Footwear, Foot Dimensions and Physical Activity in Children with Down syndrome

Submitted by

Nirmeen Hassan

Bachelor of Health Sciences and Master of Podiatric Practice

A thesis submitted in total fulfilment of the requirement for the degree of Doctor of Philosophy.

Discipline of Podiatry

School of Allied Health, Human Services and Sport

College of Science Health and Engineering

La Trobe University

Victoria 3086

Australia

September 2020

Table of contents

List of figures

List of tables

Statement of authorship

I, Nirmeen Hassan, declare that this is a submission of original work and except where reference is made in the text of the thesis, this thesis contains no material published elsewhere or extracted in whole or part from a thesis submitted for the award of any other degree or diploma.

I, Nirmeen Hassan, also declare that no work by others has been used without due acknowledgement in the main text of the thesis. This thesis has not been submitted for the award of any other degree or diploma at any other tertiary institution. In terms of the extent of collaboration with other authors, although publications included in this thesis include joint authorship, I have made a significant and leading contribution to the work, equivalent to that expected for a traditional thesis. I am the first author on all publications included in this thesis.

All research studies reported in this thesis were approved by the relevant ethics committee ([La Trobe University Human Ethics Committee] HEC13-035, HEC16-027 and HEC19-290).

Signed: 

Date: 25/09/2020

Acknowledgements

I would like to begin by sincerely thanking my supervisors, Associate Professor Shannon Munteanu, Professor Karl Landorf and Professor Nora Shields, for their endless support and assistance throughout the duration of my candidature. Their efforts, guidance and willingness to assist me throughout the process has been invaluable. Beyond the completion of my thesis, my supervisors taught me valuable skills that will carry over to future endeavours and have introduced me to opportunities I would have otherwise, not have experienced. I would also like to thank Professor Hylton Menz, for his assistance and support throughout my candidature. His knowledge and expertise in the field of research has been priceless.

I would also like to acknowledge my parents, Mohamed Hassan and Nagwa Taha, for encouraging me to pursue a degree in the research field and for always supporting me. I would also like to thank my other half, Zaki Benamar, who has supported me since day one.

Publications and presentations

Publications

Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. *Journal of Intellectual Disability Research.* 2019;63(2):168–191. DOI: https://doi.org/10.1111/jir.12562

Hassan NM, Shields N, Landorf KB, Buldt AK, Taylor NF, Evans AM, Williams CM, Menz HB, Munteanu SE. Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study. *Disability and Rehabilitation*. 2019;22:1–10. DOI: https://doi.org/10.1080/09638288.2019.1692380

Hassan NM, Buldt AK, Shields N, Landorf KB, Menz HB, Munteanu SE. Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome. *Journal of Foot and Ankle Research.* 2020;13:31. DOI: https://doi.org/10.1186/s13047-020-00403-1

Hassan NM, Buldt AK, Shields N, Landorf, KB, Menz HB, Munteanu SE. Differences in foot dimensions between children and adolescents with and without Down syndrome. *Manuscript submitted for publication (Disability and Rehabilitation)*; June 1, 2020.

Presentations

Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. Australasian Academy of Cerebral Palsy and Developmental Medicine Conference. 21 to 24 March 2018, Auckland, New Zealand.

Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. VIC State Podiatry Conference. 27 to 28 April 2018, Melbourne, Australia.

Funding

This work was supported by an Australian Government Research Training Program Scholarship (2015) and a La Trobe University Postgraduate Research Scholarship. The Research Focus Area – Sport, Exercise and Rehabilitation also funded one study ($24,143) included in this thesis (randomised pilot study presented in Chapter 3). I was also awarded a School of Allied Health, Human Services and Sport Higher Degree Research grant ($5,000) to support research-related costs associated with projects presented in this thesis.

Summary

Down syndrome is a chromosomal condition that affects the musculoskeletal system, including the foot. Children with Down syndrome have structural variations to their feet, which complicates footwear fit. Consequently, many children with Down syndrome wear poorly-fitting footwear. This can have negative consequences, including reducing levels of physical activity. Therefore, the aims of this thesis are to determine: (i) the effectiveness of interventions (including custom-fitted footwear to improve fit) to increase physical activity in children with Down syndrome; and (ii) the foot dimensions of children with Down syndrome to better understand footwear fitting issues in this population.

Four studies were conducted. The first study was a systematic review of nine randomised controlled trials that evaluated the effectiveness of interventions to improve physical activity in individuals with intellectual disabilities. A key finding was that only a limited number of physical activity interventions were effective in increasing physical activity in this population. These were a gym-based progressive resistance training program, a multi-component diet and physical activity program and a physical activity framework and education program. No previous studies investigated the effects of footwear on physical activity in individuals with intellectual disabilities, including those with Down syndrome. This warranted further investigation because poorly-fitting footwear has been associated with reduced physical activity.

The second study was a randomised pilot study that determined the feasibility of conducting a definitive randomised trial to evaluate the efficacy of custom-fitted footwear to increase physical activity in children with Down syndrome. Thirty-three children with Down syndrome (mean age 9.7 [3.6] years, 21 girls) were randomly allocated to an intervention group (custom-fitted footwear) or a wait-list control group. Based on Bowen's framework, six domains of feasibility were evaluated: demand (recruitment), implementation (co-interventions and adherence), acceptability (satisfaction with the intervention), practicality (adverse events), limited efficacy testing (physical activity; disability associated with foot and ankle problems, and gait) and adaptation (shoe-fit). Outcome measures were assessed at baseline, 6 weeks and 12 weeks. The results showed trends for differences in physical activity that favoured custom-fitted footwear; however, the differences were small. The main finding was that the fit of the footwear intervention – despite being fitted according to the manufacturer's protocol – was no better than participants' regular footwear. The footwear the participants were provided with was too narrow to accommodate foot width. These findings justified the need to better understand the foot dimensions of children with Down syndrome.

