Improvement in VO_2 peak and possible mechanisms for these improvements

Study	Mode	Improvement in VO_2 peak	Mechanisms that may improve VO_2 peak
Burgomaster *et al*., 2008	Cycling	6%$	Skeletal muscle oxidative capacity
Gibala *et al*., 2012	Cycling	Review	Oxidative enzymes, PGC-1α, central factors
Helgerud *et al*., 2007	Running	17% $	Stroke volume
Hwang *et al*., 2011	Running Cycling	22% *	Peak oxygen pulse, cardiac output, contractile capacity, rate of CA2+ uptake, capillary density
Ingul *et al*., 2010	Running	9% $	Improved left-ventricular ejection fraction
Moholdt *et al*., 2009	Running	10% *	None provided
Rognmo *et al*., 2004	Running	18%*	None provided
Smart *et al*., 2011	Cycling	21% #	No statistical significant improvements shown for endothelial function
Talanian *et al*., 2007	Cycling	13% $	Muscle oxidative enzyme capacities
Tjønna *et al*., 2009	Running/ Walking	10% *	Improved endothelial function; peak oxygen pulse
Tjønna *et al*., 2008	Running	26% *	Improved endothelial function; PGC-1α; CA2+ reuptake; stroke volume
Warburton *et al*., 2005	Running	10% $	None provided
Wisløff *et al*., 2007	Running	46% *	PGC-1α; improved endothelial function, CA2+ reuptake; pro BNP
Whyte *et al*., 2010	Cycling	9% $	Muscle oxidative enzymes

*: A more significant improvement compared to CAT; #: no significant improvement compared to CAT; $: only IT performed and not CAT.

Reports of an enhanced skeletal muscle fat oxidation are highly probable as Gaitanos *et al*. (1993:712) and Trump *et al*. (1996:1574) demonstrated that nearing the end of an IT workout, an inhibition of anaerobic glycolysis may occur. ATP resynthesis may be provided by PCr degradation and intracylglycerol stores. The researchers explained that this finding may occur despite the continued elevation of plasma epinephrine. Putman *et al*. (1995:458) also showed that by the third of five 30-second Wingate bouts, the majority of the ATP was generated by mitochondrial oxidation. There are also studies that took skeletal muscle biopsies after an intervention period (Gibala *et al*., 2009:929; Gibala *et al*., 2008:58). These studies found an increased activity and content of mitochondrial enzymes such as citrate synthase and cytochrome oxidase.

Other reasons for an improvement in aerobic capacity with IT may be related to an improvement in lower extremity muscle strength. Nemoto *et al*. (2007:803) demonstrated that concomitant increases in isometric knee strength (flexion and extension) with IT (and not with CAT) might have helped the participants in their study to obtain an improved aerobic capacity.

The anaerobic capacity after an IT training protocol has also been shown to improve. Studies reported improvements in maximal accumulated oxygen deficit, lactate dynamics and variables associated with the Wingate anaerobic test (Whyte *et al*., 2010:1421; Trapp *et al*., 2008:688; Burgomaster *et al*., 2006:2041; Tabata *et al*., 1997:390).

2.4.3 Effect of interval training on anthropometry

Most of the studies using interval training had remarkable success to significantly manipulate anthropometric variables (Heydari *et al*., 2012:3; Ingul *et al*., 2010:853; Shepherd *et al*., 2010:11; Whyte *et al*., 2010:1421; Tjonna *et al*., 2009:317; Tjonna *et al*., 2008:346; Trapp *et al*., 2008:684; Helgerud *et al*., 2007:665; Warburton *et al*., 2005:1080; Boudou *et al*., 2003:421; Mourier *et al*., 1997:385). From the evidence presented in Table 2.6 one can see that anthropometric variables can be significantly improved with many differing types of IT. Heydari *et al*. (2012:3) showed that 20-minute sessions, three times a week at 80%–90% of peak heart rate for 12 weeks elicited significant changes in body mass, BMI, waist circumference, fat mass, fat free mass, percentage of fat mass and also showed that the percentage of fat oxidation in grams per day improved after the intervention period. Ingul *et al*. (2010:853) performed a study with 4 x 4-minute intervals at 90% of maximum heart rate, twice a week, for 13 weeks and found that percentage of body fat as well as waist circumference decreased significantly. A study by Trapp *et al*. (2008:684) utilised a somewhat different IT group (8 seconds of sprinting with 12 seconds of slow pedalling repeated 60 times) that trained three times a week for 16 weeks whereas the other group exercised continuously at 60% of VO_2 peak. Only the IT group significantly lost body and fat mass.

In recent review articles by Pattyn *et al*. (2014:687) and Boutcher (2010:176), it is explained that IT is more effective in altering body composition indices compared to regular aerobic exercise. In fact, Boutcher (2010:176) controversially stipulated that the effect of regular aerobic exercise on body fat is negligible. They explained that the main reason why high intensity training results in significant weight loss remains largely unknown. Trapp *et al*. (2008:684) also found that the IT group lost more body mass and subcutaneous and abdominal fat compared to CAT. They explained that IT might have suppressed dietary intake more. They also hypothesised that IT might result in greater lipid oxidation compared to CAT. These researchers previously demonstrated that lipid release as demonstrated by plasma glycerol and catecholamine levels were significantly elevated after IT (Trapp *et al*., 2007:2370). They also explained that fat loss was variable and those with the highest baseline fat mass lost the most fat after the intervention period (Trapp *et al*., 2008:690). These findings are supported by Boudou *et al*. (2003:421) and Mourier *et al*. (1997:385) who also indicated that the greatest fat loss occurred in overweight individuals. Another study by Tjønna *et al*. (2009:324) supported these findings and reiterated that IT was more effective in obtaining a favourable body composition compared to CAT. They explained that although fat metabolism might be higher during moderate continuous training, the total fat oxidised after an IT session might be more. Also, evidence suggested that subsequent to a 2-minute high-intensity exercise, oxidation was supplied largely by fat fuels during the post exercise recovery (Medbø & Jebens, 2002:211). Even when training volumes were matched between IT and CAT, IT seemed to be more efficient in reducing body mass in adolescents and adults (Tjønna *et al*., 2008:346; Gutin *et al*., 2005:746; Gutin *et al*., 2002:818). However, there is a study that reported similar improvements in body mass loss after IT or CAT training (Schjerve *et al*., 2008:291).

Boutcher (2010:176) highlighted a few possible mechanisms for increased fat or body mass loss after IT even though the cause remains largely unknown. Firstly, IT has had discernible increases in body and skeletal muscle fatty acid oxidation. This finding has already been discussed (Walman *et al*., 2009:165; Trapp *et al*., 2007:2370; Medbø *et al*., 2002:211). Secondly, IT may inhibit appetite. High intensity exercise was shown to reduce food intake in rats by facilitating the release of corticotrophin (Bilski *et al*., 2009:82). Thirdly, increased fat oxidation after IT might ensue to remove excess lactate and H+ and to resynthesise glycogen. Lastly, there is some reason to believe the excess post-exercise oxygen consumption (EPOC) might be related to post-exercise fat metabolism (Boutcher, 2010:183).

Studies also showed a significant decrease in waist circumference after only two weeks of IT (Whyte *et al*., 2010:1421). It was also shown that individuals with metabolic syndrome can significantly ameliorate their waist circumference (mean loss of 6 cm) in both an IT and CAT group after 12 weeks of training (Tjonna *et al*., 2008:348). However, one year later these researchers showed that a mean loss of 6.8 cm was attained only after 12 months of IT in overweight adolescents (Tjonna *et al*., 2009:319). Findings of this nature are important as it was reported that waist circumference predicts obesity-related health risk and noninsulin-dependent diabetes mellitus more effectively compared to BMI and other

anthropometric indices (Janssen *et al*., 2004:379; Wei *et al*., 1997:16). However, BMI was also shown to decrease with both IT and CAT training (Tjønna *et al*., 2008:348).

Studies that have used combined IT and CAT also reported a 48% reduction in visceral fat as determined by an MRI scan (Mourier *et al*., 1997:385). Therefore, it may be more feasible to combine IT and CAT compared to IT alone. Another study compared the effects of two interval training protocols with the effect of CAT in a physically active population (Cicioni-Kolsky *et al*., 2013:304). They demonstrated that body mass loss was negligible in all three groups. No explanations were given as to why this was the case. The population studied was not overweight and were physically active. Perhaps the training intervention (6 weeks) was too short to induce significant changes on an already active and lean population group.

Table 2.6: Interval training and manipulation of anthropometric-associated variables

Study	n	Body mass (kg)	Waist circumference	Subcutaneous fat (kg)	Abdominal fat (kg)
Boudou *et al*., 2003	16	2 kg loss	Not recorded	18% loss	44% loss
Burgomaster *et al*., 2008	20	No change	Not recorded	Not recorded	Not recorded
Helgerud *et al*., 2007	40	1 kg loss	Not recorded	Not recorded	Not recorded
Helgerud *et al*., 2007	40	1.5 kg loss	Not recorded	Not recorded	Not recorded
Heydari *et al*., 2012	46	1.5 kg loss	3.5 cm loss	1.5% loss	1.5 kg loss
Ingul *et al*., 2010	10	No change	3.4 cm loss	25% loss	Not recorded
Moholdt *et al*., 2009	59	Not recorded	Not recorded	Not recorded	Not recorded
Mourier *et al*., 1997	24	1.5 kg loss	1 cm loss	18% loss	48% loss
Perry *et al*., 2008	25	0.2 kg loss	Not recorded	Not recorded	Not recorded
Rognmo *et al*., 2004	21	Not recorded	Not recorded	Not recorded	Not recorded
Shepherd *et al*., 2010	8	Not recorded	Not recorded	5% loss	Not recorded
Tjønna *et al*., 2008	32	2.3 kg loss	5 cm loss	Not recorded	Not recorded
Tjønna *et al*., 2009	62	No change	Not recorded	7% loss	8% loss
Trapp *et al*., 2008	45	1.5 kg loss	Not recorded	10% loss	10% loss
Tremblay *et al*., 1994	15	0.1 kg loss	Not recorded	15% loss	12% loss
Wallman *et al*., 2009	38	No change	Not recorded	0.3 kg loss	Not recorded
Warburton *et al*., 2005	14	3 kg loss	Not recorded	Not recorded	Not recorded
Whyte *et al*., 2010	10	1 kg loss	2.4 cm loss	Not recorded	Not recorded
Wisløff *et al*., 2007	27	Not recorded	Not recorded	Not recorded	Not recorded

n: sample size

2.4.4 Effect of interval training on functionality

Improvement in the quality of life remains a problem for many individuals, especially those with physical and intellectual disability, the obese and those with chronic clinical conditions. The study by Wisloff and colleagues (2007:3086) demonstrated that patients with heart failure (subsequent to a myocardial infarction) improved on the MacNew global score for quality of life in cardiovascular diseases after an interval training program of 12 weeks. The authors suggested that higher-intensity exercise is more rewarding (Wisloff *et al*., 2007:3093). The mechanisms for an improved quality of life in this functionally stricken population are currently unknown but improved physiological adaptations and subsequent functional ability may partly explain this phenomenon. Four weeks of IT also improved the quality of life in another clinical population (coronary artery bypass surgery) (Moholdt *et al*., 2009:1031). Moreover, the patients improved quality of life on the social, physical, and emotional domain and maintained this improvement for six months with home-based training subsequent to the intervention period (Moholdt, 2009:1036). The improvement was not only restricted to the IT group as the CAT also improved significantly. Nilsson *et al*. (2008:1361) performed a similar study on patients with chronic heart failure. After 16 weeks of interval training, functional capacity as measured with a 6-minute-walk distance test and a cycle ergometer test, increased by 13 and 16 % respectively. Quality of life as measured by the Minnesota questionnaire for patients with chronic heart failure also improved significantly (effect size equal to 0.7). Moreover the improvement in quality of life was related to the improvement in functional capacity. Improvements of this nature in a chronic heart failure population with a mean age of 70 years are clinically significant. Another study hypothesised (due to various physiological improvements after 16 weeks of IT) that activities of daily living will be performed with greater ease and less time in patients with coronary artery disease (Warburton *et al*., 2005:1084). There is also a study that showed that IT is more significant to increase functional capacity compared to CAT in a chronic heart failure population (Smart *et al*., 2011:205). Quality of life as measured by the Hare-Davis Cardiac Scale improved in the IT group only (Smart *et al*., 2011:209).

2.4.5 Effect of interval training on metabolic markers

Interval training was shown to improve various metabolic risk factors for cardiovascular disease (diabetes, insulin sensitivity, blood glucose, blood pressure, and lipids) in different chronic clinical or overweight populations (Hood *et al*., 2011:1849; Moholdt *et al*., 2009:1034; Tjønna *et al*., 2009:317; Wallman *et al*., 2009:167; Tjønna *et al*., 2008:348). In some cases IT showed superior improvements compared to CAT.

2.4.5.1 Insulin sensitivity and diabetes

Most studies that conducted interval training on their study populations found an improvement in insulin sensitivity of between 22% and 57% (Whyte *et al*., 2010:1421; Tjønna *et al*., 2009:320; Tjønna *et al*., 2008:348; Boudou *et al*., 2003:421; Mourier *et al*., 1997:385). The significant improvements can likely be explained by skeletal muscle adaptations allowing for a greater capacity of fatty acid oxidation and glycolytic enzyme content (Boutcher, 2010:176). Also, IT consisting of repeated high-intensity exercise is more effective in triggering insulin activity than CAT in people with metabolic syndrome. Therefore, the high intensity of exercise may explain the improved fasting glucose tolerance (Moholdt *et al*., 2009:1034; Tjønna *et al*., 2008:346). Hwang *et al*. (2011:378) reiterated this finding when analysing IT compared to CAT. Tjonna *et al*. (2009:324) also reported lower glucose values after three months of IT (compared to a multi-treatment group) in obese individuals without diabetes and stated that it was consistent with a lower intake of carbohydrates. The participants' improved dietary habits were motivated by an improved aerobic capacity and anthropometry. Also, GLUT-4 increases were associated with an increased insulin sensitivity which also contributed to attenuated blood glucose (Host *et al*., 1998:798). Tjønna *et al*. (2008:348) similarly reported that insulin sensitivity and high-density lipoprotein increased more significantly in the IT compared to the CAT group. The increased insulin sensitivity post exercise was proposed to result from peripherally enhanced insulin response and signalling (Koval *et al*., 1999:998). Whyte and colleagues (2010:1421) showed that high-intensity interval training significantly improved various metabolic factors in overweight and sedentary men. The significant improvements were studied after a mere two weeks. Similarly, Hood *et al*. (2011:1849) demonstrated that IT might reduce the risks of many inactivity-related disorders. Muscle oxidative capacity, glucose transporter protein content and insulin sensitivity significantly improved after only two weeks of training. In fact, a study by Shepherd *et al*. (2010:11) revealed that insulin sensitivity improved by 27% after six weeks of IT. Tjønna *et al*. (2008:346) demonstrated that more risk factors were eliminated in those suffering from metabolic syndrome with IT compared to continuous training. Specifically endothelial function and insulin signalling in fat and skeletal muscle were improved. A greater reduction in blood glucose and lipogenesis in adipose tissue were noted.

IT was also shown to be a useful intervention strategy in those suffering from Type 2 diabetes (Little *et al*., 2011:1554). Glucose control was rapidly improved after only two weeks of IT (24 hour continuous monitoring) associated with improvements in various markers of mitochondrial capacity. The improved metabolic health was associated with the adaptations that ensued in the skeletal muscle. A study by Tjønna *et al*. (2009:317) similarly revealed that three months of IT compared to three months of multi-treatment (three activity sessions, dietary and psychological advice) triggered more favourable glucose and insulin control. These results were maintained 12 months after the initiation of the program as the participants were asked to continue their twice-weekly IT or multi-treatment sessions. A study that used a combined interval and resistance training program found significant improvements in mean arterial

blood pressure, fasting plasma glucose and non-esterified fatty acids in deconditioned Type 2 diabetic patients (Praet *et al*., 2008:163). Contra-indicatively, Babraj *et al*. (2009:1) did not find improvements in fasting plasma glucose values but the area under the plasma glucose and insulin curves were all reduced. This finding transpired up until three days after the last exercise session. The authors concluded that insulin action improved substantially in these individuals and could be used to minimise metabolic risk factors. Similarly, Shepherd *et al*. (2010:11) showed that the area under the plasma glucose and insulin curve improved significantly with 17% and 31% respectively pre- and post-intervention after a glucose tolerance test. Overall, a review article by Boutcher (2010:176) indicated that IT was effective in lowering insulin resistance due to a number of skeletal muscle adaptations that ultimately resulted in increased glucose tolerance.

2.4.5.2 Blood pressure

Elevated blood pressure is also indicated to respond positively to exercise interventions. The current evidence in the general population indicate that exercise may result in a hypotensive effect for up to 22-hours post-exercise. Tjønna *et al*. (2008:348) reported that both IT and CAT were equally effective in reducing mean arterial and systolic blood pressure after 16 weeks of training in patients with metabolic syndrome. Both training groups reduced their resting systolic blood pressure by a mean value of 8 mmHg. Improvements of this magnitude approximates to a 35% decrease in premature deaths resulting from stroke and ischemic heart disease (Lewington, 2003:1060). Guimaraes *et al*. (2010:627) also found that 16 weeks of either IT or continuous training in a group of hypertensive subjects elicited similar benefits in reducing ambulatory blood pressure, but that arterial stiffness was improved in the IT group only. Ciolac *et al*. (2011:824) performed an interesting study where they assessed the effect of IT and CAT on non-hypertensive offspring to hypertensive parents (high familial risk for hypertension). Both training groups, but IT to a more significant effect, improved various physiological markers of cardiorespiratory fitness. Their study provided guidelines for designing exercise programs for the prevention of an inherited hypertensive disorder. Nemoto *et al*. (2007:803) also presented findings that high intensity walking intervals elicited more benefits than a continuous program in many parameters including a greater reduction in resting systolic blood pressure. On the other hand, Wallman *et al*. (2009:166) demonstrated no change in resting systolic and diastolic blood pressure after IT and explained that it was probably due to the mean normotensive values in the sample cohort (125/80 mmHg).

To summarise, the research indicate that most studies demonstrated a significant improvement in blood pressure after IT or CAT. A few studies showed that IT had a larger effect on the blood pressure response after training but there were also studies that showed no change after either training protocols (Wallman *et al*., 2009:166; Nemoto *et al*., 2007:803; Rognmo *et al*., 2004:219). A meta-analysis, however,

reported that IT did not have a significantly superior effect on resting blood pressure after an intervention compared to CAT in the general population (Hwang *et al*., 2011:383).

2.4.5.3 Lipids

Musa *et al*. (2009:587) showed that eight weeks of IT favourably influenced high-density lipoprotein in 36 healthy untrained young men. However, no change occurred in total cholesterol. Another study in patients with metabolic syndrome also reported improvements in HDL cholesterol with IT but not in a multi-treatment group that performed CAT and received dietary and psychological advice. Although a study by Stein *et al*. (1990:277) did not include IT in their different intervention groups, it was shown that a higher training intensity (more than 75% of maximal heart rate) was needed to significantly improve high density lipoprotein cholesterol after 12 weeks. A more recent study demonstrated no improvements in total cholesterol, HDL cholesterol, LDL cholesterol or triglycerides after a combined IT and diet program (Wallman *et al*., 2009:167). The authors explained that although exercise and diet were the main triggering mechanisms to ameliorate cholesterol-associated variables, the current study's baseline values were well within the normal range. Other studies in a clinical population showed improvements regarding triglycerides, HDL and LDL and cholesterol but they were not significant in participants with heart failure and those who received coronary artery bypass surgery (Moholdt *et al*., 2009:1031; Wisløff *et al*., 2007:3088). No explanations were given for this finding. We are not aware of any studies that have shown improvements with LDL cholesterol in association with IT. In fact, CAT and not IT was shown to improve LDL cholesterol in obese adults after 12 weeks of training (Schjerve *et al*., 2008:283). Future studies should further explore the effect of IT on this measure and provide possible mechanisms as to why CAT may have a more protective effect.

2.4.6 Interval training in an intellectually disabled or Down syndrome population

Despite IT showing many physiological, functional, metabolic, health and anthropometric benefits, it has also served as a motivational and time-saving aid. Participants have reported more enjoyment and greater variability with its use. This has also helped researchers and conditioning coaches with issues pertaining to motivation, commitment and ultimately program adherence.

Results on an interval-based exercise intervention in a population of ID or DS individuals could not be found in the published literature. It is not known whether physical, metabolic and anthropometric benefits will be reported as routinely studied in those with chronic medical conditions. This is important since a large percentage of individuals with ID and DS are overweight or obese (Terblanche & Boer, 2013:830; Pitetti *et al*., 2013:47; Melville *et al*., 2008:425; Salaun & Berthouze-Aranda, 2011:333; de Winter *et al*., 2012a:1722; Rubin *et al*., 1998:175). Adults with ID and DS are also at risk of developing diabetes, cardiovascular disease, high blood pressure, metabolic syndrome and high total cholesterol (de

Winter *et al*., 2012a:1726; Jeevendam *et al*., 2009:462; Hermon *et al*., 2001:167). Obese individuals with ID and DS are more prone to develop secondary conditions such as diabetes, hypertension, cholesterol and depression compared to healthy ID individuals (Rimmer *et al*., 2010:787; Draheim *et al*., 2002b:441). Also, individuals with ID generally have a poor aerobic capacity with DS individuals having an even poorer VO_2 peak compared to their ID counterparts without DS (Baynard *et al*., 2008:1984; Guerra *et al*., 2003:1604). Most CAT programs have elicited marginal improvements in anthropometric and aerobic capacity in an ID and DS population. The adaptations needed for a greater VO_2 peak or change in anthropometry may require a longer training period and/or a higher training intensity in a DS and ID group. That is why IT may be specifically advantageous to this population group as its superior anthropometric, metabolic and physical benefits and associated mechanisms (compared to CAT) have been reported not only in the general population but also in those with chronic medical conditions. Moreover, the poor functional fitness of those with chronic medical conditions mimic many of those limitations reported in the ID and DS population.

2.5 Summary

Research in the field of exercise in persons with intellectual disabilities is limited, particularly those with DS. In the aforementioned chapter it is reported that individuals with ID and DS suffer from many health related conditions, live sedentary lives, are overweight, and have poor functional and physical abilities. These factors, together with an increased life expectancy may hamper the quality of life and independence of these individuals as they age. The advantages of regular exercise in the general population of persons without ID or DS, is well described and interventions strategies well documented. Appropriate standardised testing and intervention options are necessary to pro-actively prevent age associated diseases, conditions and physical disabilities. Future studies should aim to standardise functional fitness testing in order to improve assessment of functional fitness and to determine the effect of exercise interventions on functional fitness. The benefits of high intensity interval training in persons with no ID, has been evidenced, further studies should determine if these exercise strategies are also beneficial for persons with ID and DS. The benefits of filling the gaps in the current body of knowledge as stated above, will inform health educators, exercise professionals and caregivers of the benefits that new approaches to exercise interventions can bring to special populations.

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CHAPTER THREE

Test-retest reliability and minimal detectable change scores of twelve functional fitness tests in adults with Down syndrome

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Abstract

Aim: The purpose of the study was to explore the test-retest reliability and minimal detectable change of selected functional fitness test items in adults with Down syndrome. **Methods:** Forty-three adults with Down syndrome (24 men and 19 women) aged 18–50 years completed a battery of tests twice in a two-week period. The battery of tests consisted of two balance items, two flexibility items, five muscular strength and endurance items, two aerobic items, and one functional task. All items were considered valid and reliable tests in a general elderly or intellectually disabled population. The test-retest relative reliability for all repeated tests was assessed with intraclass correlation coefficient performing one-way analysis of variance. The test-retest absolute variability was measured by using the standard error of measurement (SEM) to calculate the minimal detectable change at the 90% confidence interval (MDC_{90}). Reliability data was visualised with a Bland-Altman plot. **Results:** All tests showed excellent intraclass correlation coefficients (ICC's>0.9). All SEM values demonstrated acceptable measurement precision (SEM<SD/2). Values for MDC_{90} are provided for all 12 tests. The analyses indicated that there was no major systematic bias in the plots. The scatter around the Bland-Altman was distributed randomly **Conclusion:** All twelve functional fitness tests demonstrated adequate feasibility and relative and absolute test-retest reliability in adults with Down syndrome in South Africa. MDC scores at 90% confidence interval for these tests will help determine whether the change measured in future experimental studies are due to error or treatment. Information of this nature will help to monitor performance alterations over time and success of training interventions.

Highlights

1: Test-retest reliability of all 12 functional fitness tests in adults with Down syndrome is excellent.

2: Standard Error of Mean values demonstrated acceptable measurement precision.

3: Minimal Detectable Change scores at 90% confidence interval for these tests are provided.

4: All tests are user-friendly, easy to perform and field-based.

1. Introduction

Down syndrome (DS) occurs when there is a full or partial extra copy of the 21st chromosome. DS is the most prevalent chromosomal cause of intellectual disability (ID) (National Down Syndrome Society (NDSS), 2014; Barnhart & Connolly, 2007). Individuals with DS are born with many health-related disorders, of which congenital heart disease (61% of all cases) is the most common (Abbag, 2006). These individuals have a greater risk of developing thyroid problems, leukaemia, epilepsy, diabetes and Alzheimer's disease (NDSS, 2014; Hermon, Alberman, Beral, & Swerdlow, 2001). Nowadays, most of these conditions are medically treatable, which explains the increase in life expectancy from 25 years (20 years ago) to 60 years (NDSS, 2014; Torr, Strydom, Patti, & Jokinen, 2010; Chicoine & McGuire, 1997).

Individuals with DS have sedentary lifestyles (Nordstrøm, Hansen, Paus, & Kolset, 2013; Esposito, MacDonald, Hornyak, & Ulrich, 2012; Shields, Dodd, & Abblitt, 2009) and the majority of these individuals are overweight or obese (Terblanche & Boer, 2013; Rubin, Rimmer, Chicoine, Braddock, & McGuire, 1998). Moreover, the aerobic capacity, muscular strength and functional capacity of individuals with DS are poor, not only when compared to the general population but also to individuals with ID without DS (Baynard, Pitetti, Guerra, Unnithan, & Fernhall, 2008; Baynard, Pitetti, Guerra, & Fernhall, 2004; Carmeli, Kessel, Merrick, & Bar-Chad, 2004; Carmeli, Barchad, Lenger, & Coleman, 2002; Croce, Pitetti, Horvat, & Miller, 1996; Fernhall et al., 1996; Pitetti, Climstein, Mays, & Barrett, 1992). Poor physical fitness and functional capacity are factors that further predisposes this population to the early development of serious health problems (González-Agüero, Vicente-Rodriguez, Moreno, Guerra-Balic, Ara, & Casajus, 2010; Torr et al., 2010; Carmeli et al., 2004). Furthermore, individuals with DS are known to age prematurely, characterised not only by a lower life expectancy compared to the general population but also by plummeting functional fitness and weight values at an significantly earlier age (Terblanche & Boer, 2013; Torr et al., 2010; Carmeli et al., 2004; Rubin et al., 1998).

The undesirable combination of an improved life expectancy and premature functional limitations in a population with Down syndrome could possibly impact on the quality of life and independence, especially in old age. Fortunately, outcomes of this nature can be prevented or reversed through structured exercise training as shown in a DS population (Mendonca, Pereira, & Fernhall, 2011; Cowley et al., 2010; Shields, Taylor, & Dodd 2008; Rimmer, Heller, Wang, & Valerio, 2004). A description of the functional fitness capacities of individuals with DS is essential in order to optimise program prescription for improved functional capacity. Therefore, a specified scientific exercise program prescription should be based on a reliable, valid and specific measuring instrument for the population group studied. Rikli and Jones (2013) and Hilgenkamp, van Wijck, and Evenhuis (2012) developed specific instruments in the general elderly and intellectually disabled populations respectively. However, due to their discernible functional fitness impairments, adults with DS should not be pooled with

populations with general ID (Winnick & Short, 2014; Baynard et al., 2008). In fact, Winnick and Short (2014) acknowledged that an instrument they developed and validated made no distinction between intellectually disabled children and adolescents with and without DS, despite evidence that the presence of DS negatively affects fitness test performance. In addition, many adults with DS struggled to perform some of the tests and it was recommended that individuals with DS have their own functional fitness battery suited to their unique needs.

Using the information provided by Rikli and Jones (2013), Hilgenkamp et al. (2012) and the American College of Sports Medicine (2005), we proposed five parameters to describe functional fitness (balance, flexibility, functional ability, muscular strength and endurance, cardiovascular endurance). A thorough literature search was conducted to identify all possible measuring instruments based on their reliability and validity (especially tests used in an elderly or intellectually disabled population). A three-month pilot study ensued, after which the test choice was further refined with the help of experts in the field of DS research, physiotherapy, occupational therapy and disability sport. The pilot study was very significant in the pre-test planning for evaluating factors such as familiarisation with tests, development of test procedures and directions, preparation of participants, number of trials, obtaining the feel for equipment and space needed, preparation of worksheets, equipment checklists, and time needed to perform tests. Final test items were selected from the Bruininks-Oseretsky Test of Motor Proficiency (BOTMP), Brockport Physical Fitness Test (BPFT) and Senior Fitness Test (SFT) manuals. The feasibility of these tests in adults with DS has already been shown to be excellent in a large epidemiological study (n=371) (Terblanche & Boer, 2013). However, before these tests can be used in adults with DS, further psychometric analyses are needed. It is imperative to establish population-specific reliability coefficients for adults with DS as has been established in the general, general elderly and ID population before embarking on epidemiological or experimental research studies (Rikli & Jones, 2013; Winninck & Short, 2014; Hilgenkamp et al., 2012; Welk & Meredith, 2008). Therefore, the primary aim of the study was to explore the test-retest reliability and minimal detectable change observed of selected functional fitness test items in an exclusively DS adult population.