The third study investigated the reproducibility of measuring foot dimensions of children with Down syndrome. Three-dimensional (3D) foot scans of 30 children with Down syndrome (mean age 10.6 [3.9] years, 18 girls) were obtained to determine the intra- and inter-rater reproducibility of 13 unique foot dimension measurements. These measurements related to length (foot length, ball of foot length, outside ball of foot length), width (diagonal and horizontal foot width, heel width), girth (ball and instep girth), height (first and fifth toe height, instep height), forefoot shape (length of the digits, classified according to longest toe) and the ratio of foot length to foot width (Wejsflog Index). Two raters completed the measurements independently, 2 weeks apart using 3D Tool© and Canvas© software programs. All measurements were found to have moderate to excellent reliability (intra-class correlation coefficients [ICCs] ranging from 0.73 to 0.99). Seven measurements had narrow limits of agreement (LOA) values (foot length, diagonal foot width, horizontal foot width, Wejsflog Index, instep height, first toe height and fifth toe height), but the remaining measurements (ball of foot length, outside ball of foot length, heel width, ball girth and instep girth) had wider LOA values indicating poorer agreement.

As the measurements of foot dimensions were found to be reproducible, the fourth study was a cross-sectional observational study that compared the foot dimensions of children with and without Down syndrome. 3D foot dimensions of 51 children with Down syndrome were compared to 51 typically developing children (using 3D foot scans), who were age (±2 years) and sex-matched (mean age 10.8 [3.7] years, 28 girls). Data were analysed as absolute and normalised (for scale) differences due to the wide age range (5 to 20 years) of participants. Results showed children with Down syndrome have smaller feet (absolute length, width and girth measurements). Further, after normalising for scale, children with Down syndrome were found to have a shorter foot length (heel to toe measurement), but a longer ball of foot length (heel to ball measurement), wider forefoot, a greater girth circumference (ball and instep girth) and greater fifth toe height. These differences in dimensions may explain why children with Down syndrome commonly wear poorly-fitting footwear.

Collectively, these studies identified that footwear fit is a problem for children with Down syndrome. Footwear may potentially improve physical activity in children with Down syndrome; however, improving footwear fit is a necessary first step. At present, commercially available footwear is unlikely to accommodate the unique foot structure of children with Down syndrome. Children with Down syndrome need footwear that is wide, has adequate girth and a deep toe box height at a given size (length fit). Footwear with the required extra-depth and width (known as medical-grade footwear) is not readily available for children. Developing footwear according to normative dimensional data for

children with Down syndrome, or mass-customisation based on new technologies could solve the issue of poorly-fitting footwear for children with Down syndrome.

Chapter 1 – Introduction

1.1 Down syndrome

Down syndrome is a chromosomal disorder, characterised by unique facial features, intellectual disability and impairments to all body systems [1]. It affects 1 in 650 to 1,000 births worldwide. It is estimated that 5,400 infants are born each year with Down syndrome in the United States and approximately 290 per year in Australia [2]. The prevalence of Down syndrome is increasing when both live births and terminated pregnancies are included [3]. There are two main reasons for this. First, a greater proportion of women are giving birth later in life. Approximately 22% of women who gave birth in 2004 were 35 years or over, which is substantially higher than the 8% of women who were aged 35 or over in 1985 [3]. Second, there has been an improvement in the sensitivity of diagnostic screening during early pregnancy to detect foetuses with Down syndrome [3, 4]. Children with Down syndrome have increased risks of health issues that lead to reduced life expectancy. However, the life expectancy of those with Down syndrome has improved in recent years, primarily due to advances in cardiac surgery [5]. At present, the estimated life expectancy of adults with Down syndrome is 60 years [6, 7]. Approximately 90% of children with Down syndrome live beyond five years and 85% live beyond 10 years [8].

1.1.1 Pathophysiology of Down syndrome

Down syndrome is the most common chromosomal abnormality and occurs as a result of additional genetic material related to chromosome 21. This may occur in three ways. The first, known as trisomy 21, occurs when an error in cell division results in an embryo with three copies of chromosome 21 instead of two [9-11]. It is the most common form of Down syndrome, with 95% of children with Down syndrome having this anomaly. The second, known as translocation, occurs when genetic material is re-arranged. There are three copies of chromosome 21, and one copy is attached (i.e. rearranged) to another chromosome, usually chromosome 14 or 15 [12]. Translocation occurs in 3 to 4% of children with Down syndrome and can be inherited [12]. The third, known as mosaicism, occurs when both normal cells and trisomy 21 cells are present, which may occur in two ways: (i) when a normal zygote with 46 chromosomes experiences an early mitotic error that leads to some cells with trisomy 21, or (ii) an early mitotic error causes some cells in an embryo with trisomy 21 to revert to a normal karyotype [13]. Mosaicism is the rarest form of Down syndrome and is present in 1 to 2% of children with Down syndrome [14].

There are a number of risk factors for Down syndrome, including paternal origin Down syndrome (where Down syndrome originated from the father), impaired folate metabolism, and increased maternal weight [15]. However, the two most well-known risk factors for Down syndrome are increased maternal age and altered recombination (a process where components of DNA are broken and recombined to produce new combinations of alleles [a variant form of a gene]) [16].

1.1.2 Systemic characteristics of Down syndrome

The clinical presentation of Down syndrome varies between individuals. However, there are common features that are present to a varied extent in individuals with Down syndrome. This includes systemic changes to several body systems, including the central nervous, cardiovascular, endocrine and musculoskeletal systems. As this thesis primarily focuses on musculoskeletal characteristics, these will be discussed separately in Section 1.1.3.

The effects of Down syndrome on the central nervous system includes intellectual disability and cognitive decline. The degree of intellectual disability is often moderate in severity but can range from mild to severe [17-19]. Additionally, cognitive decline can occur in the later stages of life. Individuals with Down syndrome are at high risk of early onset Alzheimer's disease; over 75% of individuals with Down syndrome over the age of 65 years will have a clinical diagnosis of dementia [20]. The brains of those with Down syndrome show granulovacuolar cytoplasmic changes, senile plaques and cerebrovascular amyloid similar to those with Alzheimer's disease [21]. The risk of Alzheimer's disease has emerged more recently for individuals with Down syndrome due to their increased life expectancy [22]. For this reason, regular physical activity in this population is important as it may be protective against Alzheimer's disease due to its positive effects on brain health [23]. Therefore, improving physical activity in children with Down syndrome is important to establish positive lifestyle behaviours early in life.

Congenital heart disease occurs in approximately 40 to 50% of children with Down syndrome [24]. The most common forms of congenital heart disease are atrioventricular septal defects, ventricular septal defects and atrial septic defects [25]. Congenital heart disease is the leading cause for mortality and morbidity for children with Down syndrome, particularly in the first two years of life [26]. Congenital heart defects can affect cardiorespiratory capacity and physical fitness. Cardiovascular fitness is reduced in children with Down syndrome due to reduced aerobic capacity or VO_2peak (the maximum uptake of oxygen during incremental exercise). Children with Down syndrome also have reduced peak heart rate, which also contributes to their reduced aerobic

capacity [27]. Other anatomical and physiological variations found in individuals with Down syndrome (such as smaller nasal/oral cavities, muscle hypotonia and a narrowed aorta) may also limit cardiorespiratory capacity, thereby affecting their ability to engage in physical activity [27].

Down syndrome is also associated with endocrinopathies such as thyroid dysfunction, low bone mass, and a greater predisposition to obesity [28]. The most common form of thyroid dysfunction is hypothyroidism (which occurs in 4 to 8% of children with Down syndrome [29]), but specific autoimmune thyroid disorders such as Graves or Hashimoto's disease may also occur [28, 29]. Thyroid disease in Down syndrome is often transient, and is not associated with sex, comorbidities or obesity [30]. Low bone mass affects children with Down syndrome as bone development is impaired by reduced physical activity, decreased sun exposure, obesity, mineral deficiencies (vitamin D and calcium), reduced muscle mass and malabsorption syndromes, all of which are more common in children with Down syndrome [31]. Low bone mineral density worsens with increased age [32] and may increase the risk of fractures and osteoporosis in later life [33]. Children with Down syndrome also have a higher predisposition for obesity compared to typically developing children, with the prevalence varying from 23 to 70% [34]. Children with Down syndrome have less lean muscle mass and more fat mass, even when adjusted for BMI [35]. This higher predisposition for obesity is speculated to be associated with increased leptin (a hormone that is released from fat cells to regulate energy balance), reduced basal metabolic rate, sedentary behaviour and poor diet [34]. Obesity contributes to morbidity and mortality, and plays a major role in the development of chronic health conditions such as type 2 diabetes mellitus and heart disease [36].

1.1.3 Musculoskeletal characteristics of Down syndrome

The musculoskeletal system is affected by Down syndrome in two major ways; hypotonia (reduced muscle tone) and ligamentous laxity (increased joint mobility beyond normal range of motion) [37, 38]. Together, these features can impact the function of children with Down syndrome, as evidenced by delayed developmental milestones [39], reduced gait stability, increased energy expenditure during movement and reduced physical activity [38]. Hypotonia and ligamentous laxity also alter joint kinetics and kinematics, which reduces exercise economy (a term that refers to how much speed or power is developed at a certain level of $V0_2$) [40].

Hypotonia and ligamentous laxity are risk factors for the development of musculoskeletal conditions [41], which affect approximately 20 to 63% of individuals with Down syndrome [42, 43]. Such conditions include atlanto-axial instability, scoliosis, hip subluxation and

knee instability [44-47] (Table 1). These musculoskeletal conditions can be progressive, are often characterised by pain and disability, and can adversely affect physical activity. Low physical activity can lead to secondary chronic health conditions such as osteoporosis, obesity and diabetes. In this way, musculoskeletal conditions can have significant effects on the physical activity and quality of life of individuals with Down syndrome [38, 48, 49].

Children with Down syndrome are at an increased risk of arthritis in early life. Inflammatory arthritis in children with Down syndrome is often polyarticular rheumatoid factor negative and mostly affects the small joints of the hands and wrists [50]. The prevalence of arthritis in children with Down syndrome is 8.7 to 10.2 in every 1000 children [50]. This is two to three times greater than previously reported [50]. Arthritis in children with Down syndrome is often misdiagnosed or has a delayed diagnosis from onset of disease. It may be asymptomatic but can lead to erosive damage to the joints and functional changes [50]. If left undiagnosed or untreated, it can lead to permanent joint damage and disability [50, 51].

Table 1. Common musculoskeletal conditions associated with Down syndrome.