2. Materials and methods

2.1. *Participants*

Forty-three adults with DS (24 men and 19 women) were recruited from three care centres for persons with intellectual disabilities (Huis Amelia, Uitkomsversorgd, Die Oord) in two provinces of South Africa. The study protocol was approved by the Health Research Ethics Committee (Humans) of the North-West University (NWU-00064-14-A1). The parent or legal representatives signed an informed consent form while the participant signed a consent form that was adapted for persons with intellectual disability. Professional caregivers working at the facility completed the health questionnaire (adapted Physical Activity Readiness Questionnaire). The questionnaire consists of seven questions regarding the

health of the participant. If participants answered "yes" to any of the questions, a medical practitioner was contacted for further scrutiny. Individuals were included if they fulfilled the following inclusion criteria: Down syndrome, aged between 18 and 45 years, had the cognitive ability to understand and perform the exercises in a technically correct manner, if they provided written informed consent and lastly if they successfully completed the health questionnaire. Individuals were excluded if they suffered from congestive heart disease or any other physical, mental or medical condition that was considered a contraindication for study participation. All tests were performed by the participants twice in a two-week period with a minimum of 10 days apart.

2.2. *Procedures*

All testing procedures and methods, including technique and number of trials and demonstrations, were followed exactly as outlined in the Bruininks-Oseretsky, BPFT and the SFT manuals. All tests allowed two familiarisation and practice sessions one week prior to testing. Sufficient rest was allocated during testing days. Five adults were tested simultaneously on a turn basis (i.e. participant 1 – trial 1, participant 2 – trial 1 etc.), which also provided sufficient rest between tests.

The battery of tests consisted of two balance items, two flexibility items, five muscular strength and endurance items, two aerobic items and one functional task. All testing was conducted at the centre in a large, adequately ventilated indoor venue with non-slippery flooring, free from noise or disturbances. Participants were asked to avoid alcohol or caffeine intake and unusual or intense activity 24 hours before the testing day. Five participants were tested at a time and each session lasted approximately three hours. Participants were given a 5–8-minute warm up with stretching activities under the supervision of the researcher. These activities were not tiring and involved warming up the large muscle groups (marching in place, swinging arms) and simple stretches (head turn, single arm crossover, chest stretch, calf stretch, hamstring stretch). Participants were verbally instructed and visually shown how to perform each test. Participants were tested individually for all test items. Constant motivation and encouragement were given to all participants, as described by Varela, Sardinha, & Pitetti (2001) and Waninge, Evenhuis, van Wijck, and van der Schans (2011). Encouragement was given every 15 seconds by using standardised phrases (Nasuti, Stuart-Hill, & Temple, 2013). To minimise the effects of fatigue, stations were arranged in the following order: flexibility items, 8-foot up-and-go, balance items, muscular strength and endurance (modified curl up, trunk lift, chair stand, handgrip strength), and finally the 6-minute walk distance test (6MWD). These tests were done in the same order (so that the same muscle groups were not involved in back-to-back tests), starting anywhere in the circuit, except for the 6MWD which was always performed last (after a 10-minute break). Body mass and stature were always measured first. The 16-metre shuttle run test was performed the following day. All testing was done by one researcher with specific emphasis on scoring either successful measurement with a score, or an unsuccessful measurement without any score, to avoid uncertainty. Participants performed the twelve

tests on the first and second day (session 1), and again 10 to 14 days later (session 2). The tests were performed on the same time of day.

2.3. *Test items*

The items chosen are all considered valid and reliable (BOTMP, 2005; SFT, 2013; BPFT, 2014) and are easy to administer in their respective study populations. The BOTMP provides a complete guide to measuring the gross and fine motor skills of children and adolescents. Wuang, Chang, Wang, and Lin (2013) provided evidence that this battery also has adequate reliability for adolescents with intellectual disability. The BPFT provided a complete package (27 test items) for fitness testing of youths with physical and mental disabilities. The SFT (Rikli & Jones, 2013) was performed on elderly participants 60–94 years of age, and contained seven items. Reliability was established separately for each test item in these test batteries.

2.3.1. *Body mass and stature*

Body mass was determined with a calibrated electronic scale (Beurer, Ulm, Germany) and recorded to the nearest 0.1 kg. Participants were barefoot and clothed in lightweight clothing. Stature was measured using a sliding steel stadiometer (Siber-Hegner GPM, Switzerland). The participants were barefoot with heels together and upper back, buttocks and heels against the wall. The head was positioned in the Frankfurt plane which is achieved by positioning the lower edge of the eye socket (orbital) in the same horizontal plane as the notch just above the tragus of the ear (region). The measurement is then taken from the inferior aspect of the feet to the vertex of the skull (the highest point on the skull) to the nearest 0.1 cm. The height and weight was used to calculate body mass index (BMI) in kg/m^2.

2.3.2. *Balance (BOTMP)*

Static balance was assessed with the participants standing on one leg for as long as they could to a (maximum of 10 s), while looking straight ahead and with their hands on their hips. The knee of the free leg was bent so the lower leg was parallel to the floor. The test was terminated once the hands moved off the hips and/or if too much body sway occurred. The test was performed with both legs and the best score of each leg was noted as static balance performance. Test-retest reliability for the balance subtest tests (BOTMP) was determined by Wuang & Su, (2009:847) in adolescents with intellectual disability with an intraclass correlation coefficient (ICC) value of 0.99. Construct validity has been determined by Lin, Hwang, Hu, Wu, Wang, & Huang (2004:1346) in elderly individuals. Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

Dynamic balance was assessed by walking on a 3.05 m balance beam that is 10.16 cm wide. Participants were instructed to walk with a normal stride, while maintaining hands on the hips. The number of consecutive steps completed on the balance beam, up to a maximum of six steps, was recorded. In both

tests, two trials were administered and the best score noted. Test-retest reliability for the balance subtest tests (BOTMP) was determined by Wuang et al. (2009:847) in adolescents with intellectual disability with an ICC value of 0.99. Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

2.3.3. *Flexibility*

The sit-and-reach test assesses lower body flexibility. Each leg was extended straight out in front of the hip, with the heel on the floor and the ankle flexed at 90 degrees (the other leg is bent off to the side with the foot flat on the floor). With the hands overlapped and the middle fingers even, the participant reached as far as possible to the toes. If the tip of the middle finger did not touch the toe, the distance short of the middle toe was measured and recorded as a negative score while a middle finger reached beyond the toes, the distance of overlap was measured and recorded as a positive score. The same procedure is followed with the other leg. Test-retest reliability of this test item revealed an ICC value of 0.95 in an elderly population (Rikli & Jones, 2013:37). Criterion-related validity with goniometer-measured hamstring flexibility has been estimated for elderly men (r=0.76) and elderly women (r=0.81) (Rikli & Jones 2013:32). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

The shoulder stretch assesses upper body flexibility. Participants attempted to touch the fingertips of their two hands behind their back. The participant reached with his/her right hand in external rotation over the right shoulder between the scapulae, while the left elbow was bent and internally rotated and reached upwards from the waist. If the middle fingers of the two hands did not touch, the distance was measured and recorded as a negative score. If the middle fingers overlapped, the distance of overlap was recorded as a positive score. The test was performed on the left and right side. In both tests, two trials were performed and the best score was noted. Test-retest reliability of this test item revealed ICC values ranging from 0.84 to 0.94 in adolescents with ID measured two weeks apart (Winnick & Short 1998:17). Test-retest reliability of this test item revealed an ICC value of 0.96 in the general elderly population (Rikli & Jones, 2013:37). In the BPFT for individuals with physical and intellectual disabilities, the shoulder stretch is shown to demonstrate logical validity (Winnick & Short, 2014:16). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

2.3.4. *Muscular strength and endurance*

Lower body strength was assessed with the chair stand test (Rikli & Jones, 2013:144). Participants sat on a straight-backed chair (43.18 cm in height and with no arm rests), feet flat on the floor and arms across the chest. On the signal go, the participant rose to a full stand, and then returned to a fully seated position. The score was the number of stands completed in 30 s (two trials). In the general older population, test-retest reliability was good (ICCs of 0.84 for men and 0.92 for women) (Rikli & Jones,

2013:37) and criterion-related variability as a measure of lower body strength was confirmed (r = 0.78 for men and 0.71 for women) (Jones, Rikli, & Beam, 1999:113). Test-retest reliability in older adults with ID was moderate (ICC = 0.72 for same-day interval and 0.65 for two-week interval) (Hilgenkamp et al., 2012:160). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

Handgrip strength was assessed by a grip dynamometer (Takei, Grip D,T.K.K 5401; Niigata City, Japan) with a grip space of 10 cm. Participants sat on a straight-backed chair without arms, feet flat on the floor. The elbow was flexed at 90° and the grip dynamometer was squeezed as hard as possible. Three trials were administered, with 30 s rest in between each trial. Both hands were tested as many adults with DS did not know which was their stronger hand. The device digitally recorded the participant's test score (kg). Test-retest reliability in the general elderly population was good with an ICC of 0.99 (Abizanda, Navarro, Garcia-Tomas, Lopez-Jimenez, Martinez-Sanchez, & Paterna, 2012:24). Test-retest reliability of older adults with ID was good (ICC = 0.94 for same-day interval and 0.90 for two-week interval) (Hilgenkamp et al., 2012:158). Construct validity has been shown to be significant to various functional tests in elderly individuals (Abizanda et al., 2012:23-25). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

Abdominal strength was assessed with the modified curl-up test. Participants lay in a supine position with knees bent and feet flat on the floor, hands on thighs. During the curl-up, participants slid their hands up the thighs to the kneecap and then returned to the starting position. The fingers had to slide at least 10 cm along the legs to the kneecaps. The researcher's hands were placed on the superior aspect of the kneecap, thereby assisting the subject in performing the correct technique. Fingers were not allowed to lift off the legs and the hands had to slide up simultaneously to the left and right kneecap respectively. Participants performed as many curl-ups as possible (up to a maximum of 75) for as long as possible by doing one curl-up every 3 s. The researcher verbally counted the number of curl-ups. Only one trial was administered. Test-retest reliability in adolescents with intellectual disability was adequate (ICC = 0.82) measured two weeks apart (Winnick & Short, 1998:16). This test has been shown to have logical validity (Jette, Sidney, & Cicutti, 1984:4). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

The trunk lift test assesses trunk strength. From a prone position with hands under the thighs, participants attempted to lift their chins up to a maximum height from the mat by arching the back. The measurement was taken with a tape measure from the mat to the bottom of the chin (lower jaw). Two trials were allowed and the best score was noted. Test-retest reliability in adolescents with intellectual disability was adequate (ICC = 0.89) measured two weeks apart (Winnick & Short, 1998:16). This test has been shown to have logical validity (Winnick & Short, 2014:16). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

The isometric push-up assesses upper body strength. Participants attempted to hold the push-up position for as long as they could. Hands had to be placed directly below the shoulders, arms had to be extended, the back had to be perfectly aligned with the rest of the body, and toes had to be on the floor. The time that the position was kept was taken to the nearest second. Only one trial was administered. Time was stopped as soon as the back sagged or lifted. Proper form was strictly controlled. Test-retest reliability measured two weeks apart in adolescents with ID was good (ICC = 0.98) (Winnick & Short, 1998:15). In the BPFT for individuals with physical and intellectual disabilities the isometric push-up is shown to demonstrate logical validity (Winnick & Short, 2014:17) Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

2.3.5. *Aerobic fitness*

Cardiorespiratory endurance was assessed by means of a 16-m modified shuttle-run test. The 16-m PACER test can be performed indoors with limited space, is fun, does not require self-monitored pacing, and incurs fewer motivational problems (Winnick & Short, 2014). At the sound of a tape-recorded beep, participants ran from one line to the other, 16 m away. The test score was the number of laps completed on pace and only one trial was given. It has been shown that the 16-m shuttle-run test has adequate test-retest reliability (ICC = 0.98) measured one week apart in children and adolescents with intellectual disability (Winnick & Short, 1998:15). Criterion-related validity has been shown with VO_2 peak in individuals with intellectual disability (Fernhall et al., 1996:602). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

The participants completed a standardised, self-paced 6-minute walk test. They were instructed to walk as many laps as possible within the allocated six minutes. The participants were allowed to rest at any time, but were encouraged to continue as soon as possible. The distance covered was recorded to the nearest metre. A 1:1 pacer was provided, as has been described in adults with ID by Nasuti et al. (2013). This test assesses aerobic endurance and functional ability important for many everyday living activities such as walking distances, shopping and sightseeing whilst on vacation etc. (Nasuti et al, 2013; Rikli & Jones, 2013). Test-retest reliability of this test item revealed an ICC value of 0.94 in the general elderly population (Rikli & Jones, 2013:37) and an ICC value of 0.98 in adults with ID (Nasuti et al., 2013:31). In adolescents with DS, this test item demonstrated good test-retest reliability (Casey et al., 2012:2068). Criterion-related validity has been shown with treadmill performance using the modified Balke protocol (Rikli & Jones, 1998:363) and to VO_2 peak (R=0.84) in adults with ID (Nasuti et al, 2013). However, the test did not show discriminant validity in adults with DS with or without cardiac restriction (Vis et al., 2009:1423).

2.3.6. *Functional test*

The 8-foot get-up-and-go test assessed the ability to perform an everyday functional activity. On the signal 'go' the participant got up from the chair, walked as quickly as possible to a cone placed 2.43 m away, walked around the cone and returned to the chair. After one practice trial, two test trials were administered and the best time recorded in seconds. Test-retest reliability of this test item revealed an ICC value of 0.95 in an elderly population (Rikli & Jones, 2013:37). There is no gold standard criterion to measure against, but it has been found to be significantly related to other functional tests in the elderly population (Podsiadlo & Richardson, 1991:142). Discriminant validity has been reported in adults with DS between age categories (18–25, 26–35, 36–45 and >45) (Terblanche & Boer, 2013:833).

2.4. *Statistical analysis*

All data was reported using a commercially available software package (SPSS, Version 20.0, Inc., Chicago, IL, USA). Descriptive statistics were expressed as mean and standard deviations (SD). Normality and homoscedasticity of data were assessed with the Kolmogorov-Smirnov and Leven tests respectively. Feasibility of tests was expressed as a percentage completion rates. Differences between test and retest were analysed by a paired t-test or Wilcoxon signed-rank test for continuous data. The test-retest relative reliability of data for all repeated tests was assessed with the ICC by using a one-way analysis of variance (ANOVA). This model treats all sources of measurement variation as error, providing an accurate estimate of stability reliability. The test-retest absolute variability was measured by using the standard error of the mean (SEM) to calculate the minimal detectable change at the 90% confidence interval (MDC_{90}). The following formulas were used (Stratford, 2004): $SEM = SD \times \sqrt{(1-r)}$; $MDC_{90} = SEM \times 1.65 \times \sqrt{2}$. Test-retest reliability was calculated between session 1 and session 2 of each test. ICC values of 0.6 or higher were considered acceptable and ICCs of 0.9 or higher desirable. Reliability data was also visualised with a Bland-Altman plot (Bland & Altman, 1986). The difference between the two tests is plotted against the mean of the two tests for each participant. A p value <0.05 will be considered statistically significant.

3. Results

Demographic variables such as age, body mass, stature, BMI and gender ratio are shown in Table 1.

Table 1
Demographic variables of adults with Down syndrome (n=43)

Variable	**Mean (SD)**	**Range**	**Standard error mean**
Gender (Male/Female)	24/19	N/A	N/A
Age (yrs)	33.6 (8.6)	31.0	1.32
Body mass (kg)	72.0 (10.9)	53.9	1.67
Height (cm)	154.3 (7.6)	29.7	1.16
BMI (kg/m^2)	30.3 (4.9)	20.5	0.74

Abbreviations: HGS, Handgrip strength, cm, centimetre; kg, kilogram; m, metre; yrs, years

3.1. *Feasibility*

Most of the adults with DS in the current study completed all 12 functional fitness tests (n=43). Only two adults with DS failed to complete one test each. One adult was not able to perform the shoulder flexibility test due to elbow pain and another adult with DS had difficulty in performing the isometric push-up as he felt shoulder pain.

3.2. *Test-retest reliability*

No significant differences were obtained between test and retest for all 12 functional fitness tests ($p<0.05$). The mean scores and standard deviations of all tests per session and associated intraclass correlation coefficient are demonstrated in Table 2. All tests showed excellent intraclass correlation coefficients ($r>0.9$). All SEM values attained the criterion (SEM < SD/2), suggesting an acceptable measurement precision (Table 2). Values of minimal detectable change at the 90% confidence interval are also shown in Table 2.

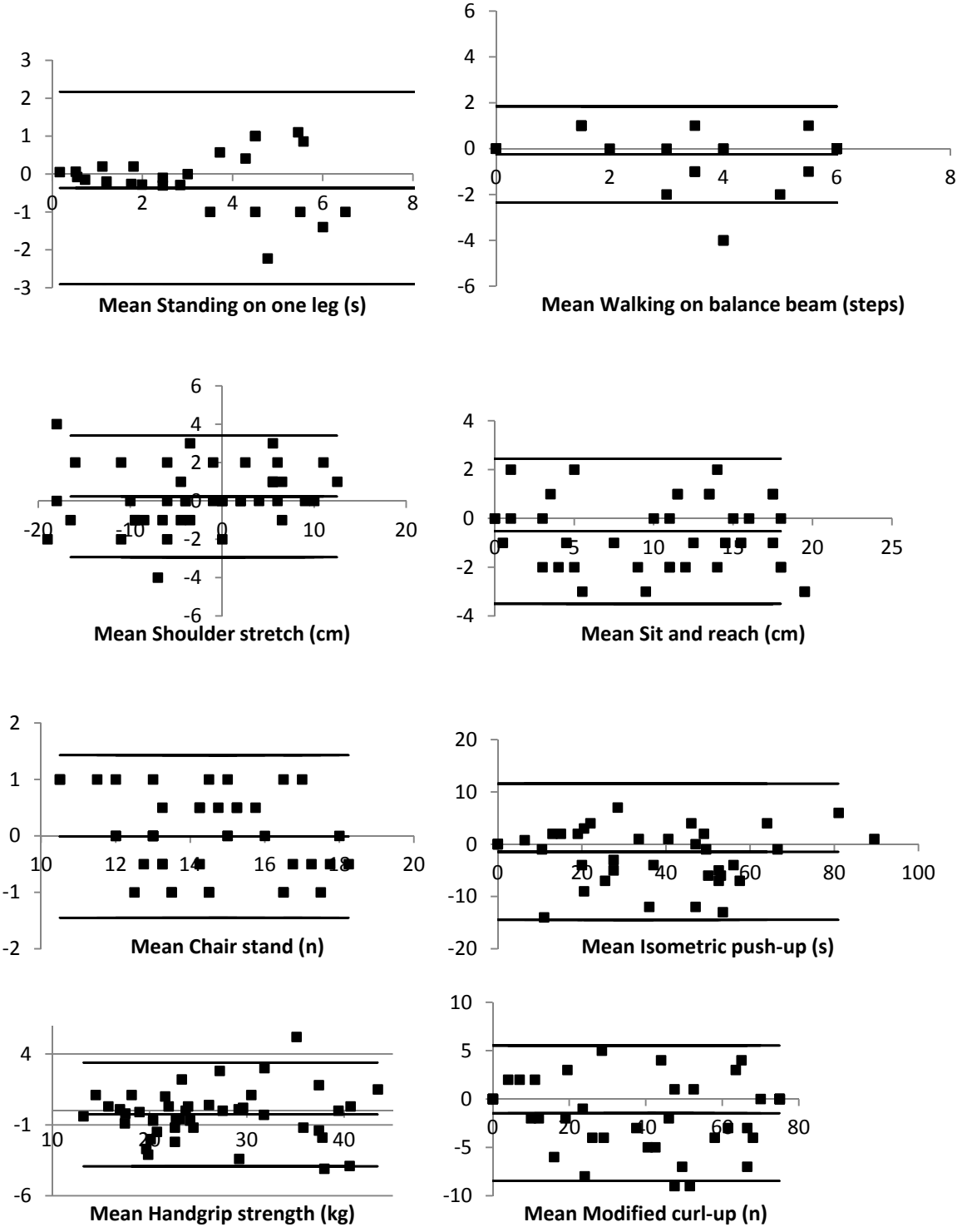
Table 2
Test-retest reliability of 12 functional test items in adult persons with Down syndrome

Test	**Mean ± SD**		**ICC (95% CI)**	**SEM**	**MDC_{90}**
	Test 1	**Test 2**			
SOOL Left (s)	5.9 (3.7)	6.0 (3.6)	0.98 (0.96-0.99)	0.53	1.23
SOOL Right (s)	5.5 (3.5)	5.9 (3.6)	0.93 (0.88-0.96)	0.92	2.14
WOBB (steps)	4.3 (2.1)	4.6 (2.1)	0.93 (0.88-0.96)	0.55	1.28
SS Left (cm)	-4.2 (10.1)	-4.1 (9.8)	0.99 (0.97-0.99)	1.24	2.89
SS Right (cm)	-2.3 (8.6)	-2.6 (8.2)	0.98 (0.97-0.99)	1.15	2.68
S&R Left (cm)	8.2 (8.8)	8.9 (9.1)	0.98 (0.97-0.99)	1.21	2.83
S&R Right (cm)	8.4 (8.7)	8.9 (9.1)	0.98 (0.97-0.99)	1.09	2.55
Chair stand (n)	14.4 (1.9)	14.4 (2.1)	0.94 (0.89-0.97)	0.48	1.12
Isometric push-up(s)	42.8 (38.8)	44.2 (37.0)	0.99 (0.97-0.99)	4.75	11.08
HGS (kg)	26.3 (8.2)	26.5 (8.1)	0.98 (0.95-0.99)	1.29	3.02
Curl-up (n)	37.8 (26.5)	39.3 (26.9)	0.99 (0.98-0.99)	2.65	6.15
Trunk lift (cm)	27.8 (9.3)	27.9 (10.5)	0.96 (0.93-0.98)	1.91	4.47
8-foot up-and-go (s)	5.5 (1.2)	5.4 (1.1)	0.94 (0.89-0.97)	0.30	0.70
16-m pacer (n)	22.8 (13.5)	23.0 (13.8)	0.99 (0.98-0.99)	1.54	3.60
6 MWD (m)	518.4 (81.5)	513.1 (82.3)	0.93 (0.88-0.96)	21.24	49.57

Data are presented as mean and standard deviation. ICC and 95% confidence intervals are shown as well as SEM and MDC_{90}. Abbreviations: 6 MWD, six minute walk distance; HGS, Handgrip strength; m, metre; MDC, minimum detectable change; SEM, standard error of measurement; SOOL, standing on one leg; SS, shoulder stretch; S&R, sit and reach; WOBB, walking on balance beam.

Figure 1 contains the Bland-Altman plots for all 12 functional fitness tests, illustrating the difference between the first and second test against the participant's mean of the two tests. Each data point

represents the difference between the two methods for each participant. The centre line indicates the mean difference between the tests, and the outer two lines indicate ± 2 SDs of the mean. The analyses indicated that there was no major systematic bias in the plots, although there were a few outliers. The scatter around the Bland-Altman was distributed randomly.



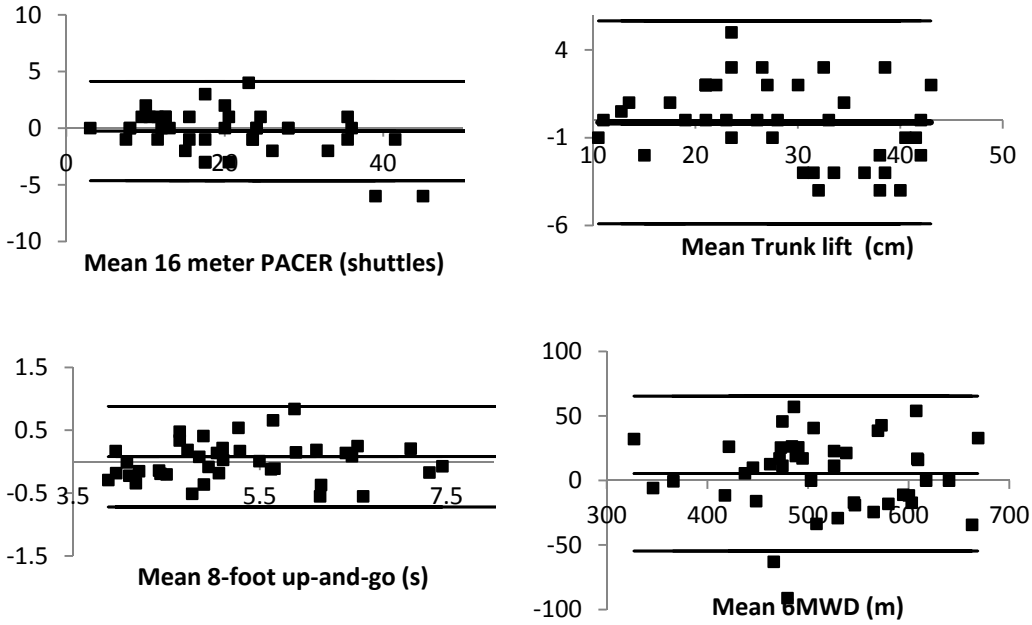


Fig. 1 Bland & Altman plots for the agreement between test and re-test of 12 functional fitness tests. The difference between the first and second test was plotted against the participant's mean two scores. The centre line equals the mean difference between the two tests and the outer lines equal ±2 SD of the mean.

4. Discussion

The main finding of the current study indicates that all twelve functional fitness tests demonstrated desirable test-retest reliability (ICC's >0.9) in adults with DS (n=43). The findings also showed that all test items were feasible, as previously demonstrated in 371 adults with DS (Terblanche & Boer, 2013). This is the first study that we are aware of, where reliability is determined for a full battery of test items in an exclusive population of adults with DS. ICC values of greater than 0.9 and alphas of greater than 0.8 were also shown for the same test items in the general elderly population and individuals with ID (Winnick & Short, 2014; 1998; Lin et al., 2004; Rikli & Jones, 2013; Abizanda et al., 2012; Wuang et al., 2009). All SEM and MDC scores at 90% confidence interval also indicated acceptable precision (SEM<SD/2) and low variability. Factors that may explain the high ICCs and low SEMs found in all tests might be as a consequence of the rigorous selection procedure of test (intensive literature review, use of test items with already desirable reliability and validity in other populations, input from various experts involved with DS individuals, three-month trial and error pilot study), practice and familiarisation sessions to avoid the learning effect, and clear, concise and simple communication with study participants. Cuing as described by Ries, Echternach, Nof, & Gagnon-Blodgett (2009) was also used with great effect. If a participant is verbally cued, physically prompted, demonstrated and progressively guided in correct and incorrect technique, understanding is more successful.

Regarding balance and flexibility test items, our study revealed ICC values very similar to what is found in elderly adults in the general population and adolescents with ID (0.94-0.99) (Wuang & Su, 2009;

Winnick & Short 1998). Concerning strength items, two of the five strength tests have been conducted on elderly individuals with ID (without DS). Hilgenkamp et al. (2012) revealed ICC values of 0.72 and 0.94 for the chair stand and handgrip strength tests respectively. The ICC value for handgrip strength is congruent with the results of our study but lower body strength as measured by the chair stand test was poorer. With reference to the remaining three strength tests (curl-up, trunk lift and isometric push-up) we are not aware of any studies that have been conducted on adults with ID or DS. Studies on adolescents with ID, reported ICC values ranging between 0.82 and 0.99 (Winnick & Short, 2014). Lastly, the only study that we are aware of that has conducted test-retest reliability with the 8-foot get-up-and-go test was performed on adults in the general elderly population. An ICC value of 0.95 is very similar to the results of our study.

It is of great value that the 16-m PACER test showed excellent test-retest reliability. Previously, Gillespie (2011) demonstrated an ICC value of 0.53 with a 20-m PACER test in adolescents with ID. We are not aware of any studies that have been performed on adults with DS. The distance of the 20-m PACER test is too great and many individuals terminate the test prematurely as they can no longer keep up with the pace, even though they are not aerobically fatigued. We observed these findings in our pilot study when test choice was further refined. The maximum heart rate in the 16-m PACER was higher compared to the 20-m PACER test. We recommend that each individual is to be tested alone, with the test investigator running alongside the participant, providing constant and standardised motivation.

Vis and colleagues (2009) demonstrated that there was no significant difference in distance walked during the 6MWD test in adults with DS. Test-retest reliability of this test item revealed an ICC value of 0.98 in adults with ID (Nasuti et al., 2013). Another study demonstrated an ICC value of 0.82 and SEM of 30 in adolescents with ID (ICC = 0.82 and SEM = 30) (Elmaghoub et al., 2011). The mean ICC value for the current study lies between the values of these two studies but the SEM was less compared to the study by Elmaghoub et al. (2011). Participants in the study by Nasuti et al. (2013) were recruited from the Special Olympics and the sample size was small (n=13) which may explain differences to our results and those by Elmaghoub et al. (2011).

For the first time, we have initiated the process of a test battery in an exclusively DS adult population. We are not aware of any physical fitness battery or normative tables for adults with DS. It is important to establish population-specific reliability coefficients exclusively for adults with DS. MDC scores at 90% confidence interval for these tests will help determine whether the change measured in future experimental studies is due to error or treatment. Magnitude of change after an intervention treatment should exceed the foreseen measurement error and variability. Information of this nature will help to determine and monitor performance alterations over time and the success of training interventions as reported in Boer & Moss (2016). The researchers in this study demonstrated that the 6MWD, 8-foot get-

up-and-go, and chair stand tests all improved over and above the MDC_{90} scores reported in the current study after 12 weeks of exercise intervention.

A limitation of the current study is that the level of ID could not be determined as the ID centres did not have any information pertaining to the intelligence quotient of the participants. However, all participants in the current study fully understood the instructions and information pertaining to test procedures and techniques. It would be very worthwhile to analyse the level of ID as a cofactor during many of the exercise tests (Vis et al., 2009). However, in the study by Boer, Meeus, Terblanche, Rombaut, Wandele & Hermans, et al. (2014) no such effect was found in an ID population without DS. Also, individuals were excluded from the study if they did not demonstrate the cognitive ability to perform exercise tests. As such, the results of the current study cannot be generalised to those with severe ID. However, a study by Waninge et al. (2011) demonstrated that the 6MWD test is feasible and reliable in individuals with severe ID. All participants in the current study had trisomy 21-type DS and the results cannot be generalised to those with mosaic DS. Lastly, if a holistic profile of balance ability is deemed in this population, it is advised that a full spectrum of balance tests such as BOTMP or Berg balance scale be conducted. Future studies should perform these tests on older adults with DS (>50 years). Furthermore, the inter-rater reliability and validity of these test items need to be confirmed.

5. Conclusion

The results of the current study yielded adequate test-retest reliability. All of the tests in the current study are user-friendly, easy to perform and field-based and can therefore be administered with great simplicity at centres for DS individuals.

Conflict of interest

There are no conflicts of interest or financial disclosures for any author of this manuscript.

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CHAPTER FOUR

Validity of the 16-metre PACER and six-minute walk test in adults with Down syndrome

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Abstract

Purpose: The purpose was to establish criterion-related validity of the 16-metre PACER and 6 minute walk distance (6MWD) to VO_2 peak, as well as predictors of VO_2 peak in adults with Down syndrome (DS). *Methods*: Adults with DS (24 male and 19 female) aged 18 to 50 years performed the three aerobic tests on non-consecutive days during a one-week period. To assess validity, peak oxygen uptake was measured directly on a motorised treadmill. Pearson-product moment correlations were performed. Predictors of VO_2 peak were assessed with a stepwise multiple regression analysis. Agreement between PACER and VO_2 peak was assessed by Bland-Altman plot. *Results*: Linear regression revealed that the PACER (R^2=0.86) and the 6MWD (R^2=0.75) was significantly related to VO_2 peak ($p<0.05$). Both the 16-metre PACER and the 6MWD significantly correlated with VO_2 peak for adults with DS. The relationship was stronger for the 16-metre shuttle run test (r=0.87) than the 6MWD (r=0.78). The correlation between VO_2 peak and both field tests, controlling for age, gender and BMI, remained significant ($r>0.7$; $p<0.05$). PACER, 6MWD and BMI are significant predictors of VO_2 peak ($p<0.05$). *Conclusion*: The 16-metre PACER and 6MWD are valid field tests for predicting aerobic capacity in adults with DS.

Introduction

Down syndrome (DS) is the most common chromosome anomaly that results in an altered development pattern and causes associated physical and intellectual characteristics. It occurs in all races and across all socio-economic levels [1]. The prevalence is 8.27 per 10 000 individuals in the USA [2]. Prevalence in developing countries like South Africa is higher [3].

The body composition, aerobic capacity and muscular strength of adults with DS is poor, not only compared to the general population but also to those with intellectual disability (ID) without DS [4,5,6,7]. A poor aerobic capacity is considered a risk factor for cardiovascular diseases [8]. These limitations have been shown to be associated with functional ability and may impede the ability to perform regular everyday living activities in this population [9,10]. Moreover, adults with DS age prematurely and suffer from an early onset of age-related diseases [9,11]. Taken together, these conditions may hamper the quality of life in old age, especially since individuals with DS live longer than in the past [11,12]. Fortunately, structured exercise training has proven to be effective in improving parameters associated with physical fitness, body composition and functional ability in adults with DS [13,14,15,16 17,18].

Specifically, the aerobic capacity of adults with DS have improved with various training modalities [13,14,15,18]. Cardiovascular endurance is best measured with a VO_2 peak test in this population [19]. Moreover, such a test and the testing protocol has been validated specifically in this population and is used widely [13,14,20,21,22]. The test is performed on a motorised treadmill and the protocol starts at an initial velocity of 4 km/h at 0% incline. After every 2 minutes, the incline increases by 2.5% until an incline of 12.5% is reached. If the participant reaches this stage, the speed is increased by 1.6 km/h every minute until exhaustion [22]. However most health practitioners do not have access to sophisticated laboratory tests to analyse and monitor the aerobic capacity of adults with DS. These tests are expensive, unavailable and time-consuming. Previously, Fernhall et al. [19] validated three cardiovascular field tests (20-metre PACER, 16-metre PACER, 600-yard run) and developed regression equations for children with ID. Two years later, the 20-metre PACER was cross validated in 17 children and adolescents with ID (6 children had DS) [23]. Guerra et al. [21] demonstrated that this prediction equation was not suitable for children and adolescents with DS. We have previously demonstrated that the 16-metre PACER test was both feasible and reliable in adults with DS [24].

The six-minute walk test (6MWD) is a simple, practical, feasible, objective and inexpensive field test that is easy to perform and suitable for individuals with DS [25]. The test is used in diverse populations including adults with obesity, cerebral palsy, Alzeimer's disease and those with severe ID [26,27,28,29]. The 6MWD is associated with aerobic capacity and is related to everyday living activities [30,31]. It involves the ability to perform large-muscle activity over an extended period of time. A walking test

may also be ideal in adults with DS, as this is their most common form of physical activity [32]. The first six stages of the standardised VO_2 peak treadmill test in this population also involve walking [22]. The rationale for standardising the time, rather than distance (e.g. 600-yard test) is to improve the discrimination ability of the test. Many elderly individuals in the general population and adults with DS cannot always complete prescribed distances. With a timed test, everybody (fit and unfit) can record a score. The 6MWD is a reliable and valid measure of aerobic capacity in adults and adolescents with ID [33,34]. Concerning adults with DS, one study determined the discriminant validity of the 6MWD but the results of the study indicated that the 6MWD was not discriminative between adults with and without cardiac disease [25] The 6MWD demonstrated excellent test-retest reliability in adults and adolescents with DS [24,34].

Considering the fact that not all adults with DS have access to confined laboratory exercise tests and since both the 16-metre PACER and 6MWD tests have shown adequate feasibility and reliability in this population, the primary purpose of this study was to establish the criterion-related validity of the 16-metre PACER and 6MWD in adults with DS. Secondly, the study also aimed to identify predictors of VO_2 peak using anthropometrical variables, gender, age, leg strength and the field tests (16-metre PACER and 6MWD).

Methods

Participants

A total of 43 healthy adults with DS (24 male and 19 female) aged 18 to 50 years performed three tests (VO_2 peak, 6MWD and 16-metre PACER) on non-consecutive days in a one-week period. Participants volunteered to participate in the study from three care centres for persons with intellectually disabilities in two provinces of South Africa. Four of the participants included have participated in the Special Olympics. The study protocol was approved by the Ethics Committee of the North-West University (NWU-00064-14-A1). Individuals were included if they had DS, were between 18 and 50 years of age, had the cognitive ability to understand and perform the exercises in a technically correct manner and if they provided written informed assent. Individuals were excluded if they suffered from congestive heart disease or any other physical or medical condition that was considered a contraindication for exercise participation. The consent form was signed by the parent or legal representatives. The health questionnaire (adapted Physical Activity Readiness Questionnaire (aPARQ)) was completed by a health professional working at the facility. The questionnaire consists of seven questions regarding the health of the participant. If participants answered “yes” to any of the questions, a physician or nurse was contacted for further examination and permission to exercise.

Procedures

Participants were familiarised with all tests and procedures on two separate occasions one week before the study commenced, as recommended by Casey et al. [35] and Fernhall et al. [22]. During these

familiarisation sessions, the tests and procedures were physically demonstrated and participants were verbally instructed (clear, concise and simple communication), physically prompted and progressively guided. It also helped with pacing strategies and gave a feel of the duration of the tests. All procedures were followed identically as outlined in the Senior Fitness Manual and Brockport Physical Fitness Test [30,36]. All tests were performed indoors in a spacious, non-slippery, temperature-controlled and well ventilated room. Tests were performed at the same time of day on each occasion for each individual (between 8 and 11 in the morning). Participants were not allowed to consume alcohol, caffeine or any diuretic medication at least 24 hours prior to testing and refrained from exercise during this period. Adults with DS were maximally motivated during all tests, as previously described by Varela et al. [37] and Millar et al. [38], with standardised motivational phrases every 15 seconds [33]. During the first visit, demographic information and body mass and length measurements were obtained. Immediately thereafter, the participant performed the chair stand test. Participants then drew a card from a hat to determine which aerobic test (VO_2 peak, 16-metre PACER, or 6MWD) would be performed on that day and the subsequent order of the tests. There was an equal probability of selecting one of the six possibilities for the order of tests. Hence, 14 participants performed the 6MWD test, 14 performed the 16-metre PACER and 15 performed the VO_2 peak test on this day. Two days later, participants performed the second test, and when another two days had elapsed, they performed the third test. Tests were always performed individually and 1:1 with a pacer to avoid the confounding influence of inter-individual competitiveness [33].

Test items

The items chosen are all considered valid and reliable [30,36] and are easy to administer.

Body mass and standing height

Body mass was determined with a calibrated electronic scale (Beurer, Ulm, Germany) and recorded to the nearest 0.1 kg. Participants were barefoot and clothed in lightweight clothing. Standing height was measured using a sliding steel stadiometer (Siber-Hegner GPM, Switzerland). The subjects were barefoot, with heels together and upper back, buttocks and heels against the wall. The head was positioned in the Frankfurt plane. The Frankfurt plane is achieved by positioning the lower edge of the eye socket (orbital) in the same horisontal plane as the notch just above the tragus of the ear (region). The measurement was taken from the inferior aspect of the feet to the vertex of the skull (the highest point on the skull) and taken to the nearest 0.1 cm. The height (m) squared and weight (kg) was used to determine body mass index (BMI).

Leg strength

Leg strength was assessed with the chair stand test [30]. Participants sat on a straight-backed chair (43.18 cm in height and with no side arms), feet flat on the floor and arms across the chest. On the signal go, the participant rose to a full stand, and then returned to a fully seated position. The score was the number

of stands completed in 30 s (two trials). In the general older population, test-retest reliability was good (ICCs of 0.84 for men and 0.92 for women) [30] and criterion-related variability as a measure of lower body strength was confirmed (r = 0.78 for men and 0.71 for women) [39]. Test-retest reliability in older adults with ID was moderate (ICC = 0.72 for a same-day interval and 0.65 for a two-week interval) [40]. Discriminant validity has been reported in adults with DS in the age categories 18–25, 26–35, 36–45 and >45 years [9].

16-metre PACER test (modified shuttle-run test)

Cardiorespiratory endurance was assessed by means of a 16-metre modified shuttle-run test. The 16-metre PACER test can be performed indoors with limited space, is fun, does not require self-monitored pacing, and incurs fewer motivational problems. At the sound of a tape-recorded beep, participants ran between two lines, 16 metres apart. The test score was the number of shuttles completed on pace. Only one trial was permitted. The 16-metre shuttle-run test has adequate test-retest reliability (ICC = 0.98) when measured one week apart in children and adolescents with ID [41]. We have previously demonstrated that the 16-metre PACER test was both feasible and reliable in adults with DS [9,24]. Criterion-related validity has been shown with the direct measurement of VO_2 peak in individuals with ID [19]. Discriminant validity has been reported in adults with DS in the age categories 18–25, 26–35, 36–45 and >45 years [9].

Six-minute walk distance test

The participants completed a standardised, self-paced six-minute walk distance test (6MWD). They were instructed to walk as many laps as possible of 50 yards within the allocated six minutes. The participants were allowed to rest at any time, but were encouraged to continue as soon as possible. The distance covered was recorded to the nearest metre. This test assesses aerobic endurance and functional ability that is important for many everyday living activities such as walking distances for shopping and sightseeing while on vacation [30,33]. Test-retest reliability of this test item revealed an ICC value of 0.94 in the general elderly population [30] and an ICC value of 0.98 in adults with ID [33]. Repeated analysis of variance showed no significant difference between tests in a group of DS adolescents provided that two practice sessions were given [35]. In adults with DS it has been reported that the 6MWD test demonstrated excellent test-retest reliability [24]. Criterion-related validity was reported with treadmill performance using the modified Balke protocol [42] and VO_2 peak (R=0.84) in adults with ID [33].

VO_2 peak test

The VO_2 peak test was performed with the (Metalyzer 3B system (Cortex, Leipzig, Germany). The test was carried out on a motorised treadmill using the standardised protocol for adults with DS. This protocol starts at an initial velocity of 4 km/h at 0% incline. After every 2 minutes, the incline increases by 2.5% until an incline of 12.5% is reached. If the participant reaches this stage, the speed is increased

by 1.6 km/h every minute until exhaustion. This protocol is used widely in scientific research concerning participants with DS [13,21], and is considered valid and reliable in a DS adult population provided that adequate familiarisation sessions are conducted [22]. An ECG was recorded during the VO_2 peak test (Custo Med, Schiller, Switzerland). The VO_2 peak was taken as the highest recorded measurement during the final 30 seconds of the tests (after the data was filtered to every 10 seconds) with an RER greater than 1.0 [31]. Participants were asked and continually encouraged to perform the test until maximal exhaustion [37]. The test was stopped if any clinically significant ST segment deviations, arrhythmias, chest pain or dizziness arose or when they requested to terminate the test. Participants were maximally motivated during all tests with standardised phrases of encouragement provided every 15 seconds [33].

Statistical analysis

All data was reported using a commercially available software package (SPSS, Version 20.0, Inc., Chicago, IL, USA). Descriptive statistics were expressed as mean and standard deviations (SD). Normality and homoscedasticity of data was assessed with the Kolmogorov-Smirnov and Leven tests respectively. The relationship between $\dot{V}O_2$ peak and field tests (16-metre PACER and 6MWD) were determined by Pearson-product moment correlation. Predictors of VO_2 peak were analysed with a stepwise multiple regression analysis. Age, height, body mass, BMI, leg strength and field tests were included in a stepwise linear regression analysis [10,19,33,34]. In the first analysis, only one field test (shuttles ran during the 16-metre PACER) along with the demographic variables (age, gender, body mass index), and leg strength was included in the prediction model. A paired sample T-test was performed between predicted and measured VO_2 peak. The predicted values were obtained using the prediction model obtained from the regression analysis equation. A Bland-Altman plot with 95% limits of agreement was constructed from the difference for VO_2 peak measured and indirect VO_2 peak from the 16-metre PACER test and the average of the VO_2 peak determined from both the field tests. In the second analysis, the other field test (6MWD) along with the demographic variables and leg strength was included in the prediction model. Level of significance was set $p \leq 0.05$.

Results

All forty-three participants completed all tests. No serious or adverse events occurred during any of the exercise testing. None of the participants stopped to rest during the 6MWD. The VO_2 peak test was continued until volitional exhaustion [43,44,45]. Descriptive characteristics of participants indicate a larger percentage of males compared to females and an average BMI of 30.0 SD 4.9 kg/m^2 (Table 1). Descriptive information regarding all physical tests are provided in Table 2. All participants were healthy. Also that five of the participants have participated in the Special Olympics. It is stated in the methods section individuals were excluded if they suffered from congenital heart disease or any other physical or medical condition that was considered a contraindication for exercise participation.

Table 1: Demographic variables of the participants with DS (n=43)

Variable	Minimum	Maximum	Mean (SD)
Gender (Male/Female)	N/A	N/A	24/19
Age (years)	19	50	33.6 (8.4)
Body mass (kg)	43	97	72.0 (10.7)
Height (cm)	143	173	154.3 (7.5)
BMI (kg/m^2)	20	41	30.0 (4.9)
Chair stand test (n)	9	19	14.5 (2.2)
VO_2 peak (ml/kg/min)	23	52	33.9 (7.5)
6MWD (m)	343	686	537.4 (85.6)
16 metre PACER (shuttles)	8	68	23.5 (14.0)

BMI, body mass index; VO2, oxygen uptake; 6MWD, six-minute walk distance; N/A, not applicable

Table 2: Descriptive information regarding fitness and physical tests (*n*=43)

Variable	Minimum	Maximum	Mean (*SD*)
Chair stand test (n)	9	19	14.5 (2.2)
6MWD (m)	343	686	537.4 (85.6)
16-metre PACER (shuttles)	8	68	23.5 (14.0)
16-metre PACER(HR)(bpm)	146	192	164.1 (9.1)
VO_2 peak (ml/kg/min)	23	52	33.9 (7.5)
VO_2 peak (L/min)	1373	3557	2384.5 (502.8)
VO_2 peak (HR)(bpm)	144	181	163.9 (10.8)
RER	1.05	1.40	1.14 (0.06)
VE (L/min)	45.2	118	79.9 (17.04)

Bpm, beats per minute; HR, heart rate; n, number; m: meter; RER, respiratory exchange ratio; SD, standard deviation; VE, minute ventilation; VO_2, oxygen uptake; 6MWD, six-minute walk distance

The relationship between VO_2 peak and the 16-metre PACER and 6MWD tests are graphically demonstrated in Figure 1.

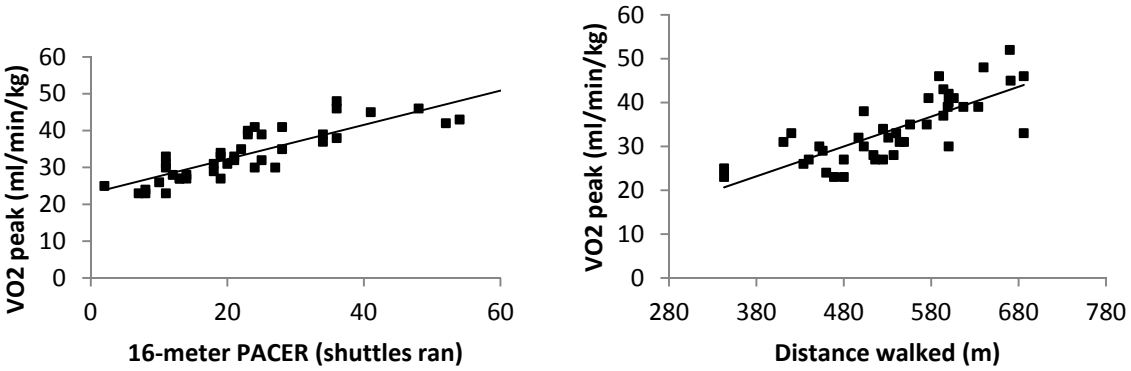

Figure 1. 16-metre PACER and distance walked (6MWD) plotted on the x-axis and VO_2 peak on the y-axis. Pearson correlation was significant for 16-metre PACER (r=0.87) and the 6MWD (r=0.78)

The results of the linear regressions are reported in Tables 3 & 5. Best subsets multiple regression revealed a significantly high $R^2 = 0.86$ to predict VO_2 peak using demographic variables (age, gender, body mass, stature, BMI) and the 16-metre PACER field test as independent variables (Table 3). Shuttles ran during the 16-metre PACER, BMI, age and gender significantly predicted 85% of the variance in VO_2 peak. Shuttles ran contributed the most to the prediction model.

Table 3: Predictors of peak VO_2 using demographic variables and the 16-metre PACER test as independent variables

Predictive variable	**Unstandardised (ß)**	**SE**	**t-value**	**p-value**	**Standardised estimate (Beta)**
Intercept	48.43	5.03	9.2	<0.01	/
16-metre PACER (n)	0.32	0.05	6.9	<0.01	0.59
BMI (kg/m^2)	-0.45	0.13	-3.5	<0.01	-0.29
Gender	-2.88	0.96	-3.0	<0.01	-0.19
Age (yrs)	-0.13	0.05	-2.5	0.02	-0.15
VO_2 peak = 48.23 +0.32 (16m PACER) -0.45 (BMI) -2.88 (Gender) -0.13 (Age) Gender (1 male, 2 female)					

BMI: body mass index; SE: standard error; yrs: years.

Predicted VO_2 peak (using our generated prediction equation from Table 2) demonstrated a strong relationship with directly measured VO_2 peak (Figure 2A). Predicted VO_2 peak was not significantly different from measured VO_2 peak (Table 4). The Bland Altman plot revealed no heteroscedasticity with a mean difference of -0.003 between predicted VO_2 peak and measured VO_2 peak and limits of agreement (5.62 and -5.63) (Figure 2B).

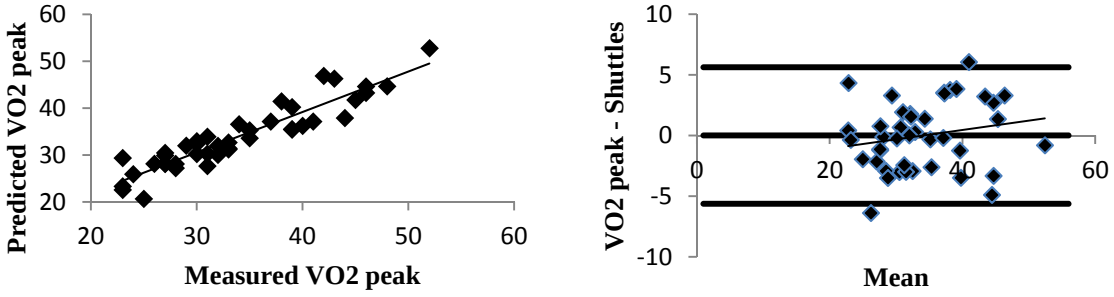

Figure 2. A. Predicted (using data from Table II) versus measured VO_2 peak (r=0.93). B. Bland-Altman plot of mean differences between measured and predicted VO_2 peak and the mean of the two respective values

Table 4: Predicted versus measured VO_2 peak

Variable	**Mean**	**SD**	**t-value**	**p-value**
Measured VO_2 peak (ml/min/kg)	33.95	7.57	-0.006	0.995
Predicted VO_2 peak (ml/min/kg)	33.96	7.03		

SD: standard deviation

Best subsets multiple regression revealed a significantly high R^2=0.75 to predict VO_2 peak using demographic variables and the 6MWD test as independent variables (Table 5). Distance walked during the 6MWD and BMI significantly predicted 75% of the variance in VO_2 peak. Distance walked contributed the most to the prediction model.

Table 5: Predictors of peak VO_2 using demographic variables and the 6MWD test as independent variables

Predictive variable	Unstandardised (ß)	SE	t-value	p-value	Standardised estimate (Beta)
Intercept	30.27	8.03	3.8	<0.01	/
6MWD (m)	0.05	0.01	5.4	<0.01	0.53
BMI (kg/m^2)	-0.70	0.15	-4.67	<0.01	-0.45

6MWD: six-minute walk distance; BMI: body mass index; SE: standard error.

Discussion

The primary aim of the study was to establish the criterion-related validity of the 16-metre PACER and 6MWD field tests. The secondary aim was to establish predictors of VO_2 peak. Linear regression analysis revealed that the PACER (R^2=0.86) and the 6MWD (R^2=0.75) was significantly related to VO_2 peak, the gold standard for cardiovascular endurance testing in adults with DS ($p<0.05$). Also, both the PACER and the 6MWD aerobic tests significantly correlated with the VO_2 peak test ($p<0.05$). The relationship was stronger for the 16-metre PACER test (r=0.87) than the 6MWD (r=0.78). The correlation and/or adjusted R^2 is slightly better than those presented in adolescents and adults with ID and DS [33,34,46]. The correlation to VO_2 peak for both field tests remains significant and greater than 0.7 even when controlling for age, gender and BMI. This is the first study that we are aware of that assessed the validity of the 16-metre PACER and 6MWD field tests in adults with DS.

Previously, Fernhall et al. [19] showed that the 16- and 20-metre PACER tests revealed a significant relationship with VO_2 peak in children with ID (r=0.74 & 0.77). Two years later, they cross validated the prediction equation [23]. However, Guerra et al. [21] showed that the prediction equation was not valid for children with DS. A more recent study also attempted to validate a prediction equation (using the 20-metre PACER test) in youth with DS [46]. However, the predictive ability was low (R^2=0.23). The limits of agreement (-9.3 to 9.3 ml/min/kg) indicated large variability. Moreover, the scatter around the Bland-Altman plot demonstrated heteroscedasticity (over-predictive in participants with low actual measured VO_2 peak and under-predictive in participants with high VO_2 peak). In our pilot study, it was realised that adults with DS struggle to participate in the 20-metre PACER test. Many participants did not reach the sixth or seventh shuttle (still level one in the test). This was confirmed by Agiovlasitis et al. [46] where the mean value of shuttles reached was 8.6. We have recently determined the feasibility, test-retest reliability and discriminant validity of the 16-metre PACER in adults with DS [9,24]. The reduction in distance (20-metre PACER compared to 16-metre PACER) allowed the participants in the current study to reach a mean of 23.5 shuttles and consequently are more likely to overcome challenges presented with high initial speed and consequently progress to higher HR maximum values (Table II). The HR maximum was not significantly different between the PACER and VO_2 peak tests.

We are only aware of studies that have established the criterion-related validity of the 6MWD in participants with DS. Two studies established the criterion-related validity of the 6MWD to VO_2 peak in adolescents (n=61) and adults (n=13) with ID [33,34]. In these studies the 6MWD correlated significantly with VO_2 peak (r=0.69) in adolescents with ID and revealed an adjusted R^2 of 0.67 in adults with ID. On the other hand, another study established that the 6MWD was not valid for examining cardiac restriction in adults with DS [25]. They did not establish criterion-related validity.

These field tests, together with the already established feasibility and reliability of the tests in adults with DS, could be used as standardised tests in epidemiological and experimental research studies. Valid and reliable tests are important as adults with DS do not have a battery of standardised test items to gauge and monitor functional fitness to the same extent as with instruments found in the general, the elderly and intellectually disabled populations. Consequently, adults with DS are often ignored or pooled with individuals with ID (without DS) despite evidence that the presence of DS negatively affects physical activity and test performance [4,6,7,13,36].

The current study also established predictors of VO_2 peak in adults with DS for both field tests as performed in previous studies consisting of individuals with ID and/or DS [10,19,33,34,46]. The number of shuttles ran, distance walked (6MWD), BMI, age and gender contributed to the variance in the dependent variable. The results demonstrate that both the 16-metre PACER and the 6MWD tests are aerobic in nature, as seen in their significant standardised estimates. The fact that BMI is a significant predictor in both models (Table 3 and 5) is not surprising, as Salaun and Berthouze-Aranda [47] demonstrated that individuals with ID who were the most overweight had the lowest cardiovascular fitness. The reason for the lower cardiovascular fitness values in this population can be partially attributed to the high incidence of obesity and sedentary behaviour in adults with DS [5,9,48]. The small but significant predictor contribution of age to (16-metre PACER) or not a predictor (6MWD) is in support of the fact Baynard et al. [4] found that VO_2 peak did not decline with age in people with DS (9–45 years of age). Absence of gender as a predictor in the 6MWD test is consistent with studies not only in the general population but also those with ID [34,49]. Lastly, leg strength was not a predictor of VO_2 peak, which was also shown in the study by Nasuti et al. [33] & Elmahgoub et al. [34], despite being significantly related to VO_2 peak in young adults with DS [50]. However, leg strength is still an important parameter of fitness in adults with DS as it has previously been shown to predict functional ability in everyday living activities [9,10].

The main objectives of the current study was to establish the criterion-related validity of the PACER and 6MWD tests and to identify predictors of VO_2 peak. We did however, conduct post regression analysis for the prediction of VO_2 peak using the 16-metre PACER test. We did not run this analysis for the 6MWD as the correlation coefficient and the predictive ability of the 16-metre PACER was greater. Also, due to the inherent nature of the 6MWD (continuous walking at the same pace) compared to the

PACER and VO_2 peak (progression of pace from start to finish that involve running) we did not feel comfortable to predict VO_2 peak from a theoretical perspective. Lastly, although we did not measure the maximum HR of the 6MWD test, it is unlikely reach values as shown in Table 2 (also reported in our pilot study). Concerning the PACER test, the predicted VO_2 peak values were not significantly different from measured VO_2 with a significant strong relationship between predicted and measured VO_2 peak (r=0.93)(Table 4). The relationship and coefficient of determination is stronger compared to the study by Fernhall et al [19] when a combination of ID and DS participants were studied. Perhaps this finding can be attributed to the heterogeneity of their study participants [19]. It has been shown that the VO_2 peak of individuals with DS is lower than those with ID, without DS [4]. However, in our study the same independent variables (16-metre PACER, BMI and gender) predicted VO_2 peak compared to Fernhall et al. [19]. The Bland-Altman plot demonstrated no systematic bias and no heteroscedasticity (increase in variance with an increase in mean VO_2). The magnitude of the limits of agreement was less than previous studies (also developing prediction equations for the measurement of VO_2 peak but with the 20-metre PACER test in individuals with DS) [23,46]. Individuals with DS can use the prediction equation provided they are between 18 and 50 years, have no congenital heart disease or any other physical or medical condition that is a contraindication for exercise performance. The guidelines and procedures as stipulated with PACER testing should be followed closely. However and importantly it is advised that the prediction equation should first be cross-validated in future studies.

In conclusion, the results of our study indicate that both the 16-metre PACER and 6MWD tests are valid indicators of cardio-respiratory fitness as assessed by moderate to strong coefficients of determination and correlation coefficients. Both field tests along with BMI, age and gender are predictors of aerobic capacity.

Limitations and future studies

It was not possible to include level of ID (profound, severe, moderate, mild) in the prediction model of the current study, as the three centres did not have any information concerning the level of ID of the 43 participants. However, only participants who understood test instructions and procedures were included in the study. There were therefore no participants with severe or profound ID in the current study. Boer et al. [51] previously also found no effect of ID on baseline performance in adolescents with ID when no participants with severe ID were included. Severe ID in adults with DS has been found to correlate significantly with a lower VO_2 peak [25].

All participants in the current study had DS type trisomy 21 and results of the current study cannot be generalised to those with mosaic-type DS (5% of population). Lastly it was not possible to randomise the tests in such a way that half the participants performed one field test and the other half the VO_2 peak and to reverse thereafter. Otherwise, participants would have had to complete the VO_2 peak test on two occasions. Two field tests were measured against the VO_2 peak test and thus the order of all tests had to be fully randomised.

Last, it was not possible to predict VO_2 peak values separately for men and women as the sample size would be too small. Future studies should focus on further refining the predictive ability of VO_2 peak using the 16-metre PACER test. Additionally and importantly a future study would need to cross-validate this prediction equation.

Future studies should focus on older people with DS (>50 years). Even though the feasibility of many tests have been established in elderly adults with DS [9], the reliability and validity remain to be determined. Also, it would be interesting to see whether the 16-metre PACER or 6MWD is more reliable or valid in this population. Perhaps the 10-metre incremental shuttle walking test as used for elderly (>50 years) individuals with ID would be a more suitable alternative [52]. It would also be worthwhile to validity of predicting VO_2 peak using the 6MWD or the 10-metre incremental shuttle walking test in older adults with DS.

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CHAPTER FIVE

The influence of sprint interval training on body composition, physical and metabolic fitness in adolescents and young adults with intellectual disability: a randomised controlled trial.