Musculoskeletal condition	Definition	Prevalence	Potential outcomes
Atlanto-axial instability [44, 52]	Increased mobility of C2 in relation to C1 (cervical vertebrae)	10 to 40%	▪ Neck pain ▪ Abnormal gait ▪ Spinal cord impingement
Scoliosis [46]	Sideways curvature of the spine	~10%	▪ Back pain ▪ Central nervous system effects ▪ Difficulty in breathing and sleeping
Hip dysplasia [47]	Abnormality of the hip socket in which the acetabulum does not fully cover the head of femur	1 to 7%	▪ Hip subluxation ▪ Abnormal gait ▪ Degenerative hip changes
Patella instability [45]	Inability of the patella to maintain its position within the trochlear groove during movement	4 to 8%	▪ Abnormal gait ▪ Pain ▪ Falls ▪ Reduced health-related quality of life

For children with Down syndrome, the two most common musculoskeletal conditions that affect the lower limb are hip dysplasia and patella instability (as outlined in Table 1

above). Hip dysplasia occurs in 1 to 7% of children with Down syndrome [47]. Although ligamentous laxity and hypotonia play a major role in hip dysplasia, it may also be attributed to anatomical irregularities, such as abnormalities of the acetabulum and the femur [53]. For example, there may be greater femoral anteversion (internal rotation of the femoral shaft), but a normal angle at the neck of femur. Instability of the hip in children with Down syndrome progresses with time [54]. Initially, there is hypermobility of the hip, which can delay the onset of walking. Following this, symptomatic hip dislocation can occur, accompanied with crepitus, limping and hip instability. Finally, the hip can begin to deviate from its centre alignment and acetabular dysplasia occurs, resulting in the hip developing into a rigid, painful position in late adolescence [54]. It is possible for hip instability to develop after skeletal maturity is reached [53]. In a study of 65 adults with Down syndrome, 28% of participants had hip abnormalities identified through radiographs, which included acetabular dysplasia and hip dislocation [55]. These radiographic changes have been correlated with reduced walking ability, suggesting that hip dysplasia may reduce independent mobility with increasing age, which is another contributor to reduced physical activity [55].

Compared to hip dysplasia, there is limited information on patella instability in children with Down syndrome. Patella instability occurs in approximately 4 to 8% of children with Down syndrome. Like hip dysplasia, ligamentous laxity and hypotonia are believed to contribute to the occurrence of patella instability. Other factors may also contribute to hip dysplasia such as femoral trochlear dysplasia and increased height of the patella [56]. Excessive foot pronation is more common in children with Down syndrome (see Section 1.2.1) and may also contribute to internal rotation of the tibia, which may worsen patella instability [57]. Patella instability can negatively affect gait and can have a subsequent adverse effect on physical activity [45].

1.2 Effects of Down syndrome on the foot

The feet of children with Down syndrome are usually affected in two ways: dermatological manifestations and structural variations. Briefly, dermatological manifestations include dermatomycoses (fungal infections of the skin, which may be related to immunological deficiencies), pressure-related lesions (which may be related to footwear fit and elevated plantar pressures), xerosis (dry skin around the heels) [58] and split toe nails [59]. Dermatological issues will not be discussed further as they are unrelated to the aims of this thesis.

Structural variations may be congenital (i.e. pes planus) or acquired (i.e. hallux valgus) and may have several negative outcomes. Structural variations may adversely affect

gait, contribute to the development of foot pain and negatively affect footwear fit. Few studies have focused on variations in foot structure and its sequalae in children with Down syndrome. This is despite approximately 30% of orthopaedic complaints in Down syndrome arising from the foot [60].

The following sections focus on three important areas, which provide the basis for this thesis: variations in foot structure, the measurement of foot structure in children with Down syndrome focussing on the potential of 3D scanning technology, and the implications of structural variations on footwear fit.

1.2.1 Variations in foot structure of children with Down syndrome

The feet of children with Down syndrome exhibit differences in structure compared to typically developing children. Structural variations that have been observed include lesser toe deformities (i.e. partial/complete syndactyly [webbing]; and sagittal plane deformities [claw or mallet toes]) [61-63], hallucal gap (space between the first and second toe) [63], isolated metatarsus primus varus [61], metatarsus primus varus with hallux valgus deformity [61], hallux valgus [61-64], pes planus [59, 61-66] and isolated calcaneal valgus [61] (Table 2). A large proportion of these structural variations (i.e. lesser toe deformities, syndactyly, clinodactyly, hallucal gap, isolated metatarsus primus varus, hallux valgus and metatarsus primus varus with hallux valgus deformity) affect the forefoot. The structural variations with the highest prevalence are a pes planus foot type (92%) [65] and hallucal gap (74%) [63]; however, hallux valgus (45%), isolated metatarsus primus varus (40%) and metatarsus primus varus with hallux valgus deformity (34%) are also common.

It is important to acknowledge that prevalence rates vary between studies. For example, one study showed that metatarsus adductus was observed in 4% of children with Down syndrome [63]. However, another study reported metatarsus adductus to be present in 48% of children with Down syndrome [65]. Another example is the prevalence of hallux valgus, which was reported as 36% in one study [63] and 45% in another study [64], both markedly higher than 10 to 15% reported in other studies [61, 62]. The differences in reported rates may be due to the methods used to collect data. For example, when clinical observation was used to report on hallux valgus, lower rates were found when compared to radiographic evaluation, which showed higher rates of hallux valgus [64]. Nevertheless, these findings signify the differences in the foot structure of children with Down syndrome, with most variations affecting the forefoot. This has implications on footwear fit as the differences in forefoot structure increase forefoot width and toe box requirements of footwear.

Table 2. A summary of studies that have evaluated the variation in foot structure in individuals with Down syndrome.