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Abstract

Objective: In this study we evaluated the effect of sprint interval training on metabolic and physical fitness in adolescents and young adults with intellectual disabilities when compared with continuous aerobic training and no training (control). **Methods:** Fifty-four persons with intellectual disabilities (age: 17 (3.0), body mass index: 27.7 (3.7), intelligence quotient: 59 (8.6)) were matched based on age, gender and intelligence quotient between sprint interval training (n = 17), continuous aerobic training (n= 15) and control (n = 14). Sprint interval training was composed of three blocks of 10 minutes at ventilatory threshold (blocks 1 and 3: 10 sprint bouts of 15 seconds, followed by 45 seconds relative rest; block 2: continuous training) twice a week for 15 weeks. Continuous aerobic training was composed of three blocks of 10 minutes continuous training. After eight weeks, intensity was increased to 110% of ventilatory threshold. The control group did not participate in supervised exercise training. Before and after the training period, body composition, physical and metabolic fitness were evaluated. **Results:** Sprint interval training showed a significant positive evolution for waist circumference, fat%, systolic blood pressure, lipid profile, fasting insulin, homeostasis model assessment of insulin resistance, peak VO_2, peak Watt, ventilatory threshold, 6-minute walk distance and muscle fatigue resistance when compared with no training ($P < 0.01$). The sprint interval training group demonstrated significant improvements for fat%, systolic blood pressure, low-density lipoprotein, fasting insulin, peak VO_2 and peak power and ventilatory threshold ($P < 0.01$) when compared with continuous aerobic training. **Conclusion:** In this study we could observe that sprint interval training has stronger beneficial effects on body composition, physical fitness and metabolic fitness compared with control. Compared with continuous aerobic training, interval training seems to result in better outcome.

Introduction

It has been well documented that children and adolescents with intellectual disability (ID) have suboptimal levels of cardiovascular fitness compared to persons with a typical development[1,2,3]. The decreased muscle strength, aerobic capacity, fat-free mass, and the increased fat mass are associated with a reduced metabolic condition (unfavorable lipid profile)[4,5].

Regular physical activity has beneficial effects on body composition, physical fitness, and cardiovascular risk amongst individuals in the general population of all ages without ID[6].

In adolescents and young adults with ID, Varela et al.[7] and Millar et al.[8] reported no effects on body composition, while Ordonez et al.[5] reported a significant decrease in fat mass after endurance training. These studies found no significant improvement in aerobic capacity but showed that work performance and time to exhaustion did significantly improve. Concerning metabolic fitness, only one study was published and found a significant positive and clinical relevant effect[9].

Concerning strength or resistance training in persons with ID, Weber and French[10] and Shields and Taylor[11] reported a significant improvement in muscular strength after weight training, but effects on metabolic fitness were not investigated.

A case study of Lewis and Fragala-Pinkham[12] showed an improvement of aerobic and anaerobic capacity in a 10-year-old child with Down syndrome after six weeks combined aerobic and strength without changes in body composition. Elmahgoub et al.[14,15] reported a significant improvement of aerobic capacity, muscle strength and lipid profile in adolescents with ID.

Another exercise training mode with a lot of potential is interval training. Interval training (IT) is defined as vigorous exercise performed at a high intensity for a brief period of time interposed with recovery intervals at low-to-moderate intensity or complete rest. Two distinct types of IT are sprint interval training (SIT) and aerobic interval training (AIT)[15]. Investigations examining IT report improvement of sub-maximal and maximal exercise capacity, mitochondrial biogenesis, enzymatic markers associated with glycolysis, aerobic metabolism and beta-oxidation better than continuous aerobic exercise training in the general population[15-18] .Only one study evaluated the effects of IT compared with endurance training in overweight individuals without ID. The endurance group performed a 30 to 60 minute continuous exercise at 80% of the peak heart rate (HR). The IT group performed 3 to 6 sets of 60-seconds sprint at 100% of the peak velocity interspersed by a 3-min active recovery period at 50% of the exercise velocity, twice a week for 12 weeks. In both groups physical and metabolic parameters, focusing on insulin sensitivity, were significantly ameliorated, but with the same magnitude[19].

There are no published reports concerning this training modality in a population of adolescents and young adults with ID. Therefore the purpose of our study is to evaluate the effect of SIT on anthropometric variables, physical and metabolic fitness in adolescents and young adults with ID compared with the effect of continuous aerobic training (CAT) and a control (CON) group.

Methods

For this study 70 adolescents and young adults with ID, all of which were attending secondary school at two Belgian special education schools (Ravelijn, Brugge and De Varens, Brugge), were eligible. From these subjects, 54 people were selected so three groups of 18 participants could be matched for age, sex and intelligence quotient (IQ). If three people with the same age, sex and IQ were available they were randomly allocated to one of three groups SIT, CAT or CON (Figure 1).

All participants were living with their families and had no severe physical impairments that could interfere with the training program or testing protocols. In addition, all participants were able to communicate and understand instructions given by the physiotherapists during training and testing.

All subjects and their parents were informed about the purpose of this study, its risks, and procedures. A signed informed consent was provided by all participants and their parents before study admission. The research study was approved by the ethics committee of the Ghent University Hospital.

Adolescents were diagnosed as fragile X syndrome, fetal alcohol syndrome, Prader–Willi syndrome, hydrocephalus, pervasive developmental disorder, Sotos syndrome and Steinert syndrome. In several participants, autism, epilepsy, or attention deficit hyperactivity disorder was associated with the ID. The medical record of nine ID individuals did not contain specific information about the associated impairments or the cause of their ID.

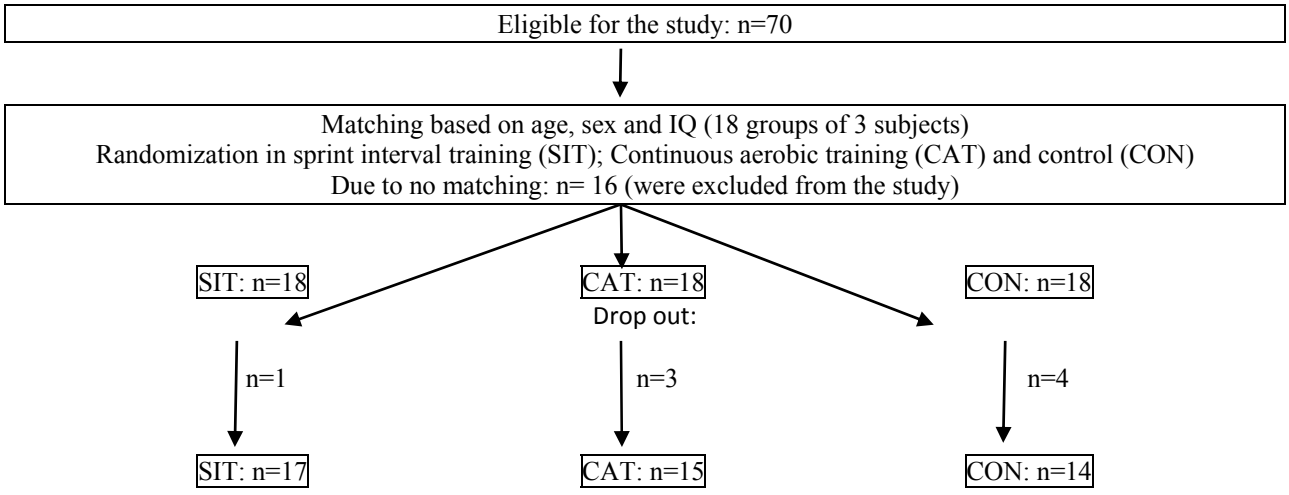

Fig. 1: Participants' flow diagram depicting the matching and randomization procedure during the trial. Drop out numbers are also indicated which were caused by other non-educational commitments.

In many participants, lower limb (flat foot, varus, and valgus knee misalignment), pelvic deformities (malposition of Spina Iliaca Posterior Superior and Spina Iliaca Anterior Superior), or both were noticed. We observed no severe interference of these deformities with their performance during the training program or testing protocols. General descriptive statistics are shown in Tables 1 and 2.

The training program was conducted in the physiotherapy wing at the special education schools. The SIT group followed a 15-week training program. The adolescents exercised for 40 minutes, twice a week. The

training was supervised by three physiotherapists and integrated into the school program replacing the usual physical education lessons. Each training session included a warm up (stretching of the large muscle groups and cardiovascular exercises at 30% of peak Watt for 5 minutes), a sprint interval block (10 minutes), continuous aerobic exercises (10 minutes), another sprint interval block (10 minutes) and cooling down (stretching of the large muscle groups and cardiovascular exercises at 30% of peak Watt for 5 minutes). For the first seven weeks, each sprint interval block consisted of 10 sprint bouts (>100 r/min), of 15 seconds at a resistance matching with the ventilatory threshold (VT_R), alternated with 45 seconds relative rest (50 r/min at VT_R). Starting from week 8 until week 15, the intensity of sprinting and relative rest was increased up to 110% of VT_R. All exercises consisted of cycling.

The protocol of the CAT group consisted of warming up (stretching of the large muscle groups and cardiovascular exercises at 30% of peak Watt for 5 minutes), cycling (10 minutes), walking/running (10 minutes), stepping (10 minutes) and cooling down (stretching of the large muscle groups and cardiovascular exercises at 30% of peak Watt for 5 minutes). During the continuous aerobic protocol (cycling, running, stepping) participants cycled for 10 minutes at a HR similar to the HT at VT (60 r/min), which was increased to 110% of VT from week 8 onwards. The subjects in the control group participated in usual everyday activities without supervised exercise training.

The quantification of all examined variables was performed by blinded assessors. All tests and measurements were conducted at the exercise laboratory of the Physiotherapy Department, Ghent University. Prior to all tests and measurements, participants were well informed and familiarized with the equipment and testing protocols. All participants from each of the three groups were tested on the same day pre- and post-intervention.

Anthropometry

Height was measured to the nearest 0.1 cm using a stadiometer (Holtain Ltd., Pembrokeshire, UK). Weight was measured to the nearest 0.1 kg on a digital balance scale (Seca, Germany) with the subject wearing lightweight clothing and no shoes. The body mass index (BMI) was calculated from weight and height. Waist circumference was measured by a tape meter at the level of the umbilicus with the subject in a standing position after normal expiration. Fat mass and fat-free mass were assessed by bio-electrical impedance analysis (Bodystat 1500 MDD, Douglas, Isle of Man, UK). Subjects were in the supine position for at least five minutes. Surface electrodes were attached to the dorsal side of the right foot and the dorsal side of the right wrist. Fat mass and fat-free mass were calculated using the formula of Wabitsch[20].

Physical Fitness

Maximal cardiopulmonary exercise test

Participants were tested on a computer-driven cyclo-ergometer (Marquette Case, Marquette Electronics, Milwaukee, WI, USA) using a ramped protocol (15 W/min) starting at 30 W. Twelve-lead electrocardiogram and HR were recorded continuously during the test, whereas blood pressure was measured with a manual sphygmomanometer every two minutes. Subjects were familiarized with the test

procedure before baseline testing. Subjects were asked to perform exercise testing to their self-determined limits of physical exhaustion or until the physician stopped the test because of severe adverse events, such as increasing chest pain, dizziness, potentially life threatening arrhythmias, clinically important ST-segment deviations, marked systolic hypotension, or hypertension. Tests were classified as maximal as respiratory exchange ratio (RER)>1.1. Respiratory gas measurements were obtained by using a Metalyzer 3B (Cortex, Leipzig, Germany). Oxygen consumption (VO_2), carbon dioxide production (VCO_2), minute ventilation (VE), tidal volume, respiratory rate, and mixed expiratory carbon dioxide concentration were measured continuously with mixed chamber analysis. Peak VO_2 was expressed as the highest attained VO_2 during the final 30 seconds of exercise according to the American Thoracic Society guidelines.
The VT was determined based on the metabolic equivalents of O_2 and CO_2 (VE/VO_2 and VE/VCO_2). The point at which the VE/VO_2 increased without an increase in VE/VCO_2 was identified as the VT [21].

Six-minute walk test

All subjects performed a standardised, self-paced 6-minute walk test (6MWT) in a 20-m-long corridor. They were asked to cover as much distance as possible within six minutes without running. Subjects were allowed to stop at any time but were encouraged to restart as soon as possible. During the test, subjects were instructed and encouraged continuously. The distance covered after six minutes was measured to the nearest meter.

Strength

Sit-to-stand test

The sit-to-stand test measures the maximum number of repetitions within 30 seconds that an individual can rise to a full stand from a seated position, without pushing off with the arms and return to a fully seated position. The participant's score is defined as the number of completed stands. This test was developed in a population of older adults and is highly correlated with strength of the lower limbs[23]. In a population with ID, there was also a high correlation of the sit-to-stand with the 1RM of lower limb strength ($r = 0.69$) [13].

Muscle fatigue resistance (MFR)

The participant had to squeeze the dynamometer with as much force as possible and for as long as possible. The test terminated when grip strength dropped to 50% of its maximum value during sustained contraction. The reliability of this test was evaluated in healthy young subjects and in a geriatric population without ID. The Intra class correlation (ICC) ranged from 0.77 to 0.94 in both groups[13].

Metabolic fitness

Blood pressure

Resting systolic and diastolic blood pressure was taken seated on a chair at the beginning of the testing after 10 minutes rest. Blood pressure was measured with a manual sphygmomanometer

Lipid profile

Blood samples were obtained after an overnight fast. The following variables were evaluated using diagnostic kits (Roche Diagnostics) for high-density lipoprotein (HDL)-C (PEG + cholesterol-oxidase), triglycerides (glycerol phosphate-PAP) and total cholesterol (cholesterol-oxidase-PAP). Low-density lipoprotein (LDL)-C was calculated from total cholesterol and HDL-C.

HOMA

Fasting plasma glucose and insulin concentrations were measured in the morning (before 10 am). HOMA-IR index was calculated as [fasting serum insulin (mU/l) * fasting plasma glucose (mmol/l)/22.5], with higher values indicating a higher degree of insulin resistance[24]. Glucose (hexokinase method) and insulin concentrations were determined on a Modular P and E respectively using Roche Diagnostics consumables (Roche Diagnostics, Mannheim, Germany).

Statistical Analyses

All data were analysed with a commercially available statistical software programme (Statistical Package for the Social Sciences, SPSS 20.0, SPSS Chicago, IL, USA). Data is expressed as mean and SD. To evaluate possible differences between groups at baseline a Univariate ANOVA was performed. To evaluate pre-post differences between groups a repeated measure ANOVA with post hoc Bonferroni test was performed. Effect sizes were calculated with Cohen's d and magnitude classified as small (0.2-0.3), medium (around 0.5), and large (0.8 to infinity).

Results

Groups were matched for age, sex and ID. There were eight dropouts, one in the interval group, three in the continuous group and four in the control group, owing to other educational commitments. Participants in the intervention groups performed a minimum of 27 sessions (40 minutes each) and a maximum of 30 sessions over 15 weeks. Interruption of training sessions was owing to illness or other commitments. No adverse or side effects were reported.

After 15 weeks of training, waist circumference and body fat percentage was decreased significantly more in the SIT compared with no training. Only percentage of body fat was decreased more in the SIT group compared with CAT (Table 1).

Systolic blood pressure decreased significantly in the SIT group when compared with the control group and CAT group ($P < 0.01$). Fasting insulin levels decreased significantly after SIT compared with the control group and to CAT. The decrease in insulin sensitivity (HOMA-IR) was more pronounced in the SIT group compared with the control group, but the change was not significantly different from CAT ($P < 0.01$). Lipid profile (triglyceride, total cholesterol, HDL, and LDL levels) ameliorated significantly in the SIT group compared with the control group but only LDL was more decreased compared with the CAT group ($P < 0.01$).

Table 1. Influence of SIT and CAT on indices of anthropometry, blood pressure and lipid profile.

Variables	SIT(n=17)		CAT(n=15)		Control(n=14)	
	Pre	Post	Pre	Post	Pre	Post
General						
Age	18 (3.2)		16.7 (3.6)		17.4 (2.4)	
Gender	11/6		10/5		9/5	
Male/Female						
IQ	59.2 (9.1)		57.3 (7.9)		59.1 (9.3)	
Anthropometry						
Weight (kg)	76.8 (18.3)	76.0 (19.1)	77.9 (9.3)	77.6 (8.5)	79.3 (14.7)	80.0 (14.4)
Height (cm)	163.7 (9.5)	164.6 (9.6)*	168.3 (7.1)	170.1(7.9)*	171.2(12.1)	171.9(11.5)
BMI (kg/m2)	28.4 (4.7)	27.7 (4.7)	27.5 (2.7)	26.9 (3.1)	26.9 (3.2)	26.9 (2.9)
Waist (cm)	95.8 (13.1)	91.5 (13.1)*	95.9 (9.6)	93.4 (9.6)*	95.0 (8.8)	95.9 (8.2)
Fat (%)	34.2 (6.9)	30.4 (7.0)*$	32.3 (7.0)	31.3 (6.6)*	32.0 (7.1)	32.0 (7.0)
Blood pressure						
SBP (mmHg)	123.5 (9.8)	113.2(8.1)*$	121.0(11.4)	119 (8.9)*	118.2(10.3)	118.6(10.1)
DBP (mmHg)	73.5 (6.8)	76.5 (8.1)	72.3 (7.8)	73.0 (8.8)	72.1 (5.4)	72.1 (4.7)
Lipid profile						
Chol. (mg/dl)	169.8(25.2)	154.8(22.9)*	162.9(26.6)	164.0(31.3)	169.6(29.7)	171.9(25.8)
HDL (mg/dl)	54.9 (13.5)	59.4 (11.4)*	48.9 (9.4)	49.7 (10.6)	59.3 (16.9)	55.9 (15.6)
LDL (mg/dl)	105.2(12.4)	95.6 (9.3)*$	96.4 (24.8)	97.4 (28.5)	92.6 (23.5)	96.0 (21.2)
Tri (mg/dl)	79.2 (22.2)	70.8 (16.7)*	91.5 (50.4)	87.5 (50.3)	96.6 (75.6)	95.0 (85.6)
HOMA-IR	2.9 (1.3)	2.3 (0.8)*	2.9 (1.3)	2.6 (1.1)*	2.6 (0.8)	2.7 (0.9)

Data are presented as mean (standard deviation). Between group differences analysis of variance with post-hoc Bonferroni was used to evaluate time and interaction effects.
SIT, sprint interval training; BMI, body mass index; CAT, continuous aerobic training; Chol, Cholesterol; DBP, diastolic blood pressure; HDL, high-density lipoprotein; IQ, Intelligence quotient; LDL, low-density lipoprotein; SBP, systolic blood pressure; Tri, triglyceriden; HOMA-IR, homeostasis model assessment of insulin resistance.
*$p < 0.05$ significant different evolution SIT vs CON or CAT vs CON
\$$p < 0.05$ significant evolution SIT vs CAT

Regarding variables associated with physical fitness (Table 2) SIT demonstrated a significant different evolution on peak VO_2, peak power, ventilatory threshold (W and L/min), 6-minute walk distance and MFR compared with no training ($P < 0.01$). Compared with CAT, there was a more pronounced increase in peak VO_2, peak power and ventilatory threshold (W and L/min) after SIT ($P < 0.01$).

Table 2. Influence of SIT and CAT on indices of physical fitness

Variables	SIT(n=17)		CAT(n=15)		Control(n=14)	
	Pre	Post	Pre	Post	Pre	Post
Physical Fitness						
Peak VO_2 (L/min)	2.4 (0.7)	2.6 (0.6)*$	2.5 (0.6)	2.4 (0.6)	2.3 (0.6)	2.2 (0.5)
Rel. peak VO_2 (ml/kg/min)	31.5 (5.2)	31.4 (4.8)	31.4 (6.8)	31.2 (6.6)	28.7 (5.7)	27.4 (4.6)
Peak Power (W)	155.0(36.6)	178.8(41.3)*$	179.0(42.6)	178.7(42.7)	166.8 (45.7)	158.9(46.8)
Peak HR (bpm)	178.4 (12.4)	175.5 (13.0)	181.3 (8.5)	174.3 (15.5)	180.3 (15.4)	177.9 (14.3)
VT (W)	99.7 (25.1)	120.6(32.2)*$	90.3 (24.5)	98.3 (20.3)*	86.1 (27.9)	82.9 (22.7)
VT (VO_2)	1.6 (0.5)	1.8 (0.5)*$	1.4 (0.3)	1.5 (0.3)	1.2 (0.3)	1.2 (0.3)
6 MWD (m)	598.2 (63.6)	665.9(69.4)*	538.7(105.0)	619 (72.2)*	567.0(69.4)	591.8 (82.7)
MFR (sec)	13.7 (7.5)	19.9 (6.8)*	19.5 (9.9)	22.5 (10.9)*	21.3 (13.8)	19.2 (11.4)
Sit-to-stand (amount/30 sec)	16.8 (4.0)	16.0 (3.4)	20.8 (6.4)	21.7 (6.5)	15.1 (4.4)	16.3 (2.8)

Data are presented as mean (standard deviation). Between group differences analysis of variance with post-hoc Bonferroni was used to evaluate time and interaction effects.
6MWD, 6-minute walk distance; MFR, muscle fatigue resistance; CAT: continuous aerobic training; SIT, sprint interval training; peakVO_2, Peak oxygen consumption; peak HR, peak heart rate; Rel peak VO_2, relative peak VO_2; VT, ventilatory threshold; *$p < 0.05$ significant different evolution SIT vs CON or CAT vs CON.
$$p < 0.05$ significant evolution SIT vs CAT

Discussion

The current study registered significant improvements in body composition (waist and fat percentage) after SIT compared with the control group and in fat percentage compared with CAT. Body composition management is an important concept in a population with ID as Salaun and Berthouze-Arende[25] recently demonstrated that the prevalence of obesity is high amongst adolescents with ID (average level of 37%). Any attempt to improve the body composition in an overweight population is welcomed, as negative chronic health consequences are well recognised[26]. Unfortunately many studies incorporating aerobic training[6,8,27,28] or combined aerobic and resistance training[27,29] have had no significant effects on body composition indices in a population with intellectual disabilities. Only one study reported small but significant weight loss with combined aerobic and resistance training[30]. The results of our study also did not reveal significant weight loss in either intervention groups. The variability of these results is difficult to explain as none of the studies controlled for dietary intake. One explanation is that after exercise training, satiety associated hormones are higher in overweight individuals[31]. If adolescents with ID have free access to food after training a negative impact on the overall outcome of body weight is a possibility. On the other hand, a decrease in waist circumference and percentage of body fat were observed in both intervention groups, but fat percentage was more pronounced in the SIT group. The effect of CAT is in line with a recent review of Alberga et al.,[32] reporting strong evidence that aerobic training decreases waist circumference and percent body fat in obese

adolescents. Only one study evaluated the effect of SIT in obese adolescents without ID. In this publication a positive effect on body composition was reported, but there was no difference between SIT and endurance training[19].

After SIT we observed significant improvement in resting systolic blood pressure, lipid profile (HDL, LDL, total cholesterol and triglycerides) and insulin sensitivity as measured by HOMA-IR compared with no training but when compared with CAT only systolic blood pressure (SBP) and LDL were more influenced by SIT ($P < 0.01$).

The effect of aerobic exercise training on systolic and diastolic blood pressure is well established. In a review by Cardoso et al.,[33] overall positive effects on systolic blood pressure, with the largest effect in those with hypertension, was observed. No effect on diastolic blood pressure was reported. Positive effects of interval training in adolescents without ID have also been reported in those with normotensive subjects[34]. Our results are in accordance with these results.

Concerning lipid profile, previously two other studies have demonstrated similar favourable results, but this was after combined aerobic and resistance training in overweight individuals with ID[13,14]. Studies in the general population have also revealed that SIT or a combination of aerobic and strength training is more beneficial on the lipid profile compared to aerobic training alone[15,35]. The effects of SIT on insulin sensitivity were recently demonstrated in a review to have moderate to strong favourable influences in a general population[36] and one study demonstrated it in adolescents with obesity without ID[19]. The variability of the results (+19% to +58%) in these studies could be explained by differences in SIT protocol, length of the intervention program and means of insulin sensitivity measurement (fasting insulin, HOMA-IR or glucose tolerance test).

Dipietro et al.,[37] concluded in their study (comparing the effects of different training intensities on insulin sensitivity) that the effects of low to moderate intensity training were lower than those of high intensity exercise. Another study that specifically compared the effects of IT and CAT in a population of young women demonstrated a 31% decrease in fasting insulin levels ($P < 0.01$) compared to the 9% of CAT (which was not significant)[38]. The results of our study revealed a significant decrease of 21% in fasting insulin levels (SIT) compared to 8% (CAT).

The SIT group demonstrated significant improvements in peak VO_2 and peak power compared with control and CAT. Also the ventilatory threshold (expressed as watts and L/min) was increased more in the SIT group compared with the control and CAT group ($P < 0.01$).

This is similar to studies in the general population[18,39-41] who also compared the effects of interval to continuous training.

Poole and Gaesser[42] demonstrated similar findings regarding the VT in which group 1 and 2 performed continuous training at 50% and 70% of VO_2 max respectively and group 3 performed SIT, three days a week for eight weeks. All three groups demonstrated significant improvements in the VT but the increase of SIT group was superior to the other groups. Enhancement of the VT is important as it improves submaximal exercise performance[42], functional performance[43] and quality of life[43].

Tjonna et al.[41] and Ciolac et al.[39] attributed this finding to improved central and peripheral physiological benefits such as cardiac function and mitochondrial capacity. Likewise, Daussin et al.[44] established that both mitochondrial oxidative capacity and capillary density increased only with SIT and not CAT. Trapp et al.[38] also showed that the SIT group improved mitochondrial capacity by 31% after 15 weeks of training. Unfortunately these variables were not measured in the current study. Considering adolescents and young adults with ID, significant improvements in VO_2 peak were reported but of small magnitude when combined aerobic and resistance was used[27,30]. On the other hand, studies incorporating continuous aerobic training only found small improvements in VO_2 peak but with no significance[7,8].

Peak power also improved significantly by 24 W in the SIT programme with no associated increase after the CAT protocol. This was also the case in both studies by Elmahgoub et al.[13,14] and the study by Calders et al.[27] who reported significant increases in peak power with combined aerobic and resistance training, but not after aerobic training, only in a population of intellectually disabled individuals.

Concerning the functional tests, 6-minute walk test and MFR ameliorated significantly more in the SIT group compared to control, but not to CAT.

A study by Calders et al.[27] previously revealed that a combined aerobic and resistance training program elicited greater benefits not only in the six-minute walk test, but also in the muscle fatigue test compared with aerobic training alone in a population of intellectually disabled individuals. They explained that the combined exercise program afforded improved quadriceps muscle strength and cardiovascular fitness. The six-minute walk test is easy and save to perform, inexpensive, field based, and it reflects one's ability to take part in everyday living activities and functional capacity[22]. The benefits of CAT and SIT shown in the current study are especially important in an overweight population with intellectual disabilities who have poor functional ability[3,29].

Finally, the functional sit-to-stand test revealed no significant improvements in the intervention groups. This can be expected as the sit-to-stand test is highly correlated with the strength of the lower legs[23]. Resistance training was not performed in either of the intervention groups.

Strengths, limitations and future studies

This is the first randomised controlled trial to investigate the effects of SIT in adolescents and young adults with ID. The participants were trained by knowledgeable, experienced and enthusiastic physiotherapists who carefully directed training sessions. As a result we obtained very accurate and favourable results with no adverse events and excellent program compliance. A possible limitation of the current study was that a strict randomised controlled trial was not possible. As strong heterogeneity exists in this population, the matching for different criteria was necessary. As such not all of the participants had an equal chance to be allocated to one of the three groups and could have biased the experimental groups. Furthermore, the participants in the study excluded those with severe musculoskeletal, neurological, and cardiovascular problems and care should be taken to generalise the results of this study. Also, most of the tests applied in this study have previously been applied in other scientific studies within this population but the validity and reliability of, for example, the MFR test have not been proven for participants with ID. However,

performances had increased after the training modalities and therefore we can conclude that training elicited beneficial effects. Finally the number of participants in each group is small (n = 14 to n = 17) and this may affect the statistical power of the various analyses. As a priori we did not nominate one primary outcome. The purpose was to focus on different aspects of body composition, physical and metabolic fitness.
The outcome of a future study exploring the effects of SIT combined with progressive resistance training and its relationship to function, health, performance and cardiovascular endurance will provide valuable insight and possible additional training recommendations for this population group. Also a study exploring the effects of an aerobic interval versus SIT may provide differences in practical findings. The same study explored in more homogenous subpopulations with ID, e.g. Down syndrome or fragile X-syndrome (whom is known to have poorer cardiovascular fitness or mitochondrial dysfunction compared with individuals with ID without DS or fragile X-syndrome) may also provide valuable feedback.

Conflict of interest

The author declares that there is no conflict of interest.

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CHAPTER 6

Effect of continuous aerobic vs interval training on selected anthropometrical, physiological and functional parameters of adults with Down syndrome

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Abstract

Background: A large percentage of adults with Down Syndrome (DS) are overweight and have extremely low aerobic capacities compared to the general population and persons with intellectual disability (ID) without DS. Previous aerobic training intervention studies showed limited potential to significantly ameliorate anthropometrical and cardiovascular variables. The primary purpose of this study was to determine the effect of continuous aerobic training (CAT) versus interval training (IT) on selected anthropometrical, health, physical and functional parameters of adults with DS. **Methods:** Forty-two adults with DS (25 men and 17 women) and a mean age of 33.8 (± 8.6) years were randomly allocated to one of three groups (IT, CAT, control). Training was performed for 12 weeks. The IT group performed ten-30 second all out sprints with 90 seconds (1:3 work-rest ratio) of low cadence, low intensity cycling or walking. The CAT group performed continuous cycling and walking at an intensity of 70 - 80% of VO_2 peak. Heart rate monitors were used for monitoring training intensities. After 6 weeks of training, the intensity of the CAT was increased to 85% of VO_2 peak, whilst the intensity of the interval training group remained "all out". An increase of 5 minutes in duration was implemented after 6-weeks for both training groups. To evaluate pre–post differences between groups, a repeated analysis of covariance (ANCOVA) with post-hoc Bonferroni test was performed. **Results:** After 12 weeks of training, body weight and BMI decreased significantly more in the IT group compared with control and CAT ($p<0.05$). Participants in the IT group decreased their body weight from 71.4±8 to 69.4±8 kg and their BMI from 29.3±4 to 28.5±4 kg/m2. Significant ameliorations for functional parameters and leg strength were shown for CAT compared to control ($p<0.05$). Participants in the CAT group improved their performance in the 6MWD (499±78 to 563±75 m), 8-foot up & go (5.9±1.2 to 4.8±0.9) and leg strength (13.1±2 to 15.2±2). VO_2 peak and time to exhaustion significantly improved in both the IT and CAT group compared to control ($p<0.01$). Moreover, a significant improvement for relative VO_2 peak was also determined for IT compared to CAT ($p<0.05$). Participants in the IT group increased their VO_2 peak from 32±8 to 37±8 ml/min/kg. Submaximal heart rate and VO_2 values improved significantly within both exercise groups ($p<0.05$). **Conclusion:** IT and CAT can both be pursued by adults with DS to positively impact on various parameters of anthropometry, fitness and functional ability, with IT more appropriate for improving body weight and aerobic capacity.