Study	Study design	Participants	Measurements	Key findings (prevalence)	
				Children with Down syndrome	**Children without Down syndrome**
Prasher et al. 1995 [59]	Case-matched study	Age: 7 to 14 years Groups: (i) 50 with Down syndrome (29 boys, 21 girls) (ii) 50 with learning disabilities (32 boys, 18 girls) (iii) 50 age-matched controls (20 boys, 30 girls)	Visual observation for podiatric anomalies (i.e. general appearance of the foot, assessment for toe abnormalities, toenail health, foot alignment and skin health)	Pes planus foot (58%) Abnormal pressure prints (34%)	*Children with learning disabilities* Pes planus foot (20%) Abnormal pressure prints (36%) *Age-matched controls* Pes planus foot (20%) Abnormal pressure prints (14%)
Concolino et al. 2006 [61]	Case-matched study	Age: 3 to 8 years Groups: (i) 50 children with Down syndrome (19 boys, 31 girls) (ii) 100 children without Down syndrome (32 boys, 68 girls)	Complete podiatric examination Podoscopic evaluation Static and dynamic baropodometric examination of the lower limb	Pes planus (60%) Isolated hallux valgus (26%) Isolated metatarsus primus varus (40%) Hallux valgus and metatarsus primus varus (34%) Syndactyly (10%) Clinodactyly (6%) Isolated calcaneal valgus (24%)	Pes planus (10%) Isolated hallux valgus (10%) Isolated metatarsus primus varus (0%) Hallux valgus and metatarsus primus varus (0%) Syndactyly (2%) Clinodactyly (0%) Isolated calcaneal valgus (6%)
Lim et al. 2014 [62]	Cross-sectional observational study	Age: 5 to 18 years Participants: 50 children with Down syndrome (28 boys, 22 girls)	Foot Posture Index Arch Index Hallux valgus Lesser toe deformities	Pes planus (76%) Hallux valgus (10%) Lesser toe deformities (12%)	N/A
El Mansour et al. 2017 [63]	Case-matched study	Age: 14.6 years (mean age of case group) and 13.5	Podoscopic assessment for foot deformities	Pes planus: Grade 2 (39%) Grade 3 (30%)	Pes planus: Grade 2 (15%), Grade 3 (30%)

		years (mean age of control group) Groups: (i) 55 children with Down syndrome (36 boys, 19 girls) (ii) 53 age-matched typically developing children (27 boys, 26 girls)	Footprints via podoscope	Hallux valgus (36%) Hallucal gap (74%) Increased space between hallux, second toe and hallux valgus (17%) Syndactyly (13%) Clinodactyly (16%)	Hallux valgus (7%) Hallucal gap (3%) Increased space between hallux, second toe and hallux valgus (0%) Syndactyly (0%) Clinodactyly (5%)
Puszczalowska et al. 2017 [66]	Case-matched study	Age: 14 to 15 years Groups: (i) 30 adolescents with Down syndrome (boys only) (ii) 30 adolescents without Down syndrome (boys only)	Foot dimensions (podoscope) Angular measurements (hallux valgus angle and angle of varus deformity of the fifth toe)	Flatter longitudinal arch Shorter foot length Narrower foot length	Not reported
Calvo-Lobo et al. 2018 [65]	Case-matched study	Age: 15 to 63 years Groups: (i) 50 with Down syndrome (ii) 55 without Down syndrome (sex-ratio not reported)	Podoscopic evaluation Static and dynamic baropodometric examination of the lower limb (as described by Concolino et al. 2006)	Pes planus (92%) Metatarsus primus adductus (48%) Hypermobile first ray (52%)	Not reported
Perotti et al. 2018 [64]	Cross-sectional observational study	Age: 5 to 18 years Groups (101 children with Down syndrome): (i) 41 children foot radiographs (27 boys, 14 girls) (ii) 60 children with ankle radiographs (31 boys, 29 girls) (iii) 15 children with ankle and foot radiographs (11 boys, 4 girls)	Evaluations of radiographs of the foot and ankle	Pes planus according to clinical observation (46%) and radiographic observation (58%) Hallux valgus according to clinical observation (15%) and radiographic observation (45%)	N/A

1.2.2 Measurement of foot structure of children with Down syndrome

Anthropometric data of the foot forms the foundation of footwear design and production [67]. Appropriately fitting footwear relies on the consideration of several foot characteristics [68], such as foot dimensions that describe the 3-dimensional (3D) structure of the foot (Table 3). There is a lack of consistency across the literature in the terms used to describe foot dimensions. However, these measurements can be classified into four general categories relating to length, width, girth and height (Figure 1).

Table 3. Definitions of foot dimension measurements.

Foot dimensions [69-71]	
Length	
Foot length	Distance between foot end (pternion) and foot tip (anterior point of most protruding toe).
Ball of foot length	Distance between foot end (pternion) and the first metatarsophalangeal protrusion.
Outside ball of foot length	Distance between foot end (pternion) and the fifth metatarsophalangeal protrusion.
Width	
Diagonal foot width	Connecting line between the first metatarsophalangeal joint and the fifth metatarsophalangeal joint.
Horizontal foot width	Orthogonal connection line starting at the first metatarsophalangeal joint to the outside curvature of the foot.
Heel width	Maximum orthogonal connection line starting at the medial side of the heel to the outside curvature of the heel.
Girth	
Ball girth	Maximum circumference at the level of the first and the fifth metatarsophalangeal joint protrusion.
Instep girth	Maximum circumference measured from the most plantar aspect of the foot to the most dorsal aspect of the foot, at the level of the navicular.
Height	
First toe height	Maximum height of the hallux measured from the most plantar aspect of the hallux to the most dorsal aspect of the hallux.
Fifth toe height	Maximum height of the fifth toe measured from the most plantar aspect of the fifth toe to the most dorsal aspect of the fifth toe.
Instep height	Measured from the most plantar aspect of the foot to the most dorsal aspect of soft tissue.

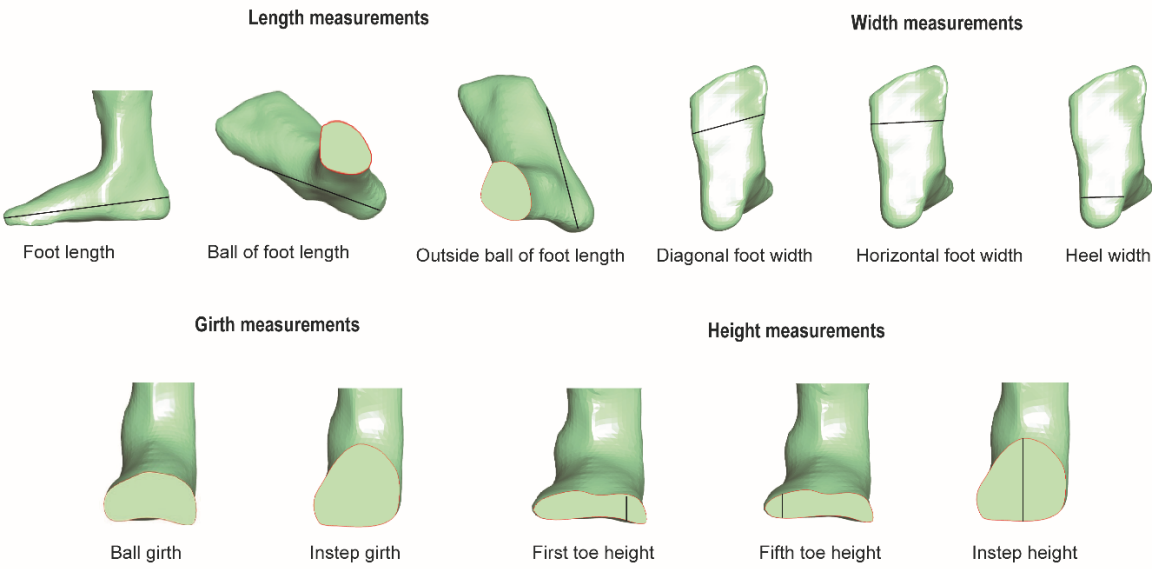


Figure 1. Foot dimensions measurements.

Historically, one- (1D) and two-dimensional (2D) measurements were used to develop the anthropometric databases used by footwear manufacturers to guide the design and manufacture of footwear in a range of sizes. There are numerous methods to collect anthropometric data, such as foot gauges (to measure length and width), callipers and footprints (which may describe the profile of the arch, such as the Arch Index). Although these methods provide useful information, 1D and 2D measurements offer limited detail of a 3D structure [72]. Additionally, these methods of measurement are subject to human error. Callipers rely on training experience and an understanding of landmark positioning, while ink-footprints can be affected by the quality of the ink. Further, neither 1D nor 2D measurements allow for important circumferential foot measurements (such as ball and instep girth) to be measured [73].

More recently, the development of surface scanning equipment such as 3D scanners and software has overcome many of the limitations of 1D and 2D measurements of foot structure. This technology allows for rapid data collection and the creation of a 3D digital representation of the human foot and has multiple applications. For example, 3D foot scanners have been used to determine the differences in foot shape between men and women [70, 71, 74, 75], and different ethnic populations [71, 76]. Understanding these variations is useful to aid the design and manufacture of footwear that can accommodate for unique foot shapes [77].

As children with Down syndrome have a characteristic foot structure, it would be beneficial to understand the detailed foot structure of children with Down syndrome and how this may differ to typically developing children. However, the reproducibility of using

3D foot scans to measure foot dimensions of children with Down syndrome has not been established. Establishing the reproducibility of such measurements is an important first step to using this technology. Applications of the technology could include exploring the foot dimensions of children with Down syndrome; using foot dimension data to support the design and manufacture of more appropriately fitting footwear for this population; and monitoring a child's foot posture over time.

1.2.3 Footwear fit in children with Down syndrome

Footwear has several important functions, such as protection, injury prevention, support and expression of personal style. These functions are governed by the appropriateness of footwear fit. When footwear is poorly-fitted, it is unable to fulfil its purpose and may cause foot pathology. This includes skin disorders (e.g. blistering, callus, corns), subungual hematomas, ingrown toenails, foot deformities (e.g. hallux valgus, lesser toe deformities) and foot pain (e.g. pain associated with the aforementioned issues) [78, 79]. Poorly-fitting footwear has been associated with reduced physical activity in children with Down syndrome [80].

Poorly-fitting footwear has been identified as an issue affecting children and adults with intellectual disability [59, 81]. Generally, the measurements of foot length and width guide footwear fitting. However, other measurements of footwear fit are also important, such as toe box depth, girth fit and instep height. Children with Down syndrome often do not wear appropriately fitting footwear (Table 4) due to the difficulty in matching their foot structure (wider forefoot relative to length) to the dimensions of footwear. Therefore, the prevalence of poorly-fitting footwear in this group is high, ranging from 60 to 88% [65, 80]. Four studies investigating footwear fit in individuals with Down syndrome (Table 4) (that have focused on length and width measures) show how foot width relative to foot length may be a key contributing factor in poorly-fitting footwear. These studies show the proportion of children with Down syndrome that wear narrow footwear is as high as 58%. One approach to address narrow fitting footwear is to purchase footwear that is longer (which subsequently has a greater width and girth), yet this is likely to result in footwear that is too long. Indeed, approximately 54% of children with Down syndrome wear footwear that is too long [80]. Commercially available footwear with additional width is difficult to find. As a consequence, children with Down syndrome are more likely to wear footwear that is an appropriate length but too narrow, or wear footwear that is too long but wide enough to accommodate their feet [62].

Table 4. Summary of studies evaluating footwear fit in individuals with Down syndrome.