Introduction

Down syndrome (DS) occurs when there is a full or partial extra copy of the 21st chromosome. Individuals with DS are born with many health related conditions such as congenital heart disease, ligamentous laxity and muscle hypotonicity (Abbag, 2006; Carmeli *et al*. 2002[a];). A large majority of individuals with DS live sedentary lifestyles, are overweight and exhibit functional limitations and extremely poor aerobic capacities (Nordstrom *et al*. 2013; Terblanche & Boer, 2013; Esposito *et al*. 2012; Cowley *et al*. 2010; Shields *et al*. 2009; Baynard *et al*. 2008). The management of body composition is an important concept as Terblanche & Boer (2013) indicated that in South Africa 79% of men and 95% of women with DS are overweight (n=371). Furthermore individuals with DS have an increased risk of obesity compared to individuals with ID without DS (Melville *et al*. 2008). This increased risk may stem from an inactive lifestyle which originates in childhood (Pitetti *et al*. 2013; Espositio *et al*. 2012). Improving variables associated with body composition in this population is clinically significant as associated chronic health conditions are well recognised (Torr *et al*. 2010; Rubin *et al*. 1998). Most individuals with DS experience accelerated ageing for which reasons are unknown but could be related to the genes on chromosome 21 or a speeded decay in functional fitness (Down syndrome South Africa (DSSA), 2014; Terblanche & Boer, 2013). Torr *et al*. (2010) also reported a functional decline in this population with an early onset of age related diseases. The combination of these factors could contribute to a poor prognosis of functionality, quality of life and independence in later years.

Provisionally, many exercise interventions have been conducted with notable success in a DS population. Two studies with an aerobic exercise intervention demonstrated significant improvements in variables such as time to exhaustion and work capacity on maximal exercise tests, however they have failed to show an improvement in VO_2 peak, loss in body weight or a loss in body fat percentage (Varela *et al*. 2001; Millar *et al*. 1993). González-Agüero *et al*. (2010) and Casey et al. (2014) stipulated that perhaps the training intensity and/or volume may not provide a sufficient stimulus for improved aerobic capacity or loss in body weight. However, an improved aerobic capacity has been shown in a study with a longer intervention period (28 weeks) and higher exercise intensities (60-85% of VO_2 peak) (Mendonca & Pereira, 2009).

Training strategies with exercise intensities greater than 85% of VO_2 peak has not been reported in a DS population. Interval training involves high intensity training interspersed with recovery periods of complete rest or lower exercise intensities. IT has been shown to purport fewer motivational problems and more perceived enjoyment in the general population (Bartlett *et al*. 2011). This exercise strategy has been used with significant effect across a wide variety of populations even in persons with chronic health conditions (Smart & Steele, 2012; Moholdt *et al*. 2009). Tjønna *et al*. (2009) demonstrated that several key cardiovascular risk factors can be reduced with IT in obese individuals in the general population. Furthermore, significant anthropometrical (waist circumference, body fat %), metabolic (lipid profile, fasting insulin) and physical improvements (VO_2 peak, ventilatory threshold, peak power) have been shown

in a population of adolescents and young adults with ID but without DS (Boer *et al*. 2014). In addition, the benefits seen with IT has matched or outweighed many of the benefits studied with continuous aerobic training (CAT) even though IT was less time consuming in some of these studies (Boer *et al*. 2014; Hwang *et al*. 2011; Burgomaster *et al*. 2008).

Against the background of the current literature, the primary purpose of this study was to ascertain the effect of continuous aerobic training versus interval training on selected anthropometrical, health, physical and functional parameters of adults with Down syndrome in South Africa.

Methods

Participants

Forty-four individuals with DS participated in this study. Study participants were recruited from three care centres for persons with intellectual disability in South Africa. The legal guardians of the participants gave consent for participation in the study. Participants had to be between 18 and 50 years to participate in the study. All participants' had to successfully complete the adapted physical activity readiness questionnaire (aPARQ). This meant that all questions had to be answered as "no". If "yes" was answered at any question/s permission from the individual's physician had to be obtained. All participants lived at the intellectually disabled centre and had no cognitive or physical impairments that prevented them from completing all the exercise tests and training protocol. The study was approved by the ethics committee of the North West University (NWU-00064-14-A1).

The participants were randomly allocated to one of three groups by picking a card out of a hat indicating the allocated group. The group sizes and drop-outs are schematically demonstrated in a flow chart (Fig 1). All participants completed baseline tests over a three day period. Prior to baseline testing, participants were familiarised with all tests and procedures on two separate occasions one to two weeks prior to testing. All tests were performed indoors in a spacious, temperature controlled and well ventilated room. Prior to day 1 of exercise testing all participants were requested to fast overnight for peripheral glucose and total cholesterol tests. In the morning of day 1, body weight, height, waist and hip circumference, blood pressure and body composition measurements were determined.

Measurements

Body weight was measured with a calibrated electronic scale (Beurer, Germany) to the nearest 0.1 kg. Participants wore light weight clothing, no shoes and emptied their bladders prior to measurement. Height was determined with a sliding steel stadiometer to the nearest 0.1 cm (Siber-Hegner GPM, Switzerland).

Waist circumference was measured at the umbilicus with the participant standing upright after a normal expiration with a flexible steel tape (Lufkin, Cooper Tools, Apex, NC). Hip circumference was measured

across the broadest part of the hips and was recorded to the nearest cm (Marfell-Jones *et al.* 2007). After the anthropometry measurements, the resting blood pressure was determined.

Resting systolic and diastolic blood pressure was taken seated on a chair with the upper arm at the level of the heart resting on a level table. Blood pressure was measured with a manual sphygmomanometer (MicroLife, Widnau, Switzerland). A resting measurement was determined after the participants were seated for at least 5 minutes. Two measurements were taken with a 5-minute interval in between of which the average of the two measurements was recorded as the resting blood pressure. Peripheral blood samples were obtained with a finger prick after an overnight fast and the blood pressure measurements. Total cholesterol and glucose was analysed using diagnostic kits (Roche Diagnostics Mannheim, Germany).

Body composition was assessed by bioelectrical impedance analysis (BIA) (Bodystat 1500 MDD, Douglas, Isle of Man, UK). Subjects were in the supine position for exactly five minutes. All procedures were carried out according to the standardised procedures as outlined in Gonzalez-Correa & Caicedo-Eraso (2012). Previous experimental studies also used BIA as a measurement of fat mass and fat free mass in individuals with ID (Boer *et al*. 2014; Salaun & Berthouze-Aranda 2012) and Down syndrome (Mendonca *et al*. 2011). BIA demonstrated adequate test-retest reliability in 20 individuals with DS in our pilot study (r=0.97).

After breakfast, testing continued with the hand grip strength ((Takei, Grip D,T.K.K 5401; Niigata City, Japan), sit-to-stand, 8-foot up-and-go test and the 6 minute walk distance test (Rikli & Jones 2013). For the hand grip strength test the participant squeezed a hand grip dynamometer (Takei, Grip D, T.K.K 5401; Niigata City, Japan) with the dominant hand with as much force as possible. Three trials were administered with at least 30 second rest periods between trials. Test-retest reliability of adults with ID was strong (ICC = 0.94) (Hilgenkamp *et al.* 2012:160) and in adolescents with DS (ICC = 0.86) (Tejero-Gonzalez *et al*. 2013:3221) and in adults with DS (ICC = 0.97) (Boer & Moss, 2016).

After the hand grip strength test the sit-to-stand test was conducted to evaluate lower body strength which is needed for many everyday living activities such as climbing stairs and getting out of a chair or car. The test assesses the amount of full stands from a seated position that can be completed in 30 seconds. One trial is administered. Test-retest reliability in adults with ID was moderate to strong (ICC = 0.72) (Hilgenkamp *et al.* 2012:160) and an ICC value of 0.94 in adults with DS (Boer & Moss, 2016).

This test was followed by the 8-foot up-and-go test to determine the agility and dynamic balance in important everyday living tasks that require quick movements such as getting off the bus in time. The test assesses how quickly one can get out of a seated position, walk 2.4 meters, and return to the seated position. Two trials were administered. Test-retest reliability of this test-item revealed and ICC value of 0.95 in the general elderly population (Rikli & Jones, 2013:37) and an ICC value of 0.95 in adults with DS (Boer & Moss, 2016).

Finally for day 1, the self-paced 6-minute walk test (6MWD) was performed. Participants were instructed to walk as many laps of 45.7 meters (50 yards) each as possible within the allocated six minutes. This test assesses aerobic endurance and functional ability important for many everyday living activities. Test-retest reliability of this test-item revealed and ICC value of 0.94 in the general elderly population (Rikli & Jones, 2013:37) and an ICC value of 0.93 in adults with DS (Boer & Moss, 2016). Repeated analysis of variance showed no significant difference between tests in a group of Down syndrome adolescents provided that two practise sessions were given (Casey et al. 2012:2068). Participants had a 20 minute break between each of these tests. On day two, the participants performed no formal physical activities.

During the morning of day 3 the VO_2 peak test (Metalyzer 3B system (Cortex, Leipzig, Germany) was performed. The test was carried out on a motorised treadmill using the standardised protocol for adults with DS. This protocol starts at an initial velocity of 4 km/h at 0% incline. After every 2 minutes, the incline increases by 2.5% until an incline of 12.5% is reached. If the participant reaches this stage, the speed is increased by 1.6 km/h every minute until exhaustion. This protocol is used widely in scientific research concerning participants with DS (Mendonca *et al*. 2011; Guerra *et al*. 2003), and is considered valid and reliable in a DS adult population provided that adequate familiarisation sessions are conducted (Fernhall *et al*. 1990). Respiratory measurements and heart rate were performed using the metalyzer 3B system (Cortex, Leipzig, Germany). An ECG was recorded during the VO_2 peak test (Custo Med, Schiller, Switzerland). The VO_2 peak was taken as the highest recorded measurement during the final 30 seconds of the tests (after the data was filtered to every 10 seconds) with an RER greater than 1.0 (American Thoracic Society & American College of Chest Physicians 2003; Goulopoulou *et al*. 2006; Baynard *et al*. 2004). Participants were asked and continually encouraged to perform the test until maximal exhaustion as described by Varela *et al*. (2001). The test was stopped if any clinically significant ST segment deviations, arrhythmias, chest pain or dizziness arose or when they requested to terminate the test. The same tests and procedures were followed after the 12-week intervention period. Participants were maximally motivated during all tests as previously described by Varela et al. (2001) and Millar et al. (1993) with standardised phrases of encouragement provided every 15 seconds (Nasuti et al., 2013).

Training intervention

All three groups continued with their normal everyday activities during the intervention period. Participants in the continuous and interval training groups trained three times a week for 12 weeks under the supervision of a sport scientist and biokineticist (accredited exercise physiologist) in a ratio of 2:6. Participants alternated the training sessions between treadmill and cycle ergometer training for the duration of the intervention period (50% of training sessions on treadmill (Johnson T8000, Taichung, Taiwan) and 50% of training sessions on the cycle ergometer (R7000, Taichung, Taiwan) due to a lack of treadmills available. Participants attended the exercise sessions in groups of six. Participants were blinded for the continuous and interval interventions. Both training groups warmed up for 5 minutes (4 km/h or 60 W), performed 20 minutes of training and cooled down for 5 minutes (4 km/h or 60 W). The interval training group performed

ten- 30 second all out sprints with 90 seconds (1:3 work-rest ratio) of low cadence, low intensity walking or cycling. The CAT group performed continuous cycling or walking at an intensity of 70 to 80% of VO_2 peak. Heart rate monitors (Polar, Kempele, Finland) was used to monitor training heart rate intensities. After 6 weeks of training, the intensity of the continuous training was increased to 85% of VO_2 peak, whilst the intensity of the interval training group remained "all out". Additionally, the duration of the exercise sessions also increased by 5 minutes for both training groups whereby the interval group performed three more sprints. Participant attendance was recorded after every exercise session. The measurement of all outcome variables were performed by blinded assessors.

Statistical analysis

It should be noted that the study followed an "intention to treat" and not the "effect of treatment" procedure. All data were analysed with a commercially available statistical software program (Statistical Package for the Social Sciences, SPSS 20.0, SPSS Chicago, IL, USA). The distribution of the variables was assessed with Kolmogorov-Smirnov test. Data are expressed as mean and standard deviation (SD). To evaluate pre–post differences between groups, a repeated analysis of covariance (ANCOVA) with post-hoc Bonferroni test was performed (group x time interaction effects) and adjusting for baseline values. Data was screened to analyse whether ANCOVA assumptions were violated.

Results

Fifty-six individuals with DS volunteered for the study. Ten individuals were excluded after the screening and familiarisation procedures as their legal guardians did not adequately complete their consent forms. After the 12 weeks of exercise training only four individuals withdrew from the study. No serious or adverse events transpired during the baseline, intervention period or post-intervention testing. IT was purported to be fun and safe by all adult participants with DS in the current study. The assumption of homogeneity of variances (Levene's test) was violated for body mass index (BMI), cholesterol, minute ventilation, and hand grip strength and assessed with the Kruskal Walis non-parametric test.

Participants attended a minimum of 28 and a maximum of 36 exercise sessions. The average percentage compliance to the IT intervention was 95% and the CAT intervention 96% respectively. Exercise sessions missed were due to illness or personal commitments. There were no significant difference between the two groups for exercise sessions attended.

There were no missing values in any of the data sets and no differences between groups for any of the variables at baseline as shown in Table 1 and 2. Results of ANCOVA with each variable (as the difference between pre and post), degrees of freedom, F-stat and significance are reported in Table 3.

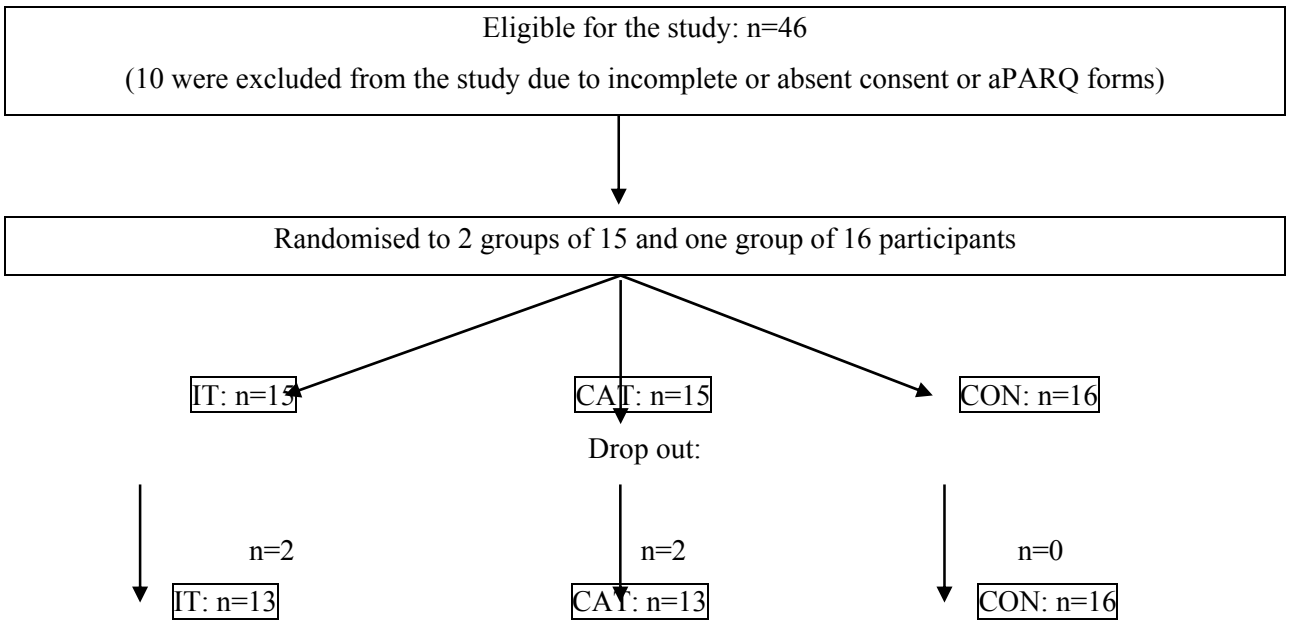

Fig. 1: Flow diagram: Participants' flow diagram depicting the matching and randomization procedure during the trial. Drop out numbers are also indicated which were death in the family (n=1), severe sickness (n=2) and attendance of insufficient training sessions due to an extended holiday (n=1). IT: interval training, CAT: continuous aerobic training; CON: control; n: number.

After 12 weeks of physical training, body weight decreased significantly more in the IT group compared with no training and CAT ($p<0.05$). A significant decrease in BMI was also found with IT as determined by a Kruskal Walis test ($p<0.01$). No significant between group changes were observed for fat mass, fat percentage, waist or hip circumference in either of the training groups (Table 1). Similarly, no significant changes were noted for fasting blood glucose, total cholesterol, systolic and diastolic blood pressure in either of the training groups compared to the control group (Table 1).

Table 1. Influence of IT and CAT on body weight, body composition, blood pressure, glucose and total cholesterol

Variables	IT(n=13)		CAT(n=13)		Control(n=16)	
	Pre	**Post**	**Pre**	**Post**	**Pre**	**Post**
General						
Age	30.0 (7.4)	30.1 (7.4)	34.2 (9.2)	34.4 (9.2)	36.6 (8.4)	36.9 (8.4)
Gender Male/Female	8/5	8/5	7/6	7/6	10/6	10/6
Anthropometry						
Weight (kg)	71.7 (8.4)	69.4 (8.3)*$	70.2 (14.6)	69.2 (14.6)*	74 (8.4)	74.1 (8.4)
Height (cm)	156.7 (7.5)	156.8 (7.5)	151.4 (6.6)	151.3 (6.6)	155.4 (7.8)	155.5 (7.8)
BMI (kg/m^2)	29.3 (4.0)	28.5 (4.0)*$	30.6 (6.1)	30.2 (6.3)	31.2 (4.6)	30.9 (4.2)
Waist circumference (cm)	94.2 (8.1)	93.8 (8.0)	95.0 (11.1)	93.7 (11.9)	99.4 (10.9)	98.0 (10.6)
Hip (cm)	101.3 (10.0)	97.6 (10.5)	102.7 (11.8)	100.9 (11.6)	105.1 (12.4)	103.1 (10.5)
W:H						
Fat mass (kg)	18.1 (7.6)	16.7 (7.2)	22.3 (8.8)	21.1 (9.0)	21.3 (9.1)	23.6 (14.5)
Fat (%)	24.7 (10.1)	23.4 (9.6)	30.4 (10.2)	29.6 (10.2)	27.8 (11.8)	27.1 (11.8)
Blood pressure						
SBP (mmHg)	117.0 (19.5)	104.9 (12.6)	111.1 (17.4)	103.3 (16.9)	123.0 (20.0)	117.3 (17.1)
DBP (mmHg)	73.8 (8.8)	65.9 (18.2)	64.8 (7.9)	61.3 (10.6)	74.4 (11.9)	66.7 (11.7)
Blood profile						
T-Chol. (mg/dl)	223.4 (25.8)	225.5 (28.5)	210.0 (38.6)	216.2 (48.4)	214.9 (30.9)	222.9 (30.6)
Glucose (mg/dl)	95.5 (14.4)	88.3 (10.8)	90.1 (9.0)	90.1 (10.8)	95.5 (10.8)	93.7 (10.8)

Data are presented as mean (standard deviation). Between groups ANOVA with post hoc Bonferroni was used to evaluate time and interaction effects.
IT, interval training; CAT, continuous aerobic training; BMI, body mass index; T-Chol, Total Cholesterol; DBP, diastolic blood pressure; SBP, systolic blood pressure.
*p <0.05 significant different evolution IT v Control or CAT v control; $ p<0.05 significant change IT v CAT

Results of the functional parameters (6MWD, 8 foot up-and-go) and leg strength (sit to stand) indicated significant ameliorations in the CAT group only when compared to the control group ($p<0.05$) (Table 2). Variables associated with physical fitness, both aerobic capacity (absolute and relative VO_2 peak) and time to exhaustion significantly ameliorated improvement in both the IT and CAT group compared to the control ($p<0.01$) (Table 2). Moreover, a significant improvement for relative VO_2 peak was also determined for IT compared to CAT ($p<0.05$). Table 4 and 5 illustrates that submaximal heart rate and VO_2 values improved significantly within both exercise groups after 12 weeks of training.

Table 2. Influence of IT and CAT on indices of physical fitness and functional ability

Variables	IT(n=13)		CAT(n=13)		Control(n=16)	
	Pre	Post	Pre	Post	Pre	Post
Physical Fitness						
Peak VO_2 (L/min)	31.9 (7.9)	37.3 (7.9)*$	32.2 (7.1)	34.4 (7.5)*	32.1 (7.1)	30.7 (6.1)
Rel. peak VO_2 (ml/kg/min)	2267.1 (578.9)	2578.3* (508.3)	2200.6 (457.5)	2312.3* (447.3)	2363.1 (533.1)	2271.2 (499.9)
VE (L/min)	67.8 (17.0)	88.8 (19.4)*	67.0 (15.4)	80.0 (13.3)	69.9 (15.9)	72.9 (15.2)
Time to exhaustion (s)	686.8(180.7)	845.4 (86.3)*	700.5 (170.2)	813.1(79.4)*	706.8 (142.0)	699.1(134.8)
Peak HR (bpm)	167.1 (10.0)	171.3 (9.0)	162.5 (12.6)	162.0 (11.2)	160.8 (12.9)	159.4 (10.4)
6 MWD (m)	519.7 (82.8)	562.6 (81.7)	499.0 (77.5)	563.2(74.9)*	476.2 (83.5)	495.9 (85.2)
HGS (kg)	27.9 (8.8)	29.9 (8.9)	23.3 (6.9)	26.1 (7.9)	23.8 (9.1)	25.5 (9.1)
8-foot up and go (s)	5.8 (1.9)	4.9 (1.1)	5.9 (1.2)	4.8 (0.9)*	6.5 (1.3)	6.2 (1.3)
Sit-to-stand (amount/30 sec)	14.4 (2.3)	15.5 (1.8)	13.1 (2.0)	15.2 (1.8)*	13.1 (2.7)	13.3 (2.3)

Data are presented as mean (standard deviation). Between groups ANOVA with post hoc Bonferroni was used to evaluate time and interaction effects.
6MWD, 6 minute walk distance; HGS, Hand grip strength; CAT, Continuous aerobic training; IT, Interval Training; peakVO_2, Peak oxygen consumption; peak HR, peak heart rate; Rel peak VO_2, relative peak VO_2; VT, ventilatory threshold; *$p < 0.05$ significant different evolution IT v Control or CAT v control
$p < 0.05$ significant evolution IT v CAT

Table 3: Results of the analysis of covariance with each variable (as the difference between pre and post), degrees of freedom, F-stat and significance

Variables (as deltas)	DF	F	Significance
Body weight (kg)	41	13.8	0.000*
BMI (kg/m^2)	41	0.7	0.000*
Waist circumference (cm)	41	0.3	0.8
Hip circumference (cm)	41	1.0	0.4
Fat mass (kg)	41	1.1	0.4
Fat percentage (%)	41	1.4	0.3
Systolic blood pressure (mmHg)	41	0.5	0.6
Diastolic bloop pressure (mmHg)	41	0.4	0.7
Total cholesterol (mg/dL)	41	1.3	0.3
Glucose (mg/dL)	41	0.8	0.4
Peak VO_2 (L/min)	41	13.3	0.000*
Relative peak VO_2 (ml/kg/min)	41	17.8	0.000*
VE (L/min)	41	6.5	0.004*
Time to exhaustion (s)	41	11.0	0.000*
6 MWD (m)	41	6.2	0.01*
Hand grip strength (kg)	41	0.6	0.57
8-ft up and go (s)	41	3.7	0.03*
Sit-to-stand (amount/30 s)	41	6.2	0.01*

*$P < 0.05$ significant different evolution between groups.

Table 4: Submaximal heart rate responses (bpm) during the maximal exercise test before and after the intervention period

Level	IT		CAT		Control	
	Pre	Post	Pre	Post	Pre	Post
4km/h @ 5%	129 (14)	120 (14)*	125 (19)	116 (19)*	128 (14)	126 (14)
4km/h @ 7.5%	135 (15)	127 (15)*	129 (16)	122 (17)*	135 (14)	135 (14)
4km/h @ 10%	143 (14)	131 (14)*	138 (17)	128 (18)*	143 (14)	142 (15)
4km/h @ 12.5%	149 (13)	137 (14)*	145 (19)	132 (19)*	147 (15)	146 (15)

*$p < 0.05$ significant different evolution (pre vs post within)

Table 5: Submaximal VO_2 responses (bpm) during the maximal exercise test before and after the intervention period

Level	IT		CAT		Control	
	Pre	Post	Pre	Post	Pre	Post
4km/h @ 5%	24 (3)	21 (3)*	22 (2)	20 (2)*	22 (2)	22 (2)
4km/h @ 7.5%	25 (4)	22 (3)*	24 (2)	22 (2)*	25 (3)	25 (3)
4km/h @ 10%	27 (4)	25 (3)*	25 (3)	23 (2)*	26 (3)	26 (3)
4km/h @ 12.5%	29 (4)	26 (4)*	27 (2)	24 (2)*	28 (3)	28 (3)

*$p < 0.05$ significant different evolution (pre vs post within)

Discussion

The primary aim of the current study was to determine the effect of IT versus CAT on selected anthropometrical, health, physical and functional parameters of adults with Down syndrome compared with a control group.

More than half of the participants in the current study were overweight and obese (54.8%) as commonly reported in persons with DS (Terblanche & Boer, 2013). The main finding of the intervention study is that IT improved body weight and BMI significantly compared to the CAT and control group. Studies in the general population also reported a significant improvement in body weight and BMI with IT (Heydari *et al*. 2012; Boutcher, 2010; Tjønna *et al*. 2008; Warburton *et al*. 2005). Moreover, IT seems to be a more effective way compared to less intense continuous types of exercise to reduce body weight in adolescents and adults in the general population (Tjønna *et al*. 2008; Trapp *et al*. 2008; Gutin *et al*. 2005). Most studies using CAT showed no improvement in body or fat mass in a DS population (Andriolo *et al*. 2011; Tsimaras *et al*. 2003; Varela *et al*. 2001), which was confirmed in a review (Dodd & Shields 2005). Another review by González-Agüero *et al*. (2010) stipulated that perhaps the training intensity and/or volume did not provide a sufficient stimulus for loss in body weight in a DS population. Boutcher (2010) highlighted a few possible mechanisms involved with an increased fat or body weight reduction after IT. Firstly IT has discernible increases in body and skeletal muscle fatty acid oxidation. Secondly, IT may inhibit appetite. High intensity exercise has been reported to reduce food intake in rats by facilitating the release of

corticotropin (Bilski *et al*. 2009). Lastly, the post exercise physiological response to exercise may influence post exercise fat metabolism possibly through the influence of heightened circulating catecholamines and assist in the removal of excess lactate, H+ and resynthesis of glycogen.

The current study reported only five individuals with DS having a SBP higher than 140 mmHg. The mean SBP for the entire group was 117 mmHg, conferring previous reports of adults with DS having lower systolic and diastolic blood pressure compared to their peers in the general population (Draheim et al., 2002). Blood pressure did not improve significantly in either of the groups compared to control similar to the study by Wallman *et al*. (2009) who also compared IT and CAT in a study that included diet education as well. This finding was also confirmed in a meta-analysis by Hwang *et al*. (2011) with the authors suggesting that a longer training period may be needed to induce chronic vasodilation. The effect of exercise training on resting blood pressure is less influential when performed on normotensive individuals (Belardinelli et al., 1999) as to hypertensive individuals (Ishikawa-Takata et al., 2003).

Fasting glucose and total cholesterol values of the majority of participants were within the normal range. Studies comparing total cholesterol concentrations between individuals with DS and the general population reported no significant differences (Pueschel *et al*. 1992). No significant improvements in fasting peripheral glucose and peripheral total cholesterol were seen in the current study. Walman *et al*. (2009) also showed no differences pre and post between groups (IT, CAT and control) for fasting glucose and total cholesterol values. The absence of changes is attributed to the largely normative values found in their study. The fact that fasting glucose decreased with IT in the study by Tjønna *et al*. (2009) may be attributed to the reported lower intake of carbohydrates. The current study however did not control for dietary intake.

An improvement in functional ability related to activities of daily living for a population of adults with DS is clinically significant as many individuals suffer from premature ageing and dependence when they age (Terblanche & Boer, 2013). The 6MWD and 8-foot up-and-go tests is easy and safe to perform and reflects one's ability to take part in everyday living activities (Rikli & Jones 2013; Elmahgoub *et al*. 2012). Both training groups significantly improved their performance on the 6MWD and 8-foot up-and-go within groups but only the CAT group reported significant improvement compared to the control group. CAT resembles the uninterrupted nature of the 6MWD test more closely and may be the reason for this finding. A recent study performed on individuals with ID reported significant improvements in the 6MWD for both IT and CAT, with the CAT group reporting more pronounced changes (+80.3 meters versus +67.7 meters) (Boer *et al*. 2014). Other populations with serious medical conditions can also benefit from IT as shown by an improvement in the distance walked on the 6MWD test in patients with chronic heart failure (Nilsson *et al*. 2008). Importantly, improvements in functional capacity were associated with an improvement in the quality of life.

With respect to the two strength tests (hand grip and sit-to-stand), no significant ameliorations were reported for hand grip strength but leg strength significantly improved in the CAT when compared to the control group. It is possible that the longer continuous cycling and walking sessions may be accountable for this finding compared to IT. However, both training groups demonstrated significant improvements within groups for leg strength. Carmeli *et al*. (2002[b]) reported that lower limb strength (assessed by an isokinetic dynamometer) was significantly improved as a result of CAT (walking on a treadmill) for 6 months in adults with DS. Improvement in lower limb muscle strength is important as this variable represent functional performance and everyday living activities in adults with DS (Terblanche & Boer, 2013; Cowley *et al*. 2010). Previous studies indicate that individuals with DS have reduced muscle strength compared to the general population and more specifically leg strength when compared to both the general population and individuals with ID without DS (Carmeli *et al*. 2002[a]; Croce *et al*. 1996; Pitetti *et al*. 1992).

All participants in the current study reached a VO_2 peak as an RER of 1.1 was reached (American Thoracic Society & American College of Chest Physicians 2003; Goulopoulou *et al*. 2006; Baynard *et al*. 2004; Fernhall & Otterstetter 2003; Fernhall et al., 2001). The VO_2 peak improved significantly in both exercise groups (IT and CAT) when compared to the control group. VO_2 peak with IT improved more significantly when compared to the CAT group similar to findings reported in the general population (Ciolac *et al*. 2011; Hwang *et al*. 2011) and those with ID (Boer *et al*. 2014). It has been purported that IT improves the pumping capacity (stroke volume) of the heart more than CAT (Tjønna *et al*. 2009; 2008; Daussin *et al*. 2008). Rest periods between work intervals allow individuals to work at higher intensities, when compared to continuous exercise. Concerning peripheral adaptations, mitochondrial capacity and CA^{2+} cycling have been shown to improve to a larger extent with IT compared to CAT (Daussin *et al*. 2008; Tjønna *et al*. 2008). Trapp *et al*. (2008) demonstrated that mitochondrial oxidative capacity improved by as much as 31% after 15 weeks of IT. Improvement in mitochondrial oxidative capacity was also shown by Heydari *et al*. (2012), Burgomaster *et al*. (2008) and Gibala & McGee (2008) which resulted from an improved concentration of oxidative enzymes (citrate synthase, cytochrome oxidase & 3-hydroxyacyl CoA dehydrogenase).

Most other studies that have performed CAT on individuals with DS have yielded no significant improvement in VO_2 peak (Varela *et al*. 2001; Millar *et al*. 1993). A lack of improvement in these studies may be that the exercise intensity during training was too low (55-70% of peak VO_2 and 65-75% of maximum heart rate) and were monitored irregularly. The sample size in the study by Millar *et al*. (1993) was very small (n=14). Also, training in the study by Varela *et al*. (2001) was performed as a non-weight bearing activity (rowing ergometery) and may be insufficient to adequately enhance physiological adaptations associated with training. It is known that individuals with DS suffer from chronotropic incompetence (Fernhall *et al*. 2009). To base training intensities on conventional percentages of peak heart rate (60-75%) may not be applicable to this population as participants will most likely train at intensities which are too low. For instance, the mean maximal heart rate in the current study was 162 bpm. Using 65%

and 75% of maximal heart rate as the study by Millar *et al*. (1993) yields training zones of 105 to 122 bpm. These submaximal values are indicative of mean walking speeds at 4 km/h (0 to 2.5% incline) when compared to the current study. These are indicative of very low training intensities (level 1 and 2 of the VO_2 peak test). The only CAT intervention study that did improve VO_2 peak in adults with was performed for 28 weeks at an exercise intensity of 60 to 85% of VO_2 peak (Mendonca & Pereira, 2009). Consequently, we also centred our CAT intensities on values between 70 and 85% of peak VO_2 and found significant improvements in VO_2 peak with 12 weeks of exercise training (using running and cycling as training modalities).

The current study did not explore the effect of training on the ventilatory threshold (VT) as we did previously on individuals with ID (Boer *et al*. 2014). It has been reported by Baynard *et al*. (2004) that the detection of VT is very difficult in this population. However, we did analyse submaximal exercise intensities as Mendonca & Pereira (2009) previously demonstrated in adults with DS. Their participants improved their submaximal exercise capacity after 28 weeks of CAT. Likewise, we demonstrated that submaximal heart rate and VO_2 values improved significantly within both exercise groups after 12 weeks of training (Table 4 & 5). Enhanced performance in submaximal exercise intensity is important as individuals with DS may adapt more efficiently to increased walking speeds (Mendonca *et al*. 2010). It is also clinically significant as submaximal exercise performance is related to functional capacity and quality of life (Aoike *et al*. 2012; Poole & Gaesser 1985).

Conclusion

A 12-week IT or CAT stimulates various physiological processes whereby anthropometrical, health, functional and physical benefits are attained. IT elucidated more significant improvements in body weight, BMI and aerobic capacity whereas CAT provided functional and lower limb strength improvements. IT and CAT are feasible and safe options for adults with DS reflected by the absence of adverse events, low drop-outs and excellent compliance during training. It is therefore advised that adults with DS partake in both IT and CAT regimens which would also add diversity to training sessions.

Limitations and future studies

The level of ID could not be determined as the ID centres did not have any information pertaining to their intelligence quotient. However all participants in the current study fully understood instructions and information pertaining to test procedures, techniques and training protocols. It would be very worthwhile to analyse the level of ID as a cofactor during many of the exercise tests. However, in the study by Boer *et al.* (2014) no such effect was found in an ID population. Most of the adults with DS lived in residential care centres. As such, it is possible that the results of the current study may not be generalised to those living with their families in private homes.

A future study could explore the combined effect of IT and CAT in this population since benefits were obtained from both training strategies. This has been demonstrated in the general population where aerobic capacity resulted in a 41% improvement (Mourier *et al*., 1997).

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CHAPTER 7
SUMMARY, CONCLUSIONS, LIMITATIONS AND RECOMMENDATIONS

7.1 Introduction

Persons with an intellectual disability, including Downs syndrome present with high levels of inactivity, low fitness and a large prevalence of overweight and obesity. Together, these factors contribute to many health related problems and poor quality of life in this population. Non-communicable diseases such as hypertension, diabetes, dyslipidemia and the metabolic syndrome are more common in individuals with ID compared to the general population. The positive changes that regular exercise can bring about, has only received the attention of researchers during the last 10 years. However, the majority of the research focussed on addressing obesity and cardiorespiratory fitness. Continuous aerobic exercise interventions were mostly the point of departure of intervention studies. The results reported for continues aerobic interventions indicate a limited effect on improving variables such as cardiorespiratory endurance and body composition. Secondly, there is no standardised battery of test items specific to adults with DS to accurately determine functional fitness and match measurements against standardised normative values. In addition, the effect of exercise interventions cannot be monitored with regard to functional fitness either. In order to address these limitations, the purpose of the current study was to determine the reliability and validity of selected functional fitness tests in adults with DS. Also, to determine the effects of IT on various anthropometric, physical, metabolic and functional parameters in adults with ID and DS. This chapter provide a summary of the study, conclusions drawn from the hypotheses tested, limitations experienced in this study and recommendations for future research.

7.2 Summary

The first chapter of this thesis presented the problem statement with regard to functional fitness testing in persons with DS and the effect of exercise interventions in persons with ID and DS. The limited information available on the reliability and validity of a standardised functional test, and the lack of evidence on exercise induced changes in body composition and aerobic capacity, resulted in the formulation of the research question for this study, namely: “What is the reliability and validity of selected functional tests and the effect of aerobic continuous and IT exercise on the physical, metabolic and anthropometric profiles of persons with ID and DS? In order to answer the research question, a review of the literature was conducted and presented in Chapter 2 as: “Literature review: functional ability and exercise in persons with intellectual disability and Down syndrome”. From the literature review a paucity of information regarding a standardised battery of test items in a DS adult population was present. Even though such batteries exist in the general, elderly and ID populations, standardised functional tests are yet to materialise in a DS adult population. Most adults with DS could not perform these tests as shown in our pilot study. Unfortunately, individuals with DS are often pooled with ID individuals even though discernible differences in functional

fitness exist. In a previous study the feasibility of various functional fitness tests were demonstrated in adults with DS (n=371) in South Africa (Boer, 2010). Tests that analyse balance, flexibility, muscular strength, cardiovascular endurance and functional ability were conducted. However, the test-retest reliability of these tests had not been performed. In addition, the validity of the two cardiovascular test items with the gold standard of VO_2 peak testing have not been determined. In order to address this void, Chapter 3 presents findings of the study that explored the test-retest reliability of twelve functional fitness tests in adults with DS. Forty-three participants (24 men and 19 women) aged 18–50 years completed a battery of tests twice in a two-week period. All items were considered valid and reliable tests in a general elderly or intellectually disabled population. The test-retest relative reliability for all repeated tests was assessed with intraclass correlation coefficient performing one-way analysis of variance. The test-retest absolute variability was measured by using the standard error of measurement (SEM) to calculate the minimal detectable change at the 90% confidence interval (MDC_{90}). Reliability data was visualised with a Bland-Altman plot. Chapter 4 investigated the criterion-related validity of the 16-metre PACER and 6MWD to the gold standard of aerobic exercise testing (VO_2 peak). Forty-three adults with DS aged 18 to 50 years performed the three aerobic tests on non-consecutive days during a one-week period. To assess validity, peak oxygen uptake was measured directly on a motorised treadmill. Pearson-product moment correlations were performed. A linear regression analysis was conducted to determine criterion related validity between the field tests and the VO_2 peak test.

As empirically shown in Chapter 2, the positive effects of IT cannot be neglected, since IT has been shown to hold superior anthropometric, metabolic, functional and physical effects compared to traditional CAT. Many experimental studies using CAT programs demonstrated a limited means to significantly impact variables such as anthropometry and aerobic capacity in both ID and DS populations. The lack of information resulted in the development of an interval training program in persons with DS and ID. Chapter 5 presents the findings of the influence of interval training on body composition, physical and metabolic fitness in young adults with intellectual disability. Forty-six persons with ID (30 men and 16 women) were matched on age, gender and IQ between IT (n=17), CAT (n=15) and control (n=14). Participants were recruited from two special education centres (Revalijn and De Varens, Brugge, Belgium). The training groups exercised for 15 weeks, twice weekly (40 minutes). IT was performed in two blocks of 10 minutes (Block 1 and 3). Participants performed 10 sprints of 15 seconds (45 seconds of relative rest) on a cycle ergometer. During block 2, continuous training was performed. The CAT group performed three blocks of 10 minutes of continuous training (cycling, walking, stepping). After 8 weeks, the intensity of training was increased. Before and after the training period, body composition and physical and metabolic fitness were evaluated. Chapter 6 presents the findings of the effect of CAT versus IT on selected anthropometrical, health, physical and functional parameters of adults with DS in South Africa. Participants were recruited from three ID care centres (Huis Amelia, Potchefstroom; Die Oord, Brits; Uitkomsversorg, Pretoria). Forty-two adults with DS (25 men and 17 women) and a mean age of 33.8 ± 8.6 years were randomly allocated to one of three groups (IT, CAT, control). Training was performed for 12 weeks (3 times a week). The IT

group performed ten-30 second all out sprints with 90 seconds (1:3 work-rest ratio) of low cadence, low intensity cycling or walking. The CAT group performed continuous cycling and walking at an intensity of 70 - 80% of VO_2 peak. Heart rate monitors were used for monitoring training intensities. After 6 weeks of training, the intensity of the CAT was increased to 85% of VO_2 peak, whilst the intensity of the interval training group remained "all out". An increase of 5 minutes in duration was implemented after 6-weeks for both training groups. To evaluate pre–post differences between groups, a repeated analysis of covariance (ANCOVA) with post-hoc Bonferroni test was performed.

The conclusion of these studies will be presented in the next section.

7.3 Conclusion

The conclusion of the current study is derived from the stated hypotheses.

Hypothesis 1

A good to moderate test-retest reliability will be obtained in the majority of the 12 fitness tests for persons with DS.

All 12 tests (balance, flexibility, muscular strength and endurance, functional ability and cardiorespiratory fitness) showed excellent intraclass correlational coefficients (ICC>0.9). All SEM values demonstrated acceptable measurement precision (SEM<SD/2). The Bland-Altman analysis indicated that there was no systematic bias and the scatter was distributed randomly. Factors that may explain the high ICCs and low SEMs found in all tests might be as a consequence of the rigorous selection procedure of test (intensive literature review, use of test items with already desirable reliability and validity in other populations, input from various experts involved with DS individuals, three-month trial and error pilot study), practice and familiarisation sessions to avoid the learning effect, and clear, concise and simple communication with study participants.

In conclusion, all 12 tests were feasible and safe in an adult DS population. They demonstrated adequate absolute and relative test-retest reliability (ICC's 0.93-0.99). Therefore the first hypothesis is ***accepted.***

Hypothesis 2

Both field tests will present adequate validity but the 16-meter PACER test will provide a more valid indication of cardiorespiratory fitness.

Linear regression revealed that the PACER (R^2=0.86) and the 6MWD (R^2=0.75) were significantly related to directly measured VO_2 peak ($p<0.05$). Both the 16-metre PACER and the 6MWD significantly correlated

with VO_2 peak for adults with DS. The relationship was stronger for the 16-metre shuttle run test (r=0.87) than the 6MWD (r=0.78). The correlation remained significant and greater than 0.7 when controlling for age, gender and BMI. To conclude, both field tests are safe and feasible for use in adults with DS and are valid indicators of aerobic capacity. Therefore, the second hypothesis is ***accepted.***

Hypothesis 3

That interval training intervention will indicate a significantly larger improvement on the physical, metabolic and anthropometric profiles compared to the continuous training intervention of persons with ID.

IT showed a significant positive change for waist circumference (-4.3 cm), body fat (-3.8%), systolic blood pressure (-11 mmHg), lipid profile of total cholesterol (-15 mg/dL), HDL-chol (+4.5 mg/dL) and LDL-chol (-9.4 mg/dL), homeostasis model assessment of insulin resistance (-0.6), peak VO_2 (+0.2 L/min), peak Watt (+23.8 W), ventilatory threshold (+21 W, +0.2 L/min), 6MWD (+67.7 m) and muscle fatigue resistance (+6.3 s) when compared with no training ($p<0.05$). Moreover the IT group demonstrated significant improvements for body fat %, systolic blood pressure, low-density lipoprotein, fasting insulin, peak VO_2, and peak power and ventilatory threshold when compared to CAT ($p<0.05$).

In this study, we found more beneficial effects of IT on body composition, physical fitness and metabolic fitness compared to control. IT also resulted in an improved outcome compared with CAT. Based on the results the third hypothesis is ***partially accepted***.

Hypothesis 4

That interval training intervention will indicate a significantly larger improvement on the physical, metabolic, functional and anthropometric profiles compared to the continuous training intervention of persons with DS.

After 12 weeks of training, body mass (-2 kg) and BMI (-0.8 kg/m^2) decreased significantly more with IT compared to CAT ($p<0.05$). No significant changes were observed for other anthropometrical and health variables between IT and CAT. VO_2 peak and time to exhaustion ameliorated significantly in both the IT and CAT compared to control ($p<0.05$). However, relative VO_2 peak improved significantly more than with CAT ($p<0.05$). Participants in the IT group increased their VO_2 peak from 31.9 ± 8 ml/min/kg to 37.3 ± 8 ml/min/kg. Significant ameliorations for functional parameters and leg strength were shown for CAT compared to control ($p<0.05$). Participants in the CAT group improved their performance in the 6MWD (499 ± 78 m to 563 ± 75 m), 8-foot up and go (5.9 ± 1 s to 4.8 ± 1 s) and leg strength (13.1 ± 2 to 15.2 ± 2 number of sit-to-stands).

Both IT and CAT are feasible and safe exercising methods that can be pursued by adults with DS to positively impact on anthropometry, health, fitness and functional ability. IT has a superior impact on body mass loss and aerobic capacity when compared to CAT. On the other hand, CAT has a larger impact on functional parameters and leg strength compared to IT. Based on the results the fourth hypothesis is ***partially accepted.***

The main findings from this study is that a standardised functional fitness battery for adults with DS was established. Previous research established the feasibility and discriminant reliability of these 12 functional fitness tests. Test-retest reliability and validity for cardiovascular fitness field tests further standardised these tests. For the first time, adults with DS have their own unique standardised set of tests to measure and monitor functional fitness (balance, flexibility, muscular strength and endurance, cardiorespiratory endurance and functional ability). Accurate evaluations of functional fitness can now be made. Norm-referenced tables can be used to elicit optimal or suboptimal levels of balance, flexibility, muscular strength and endurance, functional ability and cardiorespiratory fitness. Strengths and weaknesses on an individual to individual bases can now be pinpointed with subsequent training programs tailored to their unique needs. Many health professionals working with persons with DS do not have access to sophisticated laboratory equipment such as a VO_2 peak metabolic analyser, but they can now use standardised field tests such as the 16-metre PACER and 6MWD field tests. Monitoring the aerobic capacity of this population is crucially important since many adults with DS are obese and have extremely low aerobic capacities. The functional fitness battery will be very helpful to the unique needs of this often neglected population of DS individuals. As most adults with DS live sedentary lives, the newly developed battery may be helpful in a motivational manner to curb inactivity related behaviour. Holistically, this battery of test items can be used correctly and effectively to develop and monitor all aspects of functional fitness.

However, adults with ID also live sedentary lives and have low aerobic capacities with a large majority being overweight or obese. Furthermore, obese individuals with ID and DS are more prone to develop secondary conditions such as diabetes, hypertension, cholesterol and depression compared to healthy ID individuals. Even though, individuals with ID (without DS) have their own functional fitness battery, most studies reported limited improvement in anthropometrical and aerobic capacities with structured exercise training (as adults with DS). Exercise interventions of longer duration or high intensity was needed. To address the many functional limitations these individuals face, experimental studies with both an IT and CAT protocol was followed. IT has been used with great success in similar functionally limited populations groups without ID. Both IT and CAT demonstrated many improvements on overall functional fitness in adults with DS and ID. Especially variables such as body composition and aerobic capacity were significantly improved. The improvement was more significant with IT (in both adults with ID and DS) as demonstrated in patients with chronic conditions in the general population. These results are important since a low aerobic capacity and an unhealthy body composition are associated with many health related conditions and poorer quality of life shown specifically in adults with ID and DS. This may also have a

positive effect on independence, functional ability and inhibition of premature ageing frequently encountered in this population. The fact that CAT demonstrated superior effects on functional ability in adults with DS and similar improvements to IT in adults with ID reinforces the belief that CAT should not be excluded from training programs. Both of these training regimens should be included in exercise programs as each hold physiological advantages in different aspects of functional fitness. IT and CAT are feasible, safe and useful for adults with ID and DS. The specific outcomes of the IT compared to continuous training can now shed light on the volume of training (intensity, duration and frequency) that would be ideal for optimal health and functional capacity development in an ID and DS population. Specifically in adults with DS this information can now be adapted and applied according to their functional fitness battery. For example, if an adult with ID or DS has a very low aerobic capacity we know he may benefit more from IT due to the superior influence of high intensity training on aerobic enzymes and central adaptations such as stroke volume. That does not exclude the use of CAT as its significant benefits have also been reported in this thesis. However, as shown in adults with DS, the CAT intensity should be higher (70 to 85% of VO_2 peak) than intensities used in previous studies. If low functional ability is identified, CAT and IT should be incorporated. The uninterrupted and continuous nature of CAT also hold many physiological benefits for functional ability. In addition to aerobic capacity, and functional ability other facets of functional fitness such as balance, flexibility, and muscular strength can be monitored.

Based on the findings from this study the final conclusion that can be drawn from this study, in response to the research question posed in Chapter 1, is that all 12 functional fitness tests demonstrated excellent test-retest reliability. Both cardiorespiratory field tests, indicated sound validity with the 16-metre PACER test being more valid. Lastly, both IT and CAT provided significant improvements to physical, functional and anthropometric profiles in persons with ID and DS. The influence of IT was more significant on anthropometry and aerobic capacity compared to CAT in adults with ID and DS. However, the impact of CAT was superior on functional ability and lower limb strength in adults with DS.

The original and innovative contribution of this study to the field of ID research is that a standardised instrument of functional fitness was developed specifically for adults with DS. Such instruments exist in the general, general elderly and intellectual and physical disabled populations but not in adults with DS. This thesis, builds on previous work (Boer, 2010) and provide norm-referenced tables that can be used to elicit optimal or suboptimal levels of balance, flexibility, muscular strength and endurance, functional ability and cardiorespiratory fitness. Cardiovascular field tests were validated with the gold standard aerobic test in this population. Reliable and valid aerobic field tests for adults with DS previously did not exist despite their many cardiovascular, physical and functional limitations and sedentary lifestyle. Importantly, this study demonstrated that higher exercise intensities, such as IT and CAT (70 to 85% of VO_2 peak), significant improvements in variables such as anthropometry and aerobic capacity were realised. Previously, it was believed that these variables cannot be significantly improved due to various physiological constraints such as chronotropic incompetence, muscle hypotonicity, cardiovascular

problems and many others. This study is therefore the first to successfully investigate the effect of an interval training program in persons with ID.

The findings of this study can be used to inform policy makers, coaches, physical education teachers, physiotherapists and biokineticists on the contribution of regular exercise in special populations in particular persons with DS and ID. However, these benefits must be studied in association with the limitations discussed in the next paragraph.

7.4 Limitations and recommendations

The findings from this study should be interpreted against the limitations that were present during the study. Persons with intellectual disability, in particular DS, are classified in different types of DS. In the current study all participants had trisomy-21. The results of the current study therefore cannot be generalised to those with the translocation or mosaic type. We know that those with mosaic type DS are of higher intellectual function and have better functional ability. However, mosaic and translocation type DS occurs in only 5% of all DS cases.

We were not able to measure intelligence quotient in the current study (Chapters 3, 4 and 6). None of the three ID centres had information pertaining to the intelligence quotient or the categorisation of borderline, mild, severe or profound ID. Inclusion criteria of the current study stipulated that only those who were able to cognitively understand test techniques and training procedures were included. Also, all participants were able to communicate and understand instructions provided. A recommendation is that future studies should include categorisation and IQ levels of participants.

Most adults with DS in the current study lived in residential care centres. Very few adults with DS live in private homes in South Africa. In most cases, individuals living in private homes are scattered across the country, mostly in the rural regions. To locate, and test these individuals, especially in experimental studies (where they are assessed thrice every week for 16 weeks) is very cumbersome. Also, most individuals living in the care facilities went home every weekend during the course of the study indicating the difficulty of controlling for group versus private living versus partial private living. As a result, it is possible that the findings of this study may not be generalised to individuals living in private homes. However, the effect of group versus private living on structured exercise training or the test-retest reliability and validity of selected test items should not confound the results of the current studies to a large degree. There were too few individuals living in private homes to further subcategorise the data and explore its associated effect. A recommendation is that future studies should locate possible geographical sites where many individuals with ID live in private homes and include them in a study with individuals from a care centre to determine to possible confounding influence of living arrangement in an exercise study.

7.5 Future research

Even though the test-retest and discriminant reliability for the 12 functional fitness tests in adults with DS has been established, the effects of inter-rater reliability for these tests remain to be determined.

The current study did not evaluate individuals with DS over the age of 50 years. It is known that the life expectancy of these individuals have increased dramatically in the last two decades. A future study should explore the reliability and validity of these tests and the effect of IT on this age group. Moreover, future longitudinal studies can trace the effects of very low functional fitness in specific individuals and study possible relationships with future morbidity and mortality. This will allow for the further refinement of the most important facets of functional fitness. Individuals with low functional fitness can then be flagged and the dangers of future morbidity and mortality can be discussed with the individual and parents.

Future studies should focus their attention on the combined effect of an exercise and diet regimen on fitness, body mass and body composition. The effect of a more complete lifestyle change can then be determined. It is clear from the literature that a large percentage of adults with DS are overweight or obese. Also the effect of a combined interval and resistance training regime may provide more or additional benefits as shown in studies with combined CAT and resistance training programs in a DS population. Another experimental study that can be pursued in the future is to compare the effect of two different types of interval training (aerobic interval training versus sprint interval training).

For practical purposes the sustainability of an intervention program should be studied. For example, an assessment of the quantity of individuals that stayed active after the research study terminated. Moreover the probability of future events such as falls, physical disability, sarcopenia, frailty, morbidity and mortality could be studied in conjunction with this study.

APPENDIX A: GUIDELINES TO AUTHORS

RESEARCH IN DEVELOPMENTAL DISABILITIES

RESEARCH IN DEVELOPMENTAL DISABILITIES

AUTHOR INFORMATION PACK

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Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

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As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

References in a special issue

Please ensure that the words 'this issue' are added to any references in the list (and any citations in the text) to other articles in the same Special Issue.

Reference management software

This journal has standard templates available in key reference management packages EndNote (http://www.endnote.com/support/enstyles.asp) and Reference Manager (http://refman.com/support/rmstyles.asp). Using plug-ins to wordprocessing packages, authors only need to select the appropriate journal template when preparing their article and the list of references and citations to these will be formatted according to the journal style which is described below.

Reference style

Text: Citations in the text should follow the referencing style used by the American Psychological Association. You are referred to the Publication Manual of the American Psychological Association, Sixth Edition, ISBN 978-1-4338-0561-5, copies of which may be ordered from http://books.apa.org/books.cfm?id=4200067 or APA Order Dept., P.O.B. 2710, Hyattsville, MD 20784, USA or APA, 3 Henrietta Street, London, WC3E 8LU, UK.

List: references should be arranged first alphabetically and then further sorted chronologically if necessary. More than one reference from the same author(s) in the same year must be identified by the letters 'a', 'b', 'c', etc., placed after the year of publication.

Examples:

Reference to a journal publication:

Van der Geer, J., Hanraads, J. A. J., & Lupton, R. A. (2010). The art of writing a scientific article. *Journal of Scientific Communications, 163*, 51–59.

Reference to a book:

Strunk, W., Jr., & White, E. B. (2000). *The elements of style.* (4th ed.). New York: Longman, (Chapter 4).

Reference to a chapter in an edited book:

Mettam, G. R., & Adams, L. B. (2009). How to prepare an electronic version of your article. In B. S. Jones, & R. Z. Smith (Eds.), *Introduction to the electronic age* (pp. 281–304). New York: E-Publishing Inc.

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Submission checklist

The following list will be useful during the final checking of an article prior to sending it to the journal for review. Please consult this Guide for Authors for further details of any item.

Ensure that the following items are present:

One author has been designated as the corresponding author with contact details:

- E-mail address
- Full postal address
- Phone numbers

All necessary files have been uploaded, and contain:

- Keywords
- All figure captions
- All tables (including title, description, footnotes)

Further considerations

- Manuscript has been 'spell-checked' and 'grammar-checked'
- References are in the correct format for this journal
- All references mentioned in the Reference list are cited in the text, and vice versa
- Permission has been obtained for use of copyrighted material from other sources (including the Internet)

Printed version of figures (if applicable) in color or black-and-white

- Indicate clearly whether or not color or black-and-white in print is required.
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Additional information

The word *retarded* should be used as an adjective rather than a noun; *retardate* should be avoided. Terms that are scientifically precise should be adhered to. Therefore, *mentally retarded* will be preferred to retarded because it specifies the type of retardation, and *intellectually average* or *normal intelligence* will be preferred over *normal*. A similar format should be followed if other disabilities are involved. It is understood that all investigations have been approved by the human subjects review committee of the author's institution.

Use of the Digital Object Identifier

The Digital Object Identifier (DOI) may be used to cite and link to electronic documents. The DOI consists of a unique alpha-numeric character string which is assigned to a document by the publisher upon the initial electronic publication. The assigned DOI never changes. Therefore, it is an ideal medium for citing a document, particularly 'Articles in press' because they have not yet received their full bibliographic information. Example of a correctly given DOI (in URL format; here an article in the journal *Physics Letters B*):
http://dx.doi.org/10.1016/j.physletb.
2010.09.059
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Corresponding authors will receive an e-mail with a link to our online proofing system, allowing annotation and correction of proofs online. The environment is similar to MS Word: in addition to editing text, you can also comment on figures/tables and answer questions from the Copy Editor. Web-based proofing provides a faster and less error-prone process by allowing you to directly type your corrections, eliminating the potential introduction of errors.
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We will do everything possible to get your article published quickly and accurately. Please use this proof only for checking the typesetting, editing, completeness and correctness of the text, tables and figures. Significant changes to the article as accepted for publication will only be considered at this stage with permission from the Editor.It is important to ensure that all corrections are sent back to us in one communication. Please check carefully before replying, as inclusion of any subsequent corrections cannot be guaranteed. Proofreading is solely your responsibility.

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APPENDIX B: GUIDELINES TO AUTHORS

CLINICAL REHABILITATION

Manuscript Submission Guidelines

Clinical Rehabilitation

Clinical Rehabilitation is a highly ranked, peer reviewed scholarly journal. It is a multi-professional journal covering the whole field of disability and rehabilitation, publishing research and discussion articles which are scientifically sound, clinically relevant and sometimes provocative.

The journal acts as a forum for the international dissemination and exchange of information amongst the large number of professionals involved in rehabilitation.

The leading journal in its field, *Clinical Rehabilitation* combines clinical application of scientific results and theoretical aspects in an ideal form. It gives high priority to articles describing effectiveness of therapeutic interventions and the evaluation of new techniques and methods.

1. Peer review policy

The journal's policy is to obtain at least two independent reviews of each article. It operates a double-blind reviewing policy in which the reviewer's name is always concealed from the submitting author; authors may choose to reveal their name but the journal otherwise leaves the article anonymous. Referees will be encouraged to provide substantive, constructive reviews that provide suggestions for improving the work and distinguish between mandatory and non-mandatory recommendations.

All manuscripts accepted for publication are subject to editing for presentation, style and grammar. Any major redrafting is agreed with the author but the Editor's decision on the text is final.

2. Article types

The journal publishes original papers, systematic reviews, Rehabilitation in Practice articles correspondence relating to published papers and short reports. Other article types should be discussed with the editor before submission.

2.1 Summary of manuscript structure:

- A title page with names and contact details for all authors
- A **structured** abstract of **no more than 250 words** (the website checks this)
- The text (usually Introduction, Methods, Results, Discussion)
- Clinical Messages (2-4 bullet points, 50 words or less)
- Acknowledgements, author contributions, competing interests and funding support
- References (Vancouver style)
- Tables, each starting on a new page
- Figures, each starting on a new page
- Appendix (if any)

Please note that short reports follow a different format:

- The main text of a short report will usually be between **1000 and 1500 words** in length.
- A short report should have sufficient key references to cover all important points, but no more and usually there will be a **maximum of 15 references**.

- Tables and figures can be very efficient and effective ways of presenting data. A short report will usually have **no more than three tables and figures** (in total) and most will be restricted to two.

Further information on short reports can be found here.