Study	Study design	Participants	Measurements	Key findings	
				Children with Down syndrome	Children without Down syndrome
Prasher et al. 1995 [59]	Case-matched study	Age: 7 to 14 years Groups: (i) 50 with Down syndrome (29 boys, 21 girls) (ii) 50 with learning disabilities (32 boys, 18 girls) (iii) 50 age-matched controls (20 boys, 30 girls)	Visual observation for podiatric anomalies (i.e. general appearance of the foot, assessment for toe abnormalities, toenail health, foot alignment and skin health)	Poorly-fitting footwear (10%)	*Children with learning disabilities* Poorly-fitting footwear (6%) *Age-matched controls* Poorly-fitting footwear (0%)
Jenkins et al. 2012 [81]	Cross-sectional observational study	Age: median of 25.6 years (58.5% males, 41.5% females) Sample size: 4,094	Foot to shoe mismatch (footwear fit) using Brannock device Rate of referral	*Athletes with special needs* 29% wore shoes that were too big. 13% wore shoes that were too small. 41% had a mismatch of foot and shoe.	N/A
Shields et al. 2017 [80]	Prospective cohort study	Age: 5 to 18 years (28 boys, 22 girls) Sample size: 50 Children with Down syndrome	Foot posture (Arch Index) Visual observation of foot deformities Footwear fit Physical activity	10% of participants wore shoes that were too short. 58% wore shoes that were too narrow. 52% of participants wore shoes that were too long.	N/A
Calvo-Lobo et al. 2018 [65]	Case-matched study	Age: 15 to 63 years Groups: (i) 50 with Down syndrome (ii) 55 without Down syndrome (sex-ratio not reported)	Complete podiatric examination Podoscopic evaluation Static and dynamic baropodometric examination of the lower limb	Poorly-fitting footwear (76%) Appropriately fitting footwear (12%)	Poorly-fitting footwear (16%) Appropriately fitting footwear (84%)

Another issue that arises from wearing poorly-fitting footwear is an increased risk of reduced physical activity [80]. There is evidence that the greater the difference between footwear length and absolute foot length, the less physically active a child with Down syndrome is [80]. This may be explained by the interference of excessively long footwear on an individual's gait (i.e. reduced gait velocity to avoid tripping). This also increases the effort required to engage in physical activity, which may be exacerbated already by hypotonia [82]. Additionally, excessively long footwear affects the position of the treadline of footwear relative to the metatarsophalangeal joints. This causes the metatarsophalangeal joints to dorsiflex in a region of footwear that is not designed for flexion, increasing the risk of developing foot pain [79, 80]. Foot pain is an established barrier to physical activity [62]. Considering the link between poorly-fitting footwear and reduced physical activity, it is plausible that improving footwear fit has the potential to have a positive effect on physical activity in children with Down syndrome. If improving footwear fit increases physical activity in children with Down syndrome, it may improve health-related quality of life in this population. There may also be other benefits, such as greater opportunities for social participation and interaction, improved self-esteem and happiness. These benefits may result in reduced burden on the healthcare system as a result of improved health, and reduced burden on families due to improved social participation and health.

One approach to resolve the issue of poorly-fitting footwear in children with Down syndrome is to determine their unique foot dimensions. No studies have thoroughly investigated the foot dimensions of both male and female children with Down syndrome and the implications on footwear fit. One study found that boys with Down syndrome have shorter, narrower feet when compared to their age and sex-matched typically developing peers [66]. However, this study only included boys with a very narrow age range (14 to 15 years), which limits the generalisability of the findings. Additionally, the study measured foot dimensions in 2D, so it is unknown how 3D measurements, such as volume and girth, vary. Therefore, there is limited information on the complex, 3D structure of the foot in children with Down syndrome. The 3D shape of the foot is important for correct footwear fit, so further studies are required to establish the 3D foot dimensions of children with Down syndrome. Doing so will provide valuable data that would assist footwear manufacturers when creating footwear for this population.

1.3 Physical activity

Physical activity is defined as any bodily movement produced by skeletal muscle that results in energy expenditure [83]. Physical activity is an important component of health promotion and disease prevention for all individuals. The effect of physical activity extends beyond weight management. It reduces the risk of health conditions that impact an individual's physical and mental health and wellbeing [84]. Physical activity can increase muscular and cardiovascular fitness, improve bone density, improve sleep patterns, and improves mental health [85]. Importantly, physical activity reduces the risk of developing non-communicable chronic health conditions such as cardiovascular disease, diabetes mellitus, obesity, hypertension, and skeletal and joint diseases [84, 86], all of which are more common in individuals with Down syndrome. Therefore, regular physical activity participation is a critical component of good health for children with Down syndrome.

For children aged between 5 to 17 years, physical activity guidelines recommend a minimum of 60 minutes of moderate to vigorous physical activity daily [87]. For children with disabilities, these physical activity guidelines are applicable and should be met where possible [87]. Consistent physical activity in childhood that continues into adulthood enables individuals to maintain a favourable risk profile for chronic health conditions. For example, consistent physical activity is an important factor in reducing rates of morbidity and mortality associated with cardiovascular and metabolic conditions [88, 89]. Therefore, encouraging regular physical activity participation of all children is crucial. Physical activity in children may be achieved through play or structured exercise. However, these opportunities may not be as readily available for children with Down syndrome due to barriers that interfere with their participation in physical activity. The following sections describe (i) methods used to measure physical activity and their relative advantages and disadvantages (Section 1.3.1), (ii) physical activity in children with Down syndrome (Section 1.3.2), and (iii) barriers and facilitators to physical activity in children with Down syndrome (Section 1.3.3).

1.3.1 Measurement of physical activity

Physical activity can be quantified using subjective or objective outcome measures. This includes self-reported estimations of physical activity (often referred to as subjective measures) and measurement devices that use known physical and/or time parameters (often referred to as objective measures), with each approach having its own advantages and limitations (Table 5) [90]. Physical activity is a complex health behaviour that can be described through several dimensions, such as frequency of activity, duration of activity,

intensity of activity, and type of activity performed. Activity can also be performed for different reasons, such as exercise (structured physical activity), activity related to work, school or leisure, or activity used as a mode of transport [87]. With this in mind, the various methods of measuring physical activity vary in their ability to measure each dimension and are unlikely to measure them all. While self-reported measures of physical activity (e.g. diary entry, surveys, checklists) are simple to use and provide relevant information, they are subject to recall bias which may affect the validity of the data collected [91]. Objective measures of physical activity (such as accelerometers) provide information on several dimensions that are descriptive of physical activity (e.g. duration, frequency, intensity of physical activity) and can overcome many of the limitations of subjective measures of physical activity. Therefore, objective measures of physical activity may provide a more valid measure of physical activity. As a result, accelerometers have rapidly become a ‘gold-standard’ for the objective measurement of physical activity [92]. The following section discusses the use of accelerometers in research as this type of device was used to assess physical activity in this thesis.