3. How to submit your manuscript

Before submitting your manuscript, please ensure you carefully read and adhere to all the guidelines and instructions to authors provided below. Manuscripts not conforming to these guidelines may be returned. If you would like to discuss your paper prior to submission, please contact the Editor (Derick Wade) at: clinical.rehabilitation@sagepub.co.uk

Clinical Rehabilitation has a fully web-based system for the submission and review of manuscripts. All submissions should be made online at the *Clinical Rehabilitation* SAGETRACK website:

http://mc.manuscriptcentral.com/clinrehab

Note: Online submission and review of manuscripts is now used for all types of papers.

New User Account
Please log onto the website. If you are a new user, you will first need to create an account. Follow the instructions and please ensure to enter a current and correct email address. Creating your account is a three-step process that takes a matter of minutes. When you have finished, your User ID and password is sent immediately via email. Please edit your user ID and password to something more memorable by selecting 'edit account' at the top of the screen. If you have already created an account but have forgotten your details type your email address in the 'Password Help' to receive an emailed reminder. Full instructions for uploading the manuscript are provided on the website.

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Review your submission (in both PDF and HTML formats) and then click the Submit button You may suspend a submission at any point before clicking the Submit button and save it to submit later. After submission, you will receive a confirmation e-mail. You can also log back

into your author centre at any time to check the status of your manuscript, but not to change it.

Please ensure that you submit editable/source files only (Microsoft Word or RTF) and that your document does not include page numbers; the SAGETRACK system will generate them for you, and then automatically convert your manuscript to PDF for peer review. All correspondence, including notification of the Editor's decision and requests for revisions, will be by email.

If you would like to discuss your paper prior to submission please contact the Editor: clinical.rehabilitation@sagepub.co.uk, and if you wish to seek advice on the submission process please contact the Publishing Editor: charlotte.jardine@sagepub.co.uk

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6. Other conventions

6.1 Informed Consent

Authors are required to ensure that the following guidelines are followed, as recommended by the International Committee of Medical Journal Editors ("Uniform Requirements for Manuscripts Submitted to Biomedical Journals": http://www.icmje.org/urm_full.pdf).

Patients have a right to privacy that should not be infringed without informed consent. Identifying information, including patients' names, initials, or hospital numbers, should not be published in written descriptions, photographs, and pedigrees unless the information is essential for scientific purposes and the patient (or parent or guardian) gives written informed consent for publication. Informed consent for this purpose requires that a patient who is identifiable be shown the manuscript to be published.
Complete anonymity is difficult to achieve, however, and informed consent should be obtained if there is any doubt. For example, masking the eye region in photographs of patients is inadequate protection of anonymity. If identifying characteristics are altered to protect anonymity, such as in genetic pedigrees, authors should provide assurance that alterations do not distort scientific meaning and editors should so note.

When informed consent has been obtained it should be indicated in the submitted article.

Authors should identify individuals who provide writing/administrative assistance, indicate the extent of assistanceand disclose the funding source for this assistance.
Identifying details should be omitted if they are not essential.

6.2 Ethics

When reporting experiments on human subjects, indicate whether the procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional or regional) or with the Declaration of Helsinki 1975, revised Hong Kong 1989. Do not use patients' names, initials or hospital numbers, especially in illustrative material. When reporting experiments on animals, indicate which guideline/law on the care and use of laboratory animals was followed.

7. Acknowledgements
Any acknowledgements should appear first at the end of your article prior to your Declaration of Conflicting Interests (if applicable), any notes and your References.

All contributors who do not meet the criteria for authorship should be listed in an `Acknowledgements' section. Examples of those who might be acknowledged include a person who provided purely technical help, writing assistance, or a department chair who

provided only general support. Authors should disclose whether they had any writing assistance and identify the entity that paid for this assistance.

7.1 Funding Acknowledgement

To comply with the guidance for Research Funders, Authors and Publishers issued by the Research Information Network (RIN), *Clinical Rehabilitation* additionally requires all Authors to acknowledge their funding in a consistent fashion under a separate heading. All research articles should have a funding acknowledgement in the form of a sentence as follows, with the funding agency written out in full, followed by the grant number in square brackets:

This work was supported by the Medical Research Council [grant number xxx].

Multiple grant numbers should be separated by comma and space. Where the research was supported by more than one agency, the different agencies should be separated by semi-colons, with "and" before the final funder. Thus:

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In some cases, research is not funded by a specific project grant, but rather from the block grant and other resources available to a university, college or other research institution. Where no specific funding has been provided for the research we ask that corresponding authors use the following sentence:

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Please include this information under a separate heading entitled "Funding" directly after any other Acknowledgements prior to your "Declaration of Conflicting Interests" (if applicable), any Notes and your References.

For more information on the guidance for Research Funders, Authors and Publishers, please visit: http://www.rin.ac.uk/funders-acknowledgement.

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9. Manuscript style

9.1 File types

Only electronic files conforming to the journal's guidelines will be accepted. Preferred formats for the text and tables of your manuscript are Word DOC, and tiff or jpeg for figures (ideally figures will use journal colours). RTF, XLS and LaTeX files are also accepted. Please also refer to additional guideline on submitting artwork [and supplemental files] below.

9.2 Journal Style
Clinical Rehabilitation conforms to the SAGE house style. Click here to review guidelines on SAGE UK House Style, which is summarised in 2.1.

9.3 Reference Style
Clinical Rehabilitation operates a SAGE Vancouver reference style. Click here to review the guidelines on SAGE Vancouver to ensure that your manuscript conforms to this reference style, which is summarised in 2.1.

9.4. Manuscript Preparation
The text should be double-spaced throughout and with a minimum of 3cm for left and right hand margins and 5cm at head and foot. Text should be standard 10 or 12 point. SI units should be used throughout the text.

9.4.1 Keywords and Abstracts
The title, keywords and abstract are key to ensuring that readers find your article online through online search engines such as Google. Please refer to the information and guidance on how best to title your article, write your abstract and select your keywords by visiting SAGE's Journal Author Gateway Guidelines on How to Help Readers Find Your Article Online.

9.4.2 Corresponding Author Contact details
Provide full contact details for the corresponding author including email, mailing address and telephone numbers. Academic affiliations are required for all co-authors.

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For guidance on the preparation of illustrations, pictures and graphs in electronic format, please visit SAGE's Manuscript Submission Guidelines.

Images should be supplied as bitmap based files (i.e. with .tiff or .jpeg extension) with a resolution of at least **300 dpi** (dots per inch). Line art should be supplied as vector-based, separate .eps files (not as .tiff files, and not only inserted in the Word or pdf file), with a resolution of **600 dpi**. Images should be clear, in focus, free of pixilation and not too light or dark.

If, together with your accepted article, you submit usable colour figures, these figures will appear in colour online regardless of whether or not these illustrations are reproduced in colour in the printed version. If a charge applies you will be informed by your SAGE Production Editor. For specifically requested colour reproduction in print, you will receive information regarding the costs from SAGE after receipt of your accepted article.

All submissions should be written in a clear and succinct manner, following the style of the journal. The title page should include a descriptive title, authors' surnames and forenames, address of each author and full address, telephone, fax and email contacts for the corresponding author. In text: tables and figures are either inserted as part of a sentence, for example table 1 or in parentheses for example (figure 1). Each table should carry a descriptive heading. Each figure should be submitted either electronically or as finalised hard copy with descriptive legends on a separate sheet. In text: references (where relevant) by superscript number after punctuation.

9.4.4 Guidelines for submitting supplemental files

The journal may be able to host approved supplemental materials online, alongside the full-text of articles. Supplemental files will be subjected to peer-review alongside the article. Please contact the Editor (clinical.rehabilitation@sagepub.co.uk) in the first instance. For more information please refer to SAGE's Guidelines for Authors on Supplemental Files.

9.4.5 English Language Editing

Non-English speaking authors who would like to refine their use of language in their manuscripts might consider using a professional editing service. Visit http://www.sagepub.co.uk/authors/journal/submission.sp for further information.

10. After acceptance

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We will email a PDF of the proofs to the corresponding author. Corrections should be limited to typographical amendments. Authors' approval will be assumed if corrections are not returned by the date indicated. **Note**: the file "PDF Proof" received with the acceptance email is **not** a proof, despite its name.

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11. Further information

11.1 Important 'Instructions to Authors' – from the Editor

Further specific advice on editorial aspects of the journal and of writing for the journal are also available.

Click here for further information and advice on submitting to *Clinical Rehabilitation*.

11.2 Contact SAGE

Any correspondence, queries or additional requests for information on the Manuscript Submission process should be sent to the Editorial Office as follows:

Charlotte Jardine
Publishing Editor
SAGE Publications
1 Oliver's Yard
55 City Road
London
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+44 (0)20 7336 1244

APPENDIX C: GUIDELINES TO AUTHORS

JOURNAL OF INTELLECTUAL DISABILITY RESEARCH

JIDR Journal of Intellectual Disability Research
Published on behalf of mencap and in association with IASSID

Journal of Intellectual Disability Research

Edited By: Chris Oliver Mental Health Special Issue Editor: Sally-Ann Cooper

Impact Factor: 2.411

ISI Journal Citation Reports © Ranking: 2013: 2/37 (Education Special); 3/69 (Rehabilitation (Social Science))

Online ISSN: 1365-2788

Author Guidelines

The journal to which you are submitting your manuscript employs a plagiarism detection system. By submitting your manuscript to this journal you accept that your manuscript may be screened for plagiarism against previously published works.

Individual authors and researchers can now check their work for plagiarism before submission - please click here for details.

3.1. Getting Started

Content of Author Guidelines: 1. General, 2. Ethical Guidelines, 3. Submission of Manuscripts, 4. Manuscript Types Accepted, 5. Manuscript Format and Structure, 6. After Acceptance.

Relevant Documents: Colour Work Agreement Form

Useful Websites: Submission Site, Articles published in The Journal of Intellectual Disability Research, Author Services, Blackwell Publishing's Ethical Guidelines, Guidelines for Figures.

1. GENERAL

The Journal of Intellectual Disability Research is devoted exclusively to the scientific study of intellectual disability and publishes papers reporting original observations in this field. The subject matter is broad and includes, but is not restricted to, findings from biological, educational, genetic, medical, psychiatric, psychological and sociological studies, and ethical, philosophical, and legal contributions that increase knowledge on the treatment and prevention of intellectual disability and of associated impairments and disabilities, and/or inform public policy and practice. Such reviews will normally be by invitation. The Journal also publishes Full Reports, Brief Reports, Letters to Editor, and an 'Hypothesis' papers. Submissions for Book Reviews and Announcements are also welcomed.

The Journal of Intellectual Disability Research will feature four Annotation articles each year covering a variety of topics of relevance to the main aims of the journal or topics. Senior researchers, academics and clinicians of recognised standing in their field will be invited to write an Annotation for the journal covering an area that will be negotiated with the Associate Editor, Prof. Chris Oliver, on behalf of the Editorial team. Anyone expert in his/her particular field wishing to submit an uninvited review is advised to seek prior guidance from the Associate Editor.

All papers are assessed by expert referees.

Please read the instructions below carefully for details on the submission of manuscripts, the journal's requirements and standards as well as information concerning the procedure after a manuscript has been accepted for publication in *The Journal of Intellectual Disability Research*. Authors are encouraged to visit John Wiley & Sons Pte Ltd's Author Services for further information on the preparation and submission of articles and figures.

2. ETHICAL GUIDELINES

The Journal of Intellectual Disability Research adheres to the ethical guidelines for publication and research summarised below.

2.1. Authorship and Acknowledgements

Authorship: Authors submitting a paper do so on the understanding that the manuscript has been read and approved by all authors and that all authors agree to the submission of the manuscript to the Journal. ALL named authors must have made an active contribution to the conception and design and/or analysis and interpretation of the data and/or the drafting of the paper and ALL must have critically reviewed its content and have approved the final version submitted for publication. Participation solely in the acquisition of funding or the collection of data does not justify authorship and, except in the case of complex large-scale or multi-centre research, the number of authors should not exceed six.

The Journal of Intellectual Disability Research adheres to the definition of authorship set up by The International Committee of Medical Journal Editors (ICMJE). According to the ICMJE authorship criteria should be based on 1) substantial contributions to conception and

design of, or acquisition of data or analysis and interpretation of data, 2) drafting the article or revising it critically for important intellectual content and 3) final approval of the version to be published. Authors should meet conditions 1, 2 and 3.

It is a requirement that all authors have been accredited as appropriate upon submission of the manuscript. Contributors who do not qualify as authors should be mentioned under Acknowledgements.

Acknowledgements: Under Acknowledgements please specify contributors to the article other than the authors accredited. Please also include specifications of the source of funding for the study and any potential conflict of interests if appropriate. Suppliers of materials should be named and their location (town, state/county, country) included.

2.2. Ethical Approvals

Experimental Subjects: experimentation involving human subjects will only be published if such research has been conducted in full accordance with ethical principles, including the World Medical Association Declaration of Helsinki (version, 2002 www.wma.net/e/policy/b3.htm) and the additional requirements, if any, of the country where the research has been carried out. Manuscripts must be accompanied by a statement that the research was undertaken with the understanding and written consent of each participant and according to the above mentioned principles. A statement regarding the fact that the study has been independently reviewed and approved by an ethical board should also be included. Editors reserve the right to reject papers if there are doubts as to whether appropriate procedures have been used.

All studies using human participants or animal subjects should include an explicit statement in the Material and Methods section identifying the review and ethics committee approval for each study, if applicable. Editors reserve the right to reject papers if there is doubt as to whether appropriate procedures have been used.

Ethics of investigation: Papers not in agreement with the guidelines of the Helsinki Declaration as revised in 1975 will not be accepted for publication.

2.3 Clinical Trials

Clinical trials should be reported using the CONSORT guidelines available at www.consort-statement.org. A CONSORT checklist should also be included in the submission material (http://www.consort-statement.org/mod_product/uploads/CONSORT 2001 checklist.doc).

Manuscripts reporting results from a clinical trial must provide the registration number and name of the clinical trial. Clinical trials can be registered in any of the following free, public clinical trials registries: www.clinicaltrials.gov, clinicaltrials-dev.ifpma.org/, isrctn.org/. The clinical trial registration number and name of the trial register will be published with the paper.

The Journal of Intellectual Disability Research encourages authors submitting manuscripts reporting from a clinical trial to register the trials in any of the following free, public clinical trials registries: www.clinicaltrials.gov, clinicaltrials-dev.ifpma.org/, isrctn.org/. The clinical trial registration number and name of the trial register will then be published with the paper.

2.4 Conflict of Interest and Source of Funding

Conflict of Interest: Authors are required to disclose any possible conflict of interest. These include financial (for example patent, ownership, stock ownership, consultancies, speaker's fee). Author's conflict of interest (or information specifying the absence of conflicts of interest) will be published under a separate heading entitled 'Conflict of Interests'.

The Journal of Intellectual Disability Research requires that sources of institutional, private and corporate financial support for the work within the manuscript must be fully acknowledged, and any potential conflicts of interest noted. As of 1st March 2007, this information will be a requirement for all manuscripts submitted to the Journal and will be published in a highlighted box on the title page of the article. Please include this information under the separate headings of 'Source of Funding' and 'Conflict of Interest' at the end of your manuscript.

If the author does not include a conflict of interest statement in the manuscript then the following statement will be included by default: "No conflicts of interest have been declared".

Source of Funding: Authors are required to specify the source of funding for their research when submitting a paper. Suppliers of materials should be named and their location (town, state/county, country) included. The information will be disclosed in the published article.

2.5 Appeal of Decision

Authors who wish to appeal the decision on their submitted paper may do so by e-mailing the Editorial Office with a detailed explanation for why they find reasons to appeal the decision.

2.6 Permissions

If all or parts of previously published illustrations are used, permission must be obtained from the copyright holder concerned. It is the author's responsibility to obtain these in writing and provide copies to the Publishers.

2.7 Copyright Assignment

If your paper is accepted, the author identified as the formal corresponding author for the paper will receive an email prompting them to login into Author Services; where via the Wiley Author Licensing Service (WALS) they will be able to complete the license agreement on behalf of all authors on the paper.

For authors signing the copyright transfer agreement

If the OnlineOpen option is not selected the corresponding author will be presented with the copyright transfer agreement (CTA) to sign. The terms and conditions of the CTA can be previewed in the samples associated with the Copyright FAQs below:

CTA Terms and Conditions http://authorservices.wiley.com/bauthor/faqs_copyright.asp

2.8 OnlineOpen

If the OnlineOpen option is selected the corresponding author will have a choice of the following Creative Commons License Open Access Agreements (OAA):

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To preview the terms and conditions of these open access agreements please visit the Copyright FAQs hosted on Wiley Author Services http://authorservices.wiley.com/bauthor/faqs_copyright.asp and visit http://www.wileyopenaccess.com/details/content/12f25db4c87/Copyright--License.html.

If you select the OnlineOpen option and your research is funded by The Wellcome Trust and members of the Research Councils UK (RCUK) you will be given the opportunity to publish your article under a CC-BY license supporting you in complying with Wellcome Trust and Research Councils UK requirements. For more information on this policy and the Journal's compliant self-archiving policy please visit: http://www.wiley.com/go/funderstatement.

3. SUBMISSION OF MANUSCRIPTS

Manuscripts should be submitted electronically via the online submission site http://mc.manuscriptcentral.com/jidr. The use of an online submission and peer review site enables immediate distribution of manuscripts and consequentially speeds up the review process. It also allows authors to track the status of their own manuscripts. Complete instructions for submitting a paper are available online and below. Further assistance can be obtained from Ms Sue M Hampton-Matthews at the Editorial Office of JIDR, Second Floor, Douglas House, 18b Trumpington Road, Cambridge, CB2 2AH, UK +44 1223 746 124; e-mail: shm44@medschl.cam.ac.uk.

- Launch your web browser (supported browsers include Internet Explorer 6 or higher, Netscape 7.0, 7.1, or 7.2, Safari 1.2.4, or Firefox 1.0.4) and go to the journal's online Submission Site: http://mc.manuscriptcentral.com/jidr
- Log-in or click the 'Create Account' option if you are a first-time user.
- If you are creating a new account.
  - After clicking on 'Create Account', enter your name and e-mail information and click 'Next'. Your e-mail information is very important.
  - Enter your institution and address information as appropriate, and then click 'Next.'
  - Enter a user ID and password of your choice (we recommend using your e-mail address as your user ID), and then select your area of expertise. Click 'Finish'.
- If you have an account, but have forgotten your log in details, go to Password Help on the journals online submission system http://mcv3support.custhelp.com and enter your e-mail address. The system will send you an automatic user ID and a new temporary password.
- Log-in and select 'Author Center'.

3.2. Submitting Your Manuscript

- After you have logged in, click the 'Submit a Manuscript' link in the menu bar.
- Enter data and answer questions as appropriate. You may copy and paste directly from your manuscript and you may upload your pre-prepared covering letter.
- Click the 'Next' button on each screen to save your work and advance to the next screen.
- You are required to upload your files.
  - Click on the 'Browse' button and locate the file on your computer.
  - Select the designation of each file in the drop-down menu next to the Browse button.
  - When you have selected all files you wish to upload, click the 'Upload Files' button.
- Review your submission (in HTML and PDF format) before sending to the Journal. Click the 'Submit' button when you are finished reviewing.

3.3. Manuscript Files Accepted

Manuscripts should be uploaded as Word (.doc) or Rich Text Format (.rft) files (not write-protected) plus separate figure files. GIF, JPEG, PICT or Bitmap files are acceptable for submission, but only high-resolution TIF or EPS files are suitable for printing. The files will be automatically converted to HTML and PDF on upload and will be used for the review process. The text file must contain the entire manuscript including title page, abstract, text, references, tables, and figure legends, but no embedded figures. Figure tags should be included in the file. Manuscripts should be formatted as described in the Author Guidelines below.

Please note that any manuscripts uploaded as Word 2007 (.docx) will be automatically rejected. Please save any .docx file as .doc before uploading.

3.4. Blinded Review

All manuscripts submitted to The Journal of Intellectual Disability Research will be reviewed by two experts in the field. The Journal of Intellectual Disability Research uses double-blinded review. The names of the reviewers will thus not be disclosed to the author submitting a paper and the name(s) of the author(s) will not be disclosed to the reviewers.

To allow double-blinded review, please submit (upload) your main manuscript and title page as separate files.

Please upload:

- Your manuscript without title page under the file designation 'main document'
- Figure files under the file designation 'figures'
- The title page, Acknowledgements and Conflict of Interest Statement where applicable, should be uploaded under the file designation 'title page'.

All documents uploaded under the file designation 'title page' will not be viewable in the HTML and PDF format you are asked to review at the end of the submission process. The files viewable in the HTML and PDF format are the files available to the reviewer in the review process.

3.5. Suggest a Reviewer

The Journal of Intellectual Disability Research attempts to keep the review process as short as possible to enable rapid publication of new scientific data. In order to facilitate this process, please suggest the names and current e-mail addresses of 1 potential international reviewer whom you consider capable of reviewing your manuscript. In addition to your choice the journal editor will choose one or two reviewers as well.

3.6. Suspension of Submission Mid-way in the Submission Process

You may suspend a submission at any phase before clicking the 'Submit' button and save it to submit later. The manuscript can then be located under 'Unsubmitted Manuscripts' and you can click on 'Continue Submission' to continue your submission when you choose to.

3.7. E-mail Confirmation of Submission

After submission you will receive an e-mail to confirm receipt of your manuscript. If you do not receive the confirmation e-mail after 24 hours, please check your e-mail address carefully in the system. If the e-mail address is correct please contact your IT department. The error may be caused by spam filtering software on your e-mail server. Also, the e-mails should be received if the IT department adds our e-mail server (uranus.scholarone.com) to their whitelist.

3.8. Manuscript Status

You can access ScholarOne Manuscripts any time to check your 'Author Center' for the status of your manuscript. The Journal will inform you by e-mail once a decision has been made.

3.9. Submission of Revised Manuscripts

Revised manuscripts must be uploaded within 3 months of authors being notified of conditional acceptance pending satisfactory revision. Locate your manuscript under 'Manuscripts with Decisions' and click on 'Submit a Revision' to submit your revised manuscript. Please remember to delete any old files uploaded when you upload your revised manuscript. Please also remember to upload your manuscript document separate from your title page.

4. MANUSCRIPT TYPES ACCEPTED

Original Research Article The main text should proceed through sections of Abstract, Introduction, Methods, Results, and Discussion.

Full Reports of up to 4,500 words are suitable for major studies, integrative reviews and presentation of related research projects or longitudinal enquiry of major theoretical and/or empirical conditions.

Brief Reports of up to 1,500 words are encouraged especially for replication studies, methodological research and technical contributions.

Annotation Articles should be no more than 5,500 words long including tables and figures and should not have been previously published or currently under review with another journal. The normal instructions to authors apply. The date for submission of the article should be negotiated with the Associate Editor. An honorarium of £400 in total shall be paid to the authors(s) when the article is accepted for publication.

Three main types of Annotations will be commissioned: 1. Authoritative reviews of empirical and theoretical literature. 2. Articles proposing a novel or modified theory or model. 3. Articles detailing a critical evaluation and summary of literature pertaining to the treatment of a specific disorder.

A Hypothesis Paper can be up to 2,500 words and no more than twenty key references. It aims to outline a significant advance in thinking that is testable and which challenges previously held concepts and theoretical perspectives.

5. MANUSCRIPT FORMAT AND STRUCTURE

5.1. Format

Language: The language of publication is English. Authors for whom English is a second language must have their manuscript professionally edited by an English speaking person before submission to make sure the English is of high quality. It is preferred that manuscripts are professionally edited. A list of independent suppliers of editing services can be found at http://authorservices.wiley.com/bauthor/english_language.asp . All services are paid for and arranged by the author and use of one of these services does not guarantee acceptance or preference for publication.

Abbreviations, Symbols and Nomenclature: Spelling should conform to The Concise Oxford Dictionary of Current English and units of measurements, symbols and abbreviations with those in Units, Symbols and Abbreviations (1977) published and supplied by the Royal Society of Medicine, 1 Wimpole Street, London W1M 8AE. This specifies the use of SI units.

It is important that the term 'intellectual disabilities' is used when preparing manuscripts.

Please note that 'intellectual disability', as used in the Journal, includes those conditions labelled mental deficiency, mental handicap, learning disability and mental retardation in some counties.

5.2. Structure

All manuscripts submitted to *The Journal of Intellectual Disability Research* should include: Title, Keywords, structured Abstract, Main Text (divided by appropriate sub headings) and References.

Title Page: Please remember that **peer-review is double-blind**, so that neither authors nor reviewers know each others' identity. Therefore, **no identifying details of the authors or their institutions must appear in the submitted manuscript; author details should be entered as part of the online submission process**. However, a 'Title Page' must be

submitted as part of the submission process as a 'Supplementary File Not for Review'. This should contain the title of the paper, names and qualifications of all authors, their affiliations and full mailing address, including e-mail addresses and fax and telephone numbers.

Keywords: The author should also provide up to six keywords to aid indexing.

Abstracts: For full and brief reports a structured summary should be included at the beginning of each article, incorporating the following headings: Background, Method, Results, and Conclusions. These should outline the questions investigated, the design, essential findings, and the main conclusions of the study.

Optimizing Your Abstract for Search Engines: Many students and researchers looking for information online will use search engines such as Google, Yahoo or similar. By optimizing your article for search engines, you will increase the chance of someone finding it. This in turn will make it more likely to be viewed and/or cited in another work. We have compiled these guidelines to enable you to maximize the web-friendliness of the most public part of your article.

5.3. References

The Journal follows the Harvard reference style. References in text with more than two authors should be abbreviated to (Brown et al. 1977). Authors are responsible for the accuracy of their references.

The reference list should be in alphabetical order thus:

- Giblett E.R. (1969) Genetic Markers in Human Blood.
- Blackwell Scientific Publications, Oxford.
- Moss T.J. & Austin G.E. (1980) Preatherosclerotic lesions in Down's syndrome. *Journal of Mental Deficiency Research* **24**, 137- 41.
- Seltzer M. M. & Krauss M.W. (1994) Aging parents with co-resident adult children: the impact of lifelong caregiving. In: *Life Course Perspectives on Adulthood and Old Age* (eds M. M. Seltzer, M.W. Krauss & M. P. Janicki), pp. 3–18. American Association on Mental Retardation, Washington, DC.

Where more than six authors are listed for a reference please use the first six then 'et al.'

The Editor and Publisher recommend that citation of online published papers and other material should be done via a DOI (digital object identifier), which all reputable online published material should have - see www.doi.org/ for more information. If an author cites anything which does not have a DOI they run the risk of the cited material not being traceable.

We recommend the use of a tool such as EndNote or Reference Manager for reference management and formatting.

EndNote reference styles can be searched for here: www.endnote.com/support/enstyles.asp

Reference Manager reference styles can be searched for here:
www.refman.com/support/rmstyles.asp

5.4. Tables, Figures

Tables: Tables should include only essential data. Each table must be typewritten on a separate sheet and should be numbered consecutively with Arabic numerals, e.g. Table 1, Table 2, etc., and give a short caption.

Figures: All graphs, drawings and photographs are considered figures and should be numbered in sequence with Arabic numerals. All symbols and abbreviations should be clearly explained.

Tables and figures should be referred to in the text together with an indication of their approximate position recorded in the text margin.

Preparation of Electronic Figure for Publication

Although low quality images are adequate for review purposes, print publication requires high quality images to prevent the final product being blurred or fuzzy. Submit EPS (line art) or TIFF (halftone/photographs) files only. MS PowerPoint and Word Graphics are unsuitable for printed pictures. Do not use pixel-oriented programmes. Scans (TIFF only) should have a resolution of at least 300 dpi (halftone) or 600 to 1200 dpi (line drawings) in relation to the reproduction size (see below). Please submit the data for figures in black and white or submit a Colour Work Agreement Form (see Colour Charges below). EPS files should be saved with fonts embedded (and with a TIFF preview if possible).

For scanned images, the scanning resolution (at final image size) should be as follows to ensure good reproduction: line art: >600 dpi; halftones (including gel photographs): >300 dpi; figures containing both halftone and line mages: >600 dpi.

Further information can be obtained at guidelines for figures:
http://authorservices.wiley.com/bauthor/illustration.asp

Check your electronic artwork before submitting it:
http://authorservices.wiley.com/bauthor/eachecklist.asp

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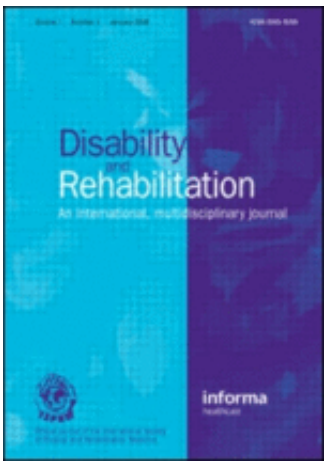
Colour Charges: It is the policy of *The Journal of Intellectual Disability Research* for authors to pay the full cost for the reproduction of their colour artwork. Therefore, please note that if there is colour artwork in your manuscript when it is accepted for publication,

John Wiley & Sons Pte Ltd require you to complete and return a Colour Work Agreement Form before your paper can be published. Any article received by John Wiley & Sons with colour work will not be published until the form has been returned.

APPENDIX D: GUIDELINES TO AUTHORS

DISABILITY AND REHABILITATION



Disability and Rehabilitation

Instructions for Authors

Disability and Rehabilitation is an international interdisciplinary journal and particularly welcomes contributions from a wide range of professional groups, including medical practitioners, occupational therapists, physiotherapists, speech and language therapists, clinical psychologists and those involved in nursing, education and engineering.

Disability and Rehabilitation is organised into sections: Reviews; Research Papers; Case Studies; Perspectives on Rehabilitation; reports on Rehabilitation in Practice, Education and Training and Correspondence.

Special Issues and specific sections on contemporary themes of interest to the Journal's readership are published. Please contact the Editor for more information.

Submissions and Peer-Review

All submissions should be made online at *Disability and Rehabilitation*'s ScholarOne Manuscripts site: http://mc.manuscriptcentral.com/dandr

Authors are given the option to remain anonymous during the peer-review process. Authors will be able to indicate whether their paper is 'Anonymous' or 'Not Anonymous' during manuscript submission, and should pay particular attention to the below:

Authors who wish to remain anonymous should prepare a complete text with information identifying the author(s) removed. This should be uploaded as the "Main Document" and will be sent to the referees. A separate title page should be included providing the full affiliations of all authors. Any acknowledgements and the Declaration of Interest statement must be included but should be worded mindful that these sections will be made available to referees.

Authors who wish to be identified should include the name(s) and affiliation(s) of author(s) on the first page of the manuscript. The complete text should be uploaded as the "Main Document".

All submissions should include a separate title page that contains contact information for the authors(s).This should be uploaded as a "Title Page" and will not be sent to referees. If a paper is deemed to be acceptable for publication pending minor revision, the author(s) names may be disclosed to the referees when the Editor's decision is made, irrespective of whether the authors names(s) were included as part of the original submission. Every effort will be made to keep the author(s) name(s) anonymous, if required, should the paper require extensive revision and further peer-review. If authors wish to remain anonymous throughout the second round of peer-review, they are reminded not to include identifying information in
the „Authors" Response" section during the upload of their revised paper.

Every paper that is revised and resubmitted must clearly indicate the parts of the manuscript that contain amendments, by highlighting the revised text in a different colour or by using 'Track Changes'(for minor revisions).

Systematic Reviews should be submitted as a "Review" and Narrative Reviews should be submitted as "Perspectives in Rehabilitation". All Systematic Reviews will be automatically submitted for the annual Best Review Paper competition.

Education and Training

This is a new section for the journal. It will publish papers relating to the education and professional training of those working in the field of rehabilitation. Papers are encouraged which develop innovatory approaches to this process and provide multi-disciplinary and international comparisons for those working in the field. Through this new section it is intended to contribute towards the development of education and training within these professional groupings.

Papers should be submitted with any tables, figures, or photographs, all of which should be of high quality suitable for reproduction. Submissions should be in English presented in double line spacing.

Submissions should include, where appropriate, a formal statement that ethical consent for the work to be carried out has been given. Photographs of patients should be avoided, but if essential, patients' consent in writing must accompany manuscript. It is not sufficient to mask identity by covering the patients' eyes.

Word Limit
There is no stated word limit to papers submitted to *Disability and Rehabilitation*. It should however be noted that space is at a premium and therefore succinct and well-constructed papers are more likely to be reviewed positively. However, the key to evaluating a paper will be the quality of the work along with the methodology adopted particularly for qualitative studies which do tend to be longer.

Disability and Rehabilitation considers all manuscripts at the Editor's discretion; the Editor's decision is final. Please see below for information on the Journal's Appeal Procedure.

Disability and Rehabilitation considers all manuscripts on the strict condition that they are the property (copyright) of the submitting author(s), have been submitted only to *Disability and Rehabilitation*, that they have not been published already, nor are they under consideration for publication, nor in press elsewhere. Authors who fail to adhere to this condition will be charged all costs which *Disability and Rehabilitation* incurs, and their papers will not be published. Copyright will be transferred to *Disability and Rehabilitation* and Informa UK Ltd., if the paper is accepted.

IMPLICATIONS FOR REHABILITATION
A feature of the Journal is a boxed insert on „Implications for Rehabilitation". This box should include between two to four main bullet points drawing out the implications for rehabilitation for your paper. **All papers including reviews, research, rehabilitation in practice, perspectives on rehabilitation, case studies and a new section on education and training for rehabilitation professionals must include this feature**. This should be

uploaded as a separate document through Manuscript Central as a single side of A4 during submission.

Included below are examples. If you have any questions, please contact the Editor.

Example 1: Leprosy
• Leprosy is a disabling disease which not only impacts physically but restricts quality of life often through stigmatisation.
• Reconstructive surgery is a technique available to this group.
• In a relatively small sample this study shows participation and social functioning improved after surgery.

Example 2: Multiple Sclerosis
• Exercise is an effective means of improving health and well-being experienced by people with multiple sclerosis (MS).
• People with MS have complex reasons for choosing to exercise or not.
• Individual structured programmes are most likely to be successful in encouraging exercise in this cohort.

Example 3: Community Based Rehabilitation
• Community Based Rehabilitation (CBR) is a Western concept that may not readily fit other cultures.
• CBR needs to be „owned" by those involved and subject to re-interpretation to be effective in other cultures.

Standardised Reporting Guidelines

We encourage Authors to be aware of, and to take into account standardised reporting guidelines when preparing their manuscripts.

The table below provides information about guidelines for different study types

Study Type	Name	Source
Case reports	CARE	www.care-statement.org
Diagnostic accuracy	STARD	www.stard-statement.org
Observational studies	STROBE	http://strobe-statement.org
Randomized controlled trial	CONSORT	www.consort-statement.org
Systematic reviews, meta-analyses	PRISMA	www.prisma-statement.org

Whilst the use of such guidelines is supported, given the multi-disciplinary nature of the Journal, it is not compulsory.

Manuscript Preparation

In writing your paper, you are encouraged to review articles in the area you are addressing which have been previously published in the Journal and where you feel appropriate, to reference them. This will enhance context, coherence, and continuity for our readers.

File preparation and types

Manuscripts are preferred in Microsoft Word format (.doc files). Documents must be double-spaced, with margins of one inch on all sides. Tables and figures should not appear in the

main text, but should be uploaded as separate files and designated with the appropriate file type upon submission. These should be submitted as “Image” files during submission. References should be given in Council of Science Editors (CSE) Citation & Sequence format (see References section for examples).

Structure of Paper

Manuscripts should be compiled in the following order: title page; abstract; main text; acknowledgments; Declaration of Interest statement; appendices (as appropriate); references; tables with captions (uploaded as separate files); figures with captions (uploaded as separate files).

An introductory section should state the purpose of the paper and give a brief account of previous work. New techniques and modifications should be described concisely but in sufficient detail to permit their evaluation; standard methods should simply be referenced. Experimental results should be presented in the most appropriate form, with sufficient explanation to assist their interpretation; their discussion should form a distinct section. Extensive tabulations will not be accepted unless their inclusion is essential.

Title Page

A title page should be provided comprising the manuscript title plus the full names and affiliations of all authors involved in the preparation of the manuscript. One author should be clearly designated as the corresponding author and full contact information, including phone number and email address, provided for this person. Keywords that are not in the title should also be included on the title page. The keywords will assist indexers in cross indexing the article. The title page should be uploaded separately to the main manuscript and designated as “title page” on ScholarOne Manuscripts. This will not get sent to referees.

Abstracts

Structured abstracts are required for all papers, and should be submitted as detailed below, following the title page, preceding the main text.

Purpose State the main aims and objectives of the paper.
Method Describe the design and methodological procedures adopted.
Results Present the main results.
Conclusions State the conclusions that have been drawn and their relevance to the study of disability and rehabilitation.

The abstract should not exceed 200 words.

Nomenclature and Units

All abbreviations and units should conform to SI practice. Drugs should be referred to by generic names; trade names of substances, their sources, and details of manufacturers of scientific instruments should be given only if the information is important to the evaluation of the experimental data.

Copyright Permission

Contributors are required to secure permission for the reproduction of any figure, table, or extensive (more than fifty word) extract from the text, from a source which is copyrighted - or owned - by a party other than Informa UK Ltd or the contributor. This applies both to direct

reproduction or 'derivative reproduction' - when the contributor has created a new figure or table which derives substantially from a copyrighted source.

Code of Experimental Ethics and Practice

Contributors are required to follow the procedures in force in their countries which govern the ethics of work done with human or animal subjects. The Code of Ethics of the World Medical Association (Declaration of Helsinki) represents a minimal requirement.

Tables, figures and illustrations

The same data should not be reproduced in both tables and figures. The usual statistical conventions should be used: a value written 10.0 ± 0.25 indicates the estimate for a statistic (e.g. a mean) followed by its standard error. A mean with an estimate of the standard deviation will be written 10.0 SD 2.65.

Contributors reporting ages of subjects should specify carefully the age groupings: a group of children of ages e.g. 4.0 to 4.99 years may be designated 4 +; a group aged 3.50 to 4.49 years 4 ± and a group all precisely 4.0 years, 4.0.

Tables and figures should be referred to in text as follows: figure 1, table 1, i.e. lower case. 'As seen in table [or figure] 1 ...' (not Tab., fig. or Fig).

The place at which a table or figure is to be inserted in the printed text should be indicated clearly on a manuscript:

Insert table 2 about here

Each table and/or figure must have a title that explains its purpose without reference to the text. The filename for the tables and/or figures should be descriptive of the graphic, e.g. table 1, figure 2a.

Tables

Tables should be used only when they can present information more efficiently than running text.

Care should be taken to avoid any arrangement that unduly increases the depth of a table, and the column heads should be made as brief as possible, using abbreviations liberally. Lines of data should not be numbered nor run numbers given unless those numbers are needed for reference in the text.

Columns should not contain only one or two entries, nor should the same entry be repeated numerous times consecutively. Tables should be grouped at the end of the manuscript on uploaded separately to the main body of the text.

Figures and illustrations

Figures must be uploaded separately and not embedded in the text. Avoid the use of colour and tints for purely aesthetic reasons. Figures should be produced as near to the finished size as possible.

Files should be saved as one of the following formats: TIFF (tagged image file format), PostScript or EPS (encapsulated PostScript), and should contain all the necessary font

information and the source file of the application (e.g. CorelDraw/Mac, CorelDraw/PC). All files must be 300 dpi or higher.

Please note that it is in the author's interest to provide the highest quality figure format possible.

Acknowledgments and Declaration of Interest sections

Acknowledgments and Declaration of interest sections are different, and each has a specific purpose.

The Acknowledgments section details special thanks, personal assistance, and dedications. Contributions from individuals who do not qualify for authorship should also be acknowledged here.

Declarations of interest, however, refer to statements of financial support and/or statements of potential conflict of interest. Within this section also belongs disclosure of scientific writing assistance (use of an agency or agency/ freelance writer), grant support and numbers, and statements of employment, if applicable.

Acknowledgments section

Any acknowledgments authors wish to make should be included in a separate headed section at the end of the manuscript preceding any appendices, and before the references section. Please do not incorporate acknowledgments into notes or biographical notes.

Declaration of Interest section

All declarations of interest must be outlined under the subheading “Declaration of interest”. If authors have no declarations of interest to report, this must be explicitly stated. The suggested, but not mandatory, wording in such an instance is: *The authors report no declarations of interest.* When submitting a paper via ScholarOne Manuscripts, the “Declaration of interest” field is compulsory (authors must either state the disclosures or report that there are none). If this section is left empty authors will not be able to progress with the submission.

Please note: for NIH/Wellcome-funded papers, the grant number(s) must be included in the Declaration of Interest statement.

Mathematics
Click for more information on the presentation of mathematical text.

References

References should follow the Council of Science Editors (CSE) Citation & Sequence format. Only works actually cited in the text should be included in the references. Indicate in the text with Arabic numbers inside square brackets. Spelling in the reference list should follow the original. References should then be listed in numerical order at the end of the article. Further examples and information can be found in The CSE Manual for Authors, Editors, and Publishers, Seventh Edition. Periodical abbreviations should follow the style given by Index Medicus.

Examples are provided as follows:

Journal article: [1] Steiner U, Klein J, Eiser E, Budkowski A, Fetters LJ. Complete wetting from polymer mixtures. Science 1992;258:1122-9.

Book chapter: [2] Kuret JA, Murad F. Adenohypophyseal hormones and related substances. In: Gilman AG, Rall TW, Nies AS, Taylor P, editors. The pharmacological basis of therapeutics. 8th ed. New York: Pergamon; 1990. p 1334-60.

Conference proceedings: [3] Irvin AD, Cunningham MP, Young AS, editors. Advances in the control of Theileriosis. International Conference held at the International Laboratory for Research on Animal Diseases; 1981 Feb 9-13; Nairobi. Boston: Martinus Nijhoff Publishers; 1981. 427 p.

Dissertations or Thesis: [4] Mangie ED. A comparative study of the perceptions of illness in New Kingdom Egypt and Mesopotamia of the early first millennium [dissertation]. Akron (OH): University of Akron; 1991. 160 p. Available from: University Microfilms, Ann Arbor MI; AAG9203425.

Journal article on internet: [5] De Guise E, Leblanc J, Dagher J, Lamoureux J, Jishi A, Maleki M, Marcoux J, Feyz M. 2009. Early outcome in patients with traumatic brain injury, pre-injury alcohol abuse and intoxication at time of injury. Brain Injury 23(11):853-865. http://www.informaworld.com/10.1080/02699050903283221. Accessed 2009 Oct 06

Webpage: [6] British Medical Journal [Internet]. Stanford, CA: Stanford Univ; 2004 July 10 - [cited 2004 Aug 12]; Available from: http://bmj.bmjjournals.com

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APPEAL PROCEDURE

Disability and Rehabilitation* and *Disability and Rehabilitation: Assistive Technology
The Editors of both Journals will respond to appeals from Authors relating to papers which have been rejected.

The Author(s) should email the Editor outlining the concerns and making a case for why their paper should not have been rejected. The Editor will undertake one of two courses of action:

1: The Editor Accepts the Appeal

- In this case the Editor will secure a further review making available confidentially the relevant information for the reviewer
- The Editor on receiving the review will either accept the appeal and therefore invite a resubmission for further review; or reject the appeal and no further action will be taken.
- If an appeal is rejected there will be no further right of appeal within the jurisdiction of the Journal.

2: The Editor does not uphold the Appeal

- If the Editor does not accept the appeal and is not prepared to secure further review the decision will be referred to the Editor of the relevant affiliated Journal for independent consideration. In the case of *Disability and Rehabilitation*, the Editor of *Disability and Rehabilitation: Assistive Technology* will be contacted, and if an appeal is not upheld by the Editor of *Disability and Rehabilitation: Assistive Technology*, the Editor of *Disability and Rehabilitation* will be consulted.

- II. The Editor will either confirm the decision or recommend that a further review be obtained.
- III. Therefore, if both Editors agree that the appeal should not be upheld there will be no further right of appeal within the jurisdiction of the Journal.

Dave Muller, Editor in Chief, *Disability and Rehabilitation*
Marcia Scherer, Editor, *Disability and Rehabilitation: Assistive Technology*

APPENDIX E: CONSENT FORM

CHAPTER THREE

NORTH-WEST UNIVERSITY
YUNIBESITI YA BOKONE-BOPHIRIMA
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POTCHEFSTROOM CAMPUS

Physical Activity, Sport and Recreation

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pieterhenk.boer@nwu.ac.za

The functional fitness capacity of adults with Down syndrome (Study 1)

CONSENT FORM (simpler language): RESEARCH PARTICIPANT

I, PIETER-HENK BOER, am working on a research project involving a descriptive study conducted on Down syndrome adults. We would like to invite you to give consent to participate in our study.

1. PURPOSE OF THE STUDY

We would like you to participate in a research study where we want to find out how fit people with Down syndrome are. If you decide that you want to be in this study, this is what will happen. We will come to the place where you work or stay and we will perform 12 tests on you and your friends. Tests will happen over 1 day and each session will last about 3 hours and repeated again after two weeks (tests will be performed between breakfast and lunch).

2. PROCEDURE

On first arrival, tests and procedures will be explained and demonstrated and consent forms will be handed out. On my second visit, consent forms will be handed in for those wishing to participate. Descriptive (basic) information regarding your year of birth, age, sex, marital status and employment status, level of education and type of work will be collected. We will then proceed to measure body mass and body length measurements. Subsequently, 12 physical fitness tests will be analysed including: balance, muscular strength and endurance, functional and endurance test items. All of these tests have been adapted to those with intellectual disability or elderly in the general population. Also all of these tests have been conducted previously to 371 adults with Down syndrome with no complications. In fact, all of the participants

enjoyed the testing very much and wanted to know when I am coming back. These same tests will be repeated again after two weeks.

3. CAN ANYTHING BAD HAPPEN TO ME

In the one test you will get tired and sweat a bit, your heart will beat fast, and you will breathe rapidly. People will be there to help you.

4. CAN ANYTHING GOOD HAPPEN TO ME?

This study will show you that exercise is healthy and fun and we hope to learn something that will help other people some day

5. DO I HAVE CHOICES?

You can choose not to be in this study. You may withdraw at any time you please

6. WILL ANYONE KNOW I AM IN THE STUDY?

We won't tell anyone you took part in this study. When we are done with this study, we will write a report about what we found out. We won't use your name in the report.

7. WHAT HAPPENS IF I FEEL ANXIOUS OR SCARED?

If you fall or feel unwell, someone will be there to help and comfort you and your parent/guardian will help you get better. If you feel shy, a professional worker from the place where you stay will be there to help and comfort you. Similarly if you feel anxious or scared a professional worker from the place where you stay will be there to help and comfort you.

8. WHAT IF I DO NOT WANT TO DO THIS?

You don't have to be in this study. It's up to you. If you say yes now, but you change your mind later, that's okay too. All you have to do is tell us. If you have any more questions please ask your parent/guardian or me. If you want to be in this study, please sign or print your name.

9. QUESTIONS

You are welcome to ask any questions before you decide to give consent. You can call me directly on (082 672 2729) and Prof. S.J. Moss, the project leader (018 299-1821).

10. FEEDBACK OF FINDINGS

The findings of the research will be shared with you in a feedback session to be scheduled for the participants. You are welcome to contact us regarding the findings of the research. We will be sharing the findings with you as soon as it is available.

CONSENT FORM

for participating in a research study entitled:

The functional fitness capacity of adults with Down syndrome

PARTICIPATION IN THIS RESEARCH IS VOLUNTARY (you decide whether you want to participate).
You are free to decline to take part in this study, or to withdraw at any point even after you have signed the form to give consent, without any consequences.

Should you be willing to participate you are requested to complete and sign below:

I, _____ hereby voluntarily consent to participate in the above-mentioned study. I am not coerced (forced) in any way to participate and I understand that I can withdraw at any time should I feel uncomfortable during the study. I also understand that my name will not be disclosed (revealed) to anybody who is not part of the study and that the information will be kept confidential (private) and not linked to my name at any stage. I also understand what I might benefit from participation as well as what might be the possible risks and should I need further information, someone will be available to assist me.

_____ _____

Date Signature of the participant

_____ _____

Date Signature of the person obtaining consent

APPENDIX F: CONSENT FORM

CHAPTER FOUR

NORTH-WEST UNIVERSITY
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The functional fitness capacity of adults with Down syndrome (Study 2)

CONSENT FORM (simpler language): RESEARCH PARTICIPANT

I, PIETER-HENK BOER, am working on a research project involving a descriptive study conducted on Down syndrome adults. We would like to invite you to give consent to participate in our study.

PURPOSE OF THE STUDY

We would like you to participate in a research study where we want to find out how fit people with Down syndrome are. If you decide that you want to be in this study, this is what will happen. We will come to the place where you work or stay and we will perform 4 tests on you and your friends. Tests will happen over 3 days and each session will last about 1 hour and (tests will be performed between breakfast and lunch).

PROCEDURE

On first arrival, tests and procedures will be explained and demonstrated and consent forms will be handed out. On my second visit, consent forms will be handed in for those wishing to participate. Descriptive (basic) information regarding your year of birth, age, sex, marital status and employment status, level of education and type of work will be collected. We will then proceed to measure body mass and body length measurements. A sit-to-stand (30 seconds) will also be performed. Subsequently, 1 physical fitness test will be analysed (either a 6 minute walking test or a running test on the treadmill or a running test on the field) All of these tests have been adapted to those with intellectual disability or elderly in the general population. Also all of these tests have been conducted previously to 371 adults with Down syndrome with no

complications. In fact, all of the participants enjoyed the testing very much and wanted to know when I am coming back. For two more non-consecutive days the other two tests which you have not completed but will be done. In other words, by the end of the study three tests would have been performed.

CAN ANYTHING BAD HAPPEN TO ME

In the one test you will get tired and sweat a bit, your heart will beat fast, and you will breathe rapidly. People will be there to help you.

CAN ANYTHING GOOD HAPPEN TO ME?

This study will show you that exercise is healthy and fun and we hope to learn something that will help other people some day

DO I HAVE CHOICES?

You can choose not to be in this study. You may withdraw at any time you please

WILL ANYONE KNOW I AM IN THE STUDY?

We won't tell anyone you took part in this study. When we are done with this study, we will write a report about what we found out. We won't use your name in the report.

WHAT HAPPENS IF I FEEL ANXIOUS OR SCARED?

If you fall or feel unwell, someone will be there to help and comfort you and your parent/guardian will help you get better. If you feel shy, a professional worker from the place where you stay will be there to help and comfort you. Similarly if you feel anxious or scared a professional worker from the place where you stay will be there to help and comfort you.

WHAT IF I DO NOT WANT TO DO THIS?

You don't have to be in this study. It's up to you. If you say yes now, but you change your mind later, that's okay too. All you have to do is tell us. If you have any more questions please ask your parent/guardian or me. If you want to be in this study, please sign or print your name.

QUESTIONS

You are welcome to ask any questions before you decide to give consent. You can call me directly on (082 672 2729) and Prof. S.J. Moss, the project leader (018 299-1821).

FEEDBACK OF FINDINGS

The findings of the research will be shared with you in a feedback session to be scheduled for the participants. You are welcome to contact us regarding the findings of the research. We will be sharing the findings with you as soon as it is available.

CONSENT FORM

for participating in a research study entitled:

The functional fitness capacity of adults with Down syndrome

PARTICIPATION IN THIS RESEARCH IS VOLUNTARY (you decide whether you want to participate).
You are free to decline to take part in this study, or to withdraw at any point even after you have signed the form to give consent, without any consequences.

Should you be willing to participate you are requested to complete and sign below:

I, _____ hereby voluntarily consent to participate in the above-mentioned study. I am not coerced (forced) in any way to participate and I understand that I can withdraw at any time should I feel uncomfortable during the study. I also understand that my name will not be disclosed (revealed) to anybody who is not part of the study and that the information will be kept confidential (private) and not linked to my name at any stage. I also understand what I might benefit from participation as well as what might be the possible risks and should I need further information, someone will be available to assist me.

_____ _____

Date Signature of the participant

_____ _____

Date Signature of the person obtaining consent

APPENDIX G: CONSENT FORM

CHAPTER FIVE AND SIX

NORTH-WEST UNIVERSITY
YUNIBESITI YA BOKONE-BOPHIRIMA
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pieterhenk.boer@nwu.ac.za

The influence of sprint interval training on physical, metabolic and anthropometric variables in adults with Intellectual Disability and Down syndrome (Study 3 and 4)

CONSENT FORM (simpler language): RESEARCH PARTICIPANT

I, PIETER-HENK BOER, am working on a research project involving a specific exercise intervention on individuals with Intellectual Disability and Down syndrome. We would like to invite you to give consent to participate in our study.

PURPOSE OF THE STUDY

We would like you to participate in a research study where we want to find out how interval training (repeated faster cycling for 30 seconds and then slower cycling for 30 seconds) influences body composition, health and fitness.

PROCEDURE

If you decide that you want to be in this study, this is what will happen. On first arrival, tests and procedures will be explained and consent forms will be handed out. All tests will be explained and demonstrated. On my second visit, descriptive (basic) information regarding your year of birth, age, sex, marital status and employment status, level of education and type of work will be collected. You will also be familiarised with all testing procedures and equipment.

On my third visit, we will come to the place where you work or stay and we will perform body mass, body length, body composition, blood pressure and waist circumference measurements. We will then perform finger pricks to analyse glucose, insulin triglycerides and total cholesterol. Lastly, testing will conclude with the 6 minute walk and sit-to-stand tests. During the 6 minute walk test, you walk for 6 minutes in a rectangle to cover as much distance as possible. During the sit-to-stand

test you sit on a straight back chair, feet flat on the floor, and arms across the chest. On the signal go, you rise to a full stand, then return to a fully seated position. You try to do as many stands that you can in 30 seconds

The 4th visit will continue two days later and will only involve the maximal aerobic exercise test. During this test you will cycle starting at a very slow pace which will increase progressively until you cannot keep up with the pace anymore. You can stop the test at any time that you would like to. If you feel tired or scared you my stop the test at any time. There will be a professional person from the care facility where you stay to comfort you. Additional to the fourth visit: Dietary intake and analysis will be done using the observation method (Patton 2002). Each of the participants will be openly observed by a researcher on 1 day, for between 8 and 11 h. This will be done during all meal times as well as during leisure time. Each food and beverage item and time of its consumption will be recorded by the observer. The day of observation and starting time (morning or afternoon) will be chosen at the convenience of the participant and caretakers. If you feel tired or scared you my stop the test at any time. There will be a professional person from the care facility where you stay to comfort you. If you feel that you do not want to be observed, that is also okay, and the nutritionist will stop observing you. The 12-week intervention period will start where you will be required to exercise for 30 minutes, three times a week. If you cannot exercise for 30 minutes

All of the above-mentioned tests will be repeated after the 12-week intervention.

CAN ANYTHING BAD HAPPEN TO ME

In the maximum aerobic exercise test and the 6 minute walk test, you will get tired and sweat a bit, your heart will beat fast, and you will breathe rapidly. The finger prick might hurt a little bit. A person from your care facility will be there to help you.

CAN ANYTHING GOOD HAPPEN TO ME?

This study will show you that exercise is healthy and fun and we hope to learn something that will help other people some day

DO I HAVE CHOICES?

You can choose not to be in this study. You may withdraw at any time you please

WILL ANYONE KNOW I AM IN THE STUDY?

We won't tell anyone you took part in this study. When we are done with this study, we will write a report about what we found out. We won't use your name in the report.

WHAT HAPPENS IF I GET ANXIOUS OR SCARED?

If you fall or feel unwell, someone will be there to help you and your parent/guardian will help you get better. If you feel shy, a professional worker from the place where you stay will be there to help and comfort you. Similarly if you feel anxious or scared a professional worker from the place where you stay will be there to help and comfort you.

WHAT IF I DO NOT WANT TO DO THIS?

You don't have to be in this study. It's up to you. If you say yes now, but you change your mind later, that's okay too. All you have to do is tell us. If you have any more questions please ask your parent/guardian or me. If you want to be in this study, please sign or print your name.

QUESTIONS

You are welcome to ask any questions before you decide to give consent. You can call me directly on (082 672 2729) and Prof. S.J. Moss, the project leader (018 299-1821). No remuneration will be given for participating in this study.

FEEDBACK OF FINDINGS

The findings of the research will be shared with you in a feedback session to be scheduled for the participants. You are welcome to contact us regarding the findings of the research. We will be sharing the findings with you as soon as it is available.

CONSENT FORM

for participating in a research study entitled:

The influence of sprint interval training on physical, metabolic and anthropometric variables in adults with Intellectual Disability and Down syndrome

PARTICIPATION IN THIS RESEARCH IS VOLUNTARY (you decide whether you want to participate).
You are free to decline to take part in this study, or to withdraw at any point even after you have signed the form to give consent, without any consequences.

Should you be willing to participate you are requested to complete and sign below:

I, _____ hereby voluntarily consent to participate in the above-mentioned study. I am not coerced (forced) in any way to participate and I understand that I can withdraw at any time should I feel uncomfortable during the study. I also understand that my name will not be disclosed (revealed) to anybody who is not part of the study and that the information will be kept confidential (private) and not linked to my name at any stage. I also understand what I might benefit from participation as well as what might be the possible risks and should I need further information, someone will be available to assist me.

_____ _____

Date Signature of the participant

_____ _____

Date Signature of the person obtaining consent

APPENDIX H: ETHICAL APPROVAL

(NWU-00064-14-A1)

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Prof SJ Moss
PhASRec

23 June 2014

Dear Prof Moss

Ethics Application: NWU-00064-14-A1

Effect of aerobic training on body composition, physical and metabolic parameters in adults with intellectual disability and Down syndrome

Thank you for amending your application. All ethical concerns have now been addressed and ethical approval is granted.

Yours sincerely

Dr GW Towers
Human Research Ethics Committee Vice-Chairperson

Original details: Dr GW Towers(12686417) C:\Users\13210572\Documents\ETIEK\2014 ETHICS\NWU-00051-14-S1 - PH Boer Goedkeuring.docm
23 June 2014

File reference: NWU-00064-14-S1

APPENDIX I: ADAPTED PHYSICAL ACTIVITY READINESS QUESTIONNAIRE

ADAPTED PHYSICAL ACTIVITY READINESS QUESTIONNAIRE (PAR-Q)
DATE (day/month/year): _____

Regular physical activity is fun and healthy and more people should increase their physical activity every day. Being more physically active is very safe for MOST people.

Please read the questions carefully and answer each one honestly. If you have any concerns about your health status, you should check with your doctor before becoming more physically active.

Question	Yes	No
1. Has your doctor ever said that you have a heart condition OR high blood pressure?		
2. Do you feel pain in your chest at rest, during your daily activities of living, OR when you do physical activity?		
3. Do you lose balance because of dizziness OR have you lost consciousness [fainted] in the last 12 months?		
4. Have you ever been diagnosed by a health professional as having any of the following (Check (v') all that apply: Heart Trouble High blood pressure High cholesterol Arthritis Chronic asthma Emphysema Bronchitis Back problems Foot problems Allergies Diabetes Trouble hearing Trouble seeing		
5. Are you currently taking any medication for any of the conditions listed above? Please describe:		
6. Do you have a bone or joint problem that could be made worse by becoming more physically active? (if you had a joint problem in the past e.g. knee, ankle, shoulder, please answer NO to this question).		
7. Has your Doctor, Nurse Practitioner (or health provider) ever said that you should only do medically supervised physical activity?		
I have read and understood the above health questions and direction regarding my participation in the Fit, Fun & Fully Alive! Group Fitness Classes. Your Initials: _____		

IF YOU ANSWERED YES to one or more of the questions above, you should consult your doctor or health provider first before becoming more physically active. Talk with your doctor about the kinds of activities you wish to participate in and follow his/her advice.

IF YOU ANSWERED NO to all the questions above, you can be reasonably sure that you can start becoming more physically active. Begin slowly and build up gradually. Delay becoming more active if you are not feeling well because of a temporary illness such as a cold or a fever wait until you feel better.
If your health changes so that you would answer YES to any of the PAR-Q questions, ask for advice from your health professional and let your Fitness Instructor know.

APPENDIX J: LANGUAGE EDITING PROOF

HESCOM
COMMUNICATION SERVICES

Translating.Writing.Editing

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LANGUAGE EDITING STATEMENT

2015-10-28

PhD in Human Movement Science: *Effect of continuous aerobic vs interval training on selected functional fitness parameters of adults with intellectual disability and Down syndrome*

by PH BOER

- Has been edited for language correctness and spelling.
- Has been edited for consistency (repetition, long sentences, logical flow)
- Has been checked for completeness of list of references and cited authors.

No changes have been made to the document's substance and structure (nature of academic content and argument in the discipline, chapter and section structure and headings, order and balance of content, referencing style and quality).

HESTER VAN DER WALT

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