Table 5. Advantages and disadvantages of common methods to measure physical activity.

Method of measuring physical activity [90]	**Data type**	**Advantages**	**Disadvantages**
Self-reported data (i.e. diary entry)	Subjective	Provides accurate data on type, duration and is cost-effective.	Subject to under/overestimation. Potential recall bias. May be unreliable. Dependent on motivation and ability of individuals to report accurately and consistently. May encourage physical activity during time being studied.
Direct observation	Subjective	Provides information on activity, type, frequency and duration.	Cannot measure intensity or energy expenditure.
Accelerometry	Objective	Data on intensity, duration, frequency and stores data over time to allow for long term assessment.	Cost. Not suitable for all activities (i.e. water-based exercise or cycling).
Pedometry	Objective	Improved reliability. Easy to use. Output data easy to interpret.	No storage, therefore, data must be recorded by user. Only measures steps completed. Adherence.
Doubly labelled water	Objective	Can accurately assess large number of people. Estimates energy expenditure.	Subject to error and expensive. Does not provide information on type of physical activity.
Direct calorimetry	Objective	Accurate (less than 1% error). Used to validate other forms of physical activity measures.	Limited normal activity. Not practical.
Indirect calorimetry	Objective	More practical than direct calorimetry.	Limits typical movement.
Measurement of VO_2 peak	Objective	Best used in combination with other physical activity measures.	Not practical to measure maximum effort by most people. Not applicable for all activity (e.g. weightlifting).
Monitoring of heart rate	Objective	Provides data on energy expenditure. Can indicate time, which provides data on frequency, duration and rate of activity.	May be expensive. Removeable so may not detect all activity. Does not describe type of activity performed. Need self-reported data alongside use.

Accelerometers can collect objective data on duration, intensity and frequency of physical activity. Accelerometers record time-stamped acceleration signals that are converted into time spent in physical activity intensities [93]. Intensities are based on calibrated thresholds set by manufacturers derived from research data for specific populations or age groups. Data can be categorised according to time spent in various intensities (e.g. light, moderate, vigorous), which can help determine if physical activity guidelines are met.

One challenge with accurately measuring physical activity in children with Down syndrome relates to adherence with wearing accelerometers. On average, data collected over a minimum of four days out of seven non-consecutive days are needed to be valid [94]. One weekend day has also been recommended for valid data collection [95]. Adhering to wear time protocols can be challenging for people with Down syndrome [96-98] and others with intellectual disability [99-102]. Poor adherence to wear time protocols is problematic as missing physical activity data in studies that have used accelerometery may affect the validity of the results and limit the conclusions, as it is likely that data are not missing at random [103]. Methods to improve adherence to wear time have been suggested and involve placing the accelerometer beneath clothing, concomitantly using diaries to document accelerometer use, and wearing an accelerometer for 24 hours each day [104].

There are additional limitations to the use of accelerometers. They do not automatically classify type of activity (e.g. walking versus running); therefore, this data needs to be documented separately. They also do not measure all forms of activity, such as cycling or water-based activities (as they are not water-resistant). Another issue is the various options with data processing (i.e. setting different epoch lengths) that can affect the results, particularly with children who engage in high intensity bursts of activity throughout the day. The epoch length (interval over which acceleration signals are averaged) is the usual accelerometer-stored magnitude that is recorded at fixed intervals (i.e. 1 second, 5 seconds,15 seconds, 60 seconds or longer). The index of physical activity is calculated at the end of each epoch (or fixed interval). This process is repeated until data collection is complete [105, 106]. To address this issue, recent attempts have examined an appropriate epoch length to estimate sporadic, high intensity activity that some children may engage in [105, 107, 108]. It is suggested that a shorter time-sampling interval may reduce errors when classifying physical activity estimates [105]. There are also several cut-off points to classify intensities available, which can vary interpretation of data [109].

1.3.2 Physical activity in children with Down syndrome

Many children with Down syndrome do not perform enough physical activity to meet physical activity guidelines [110]. The percentage of children with Down syndrome who meet daily physical activity targets has been reported to range from 0 to 43%. Further, the level of intensity of physical activity is reduced in children with Down syndrome compared to their peers. Children with Down syndrome tend to engage in less intense activity and more sedentary and light activity [110]. Further, the amount and intensity of physical activity that children with Down syndrome engage in declines with age [111-113]. This is supported by the findings that younger children with Down syndrome tend to engage in higher intensity physical activity when compared to adolescents with Down syndrome [112, 114]. A longitudinal study showed this trend by following participants over time, where physical activity in earlier years was not maintained or improved, rather it declined with time [113]. This was seen to continue as those children with Down syndrome who did not maintain or improve physical activity continued to maintain low levels of physical activity over time. Physical activity patterns do not appear to be influenced by the day of the week in children with Down syndrome (i.e. weekday or weekend) [112]; however, sex-based differences in physical activity over the weekend have been reported [112]. Boys with Down syndrome engage in greater amounts of physical activity as well as at higher intensities when compared to girls with Down syndrome [113]. This finding may be explained by the greater likelihood for males to be involved with organised sporting activities compared to females [112].

Despite it being well established that many children with Down syndrome do not meet physical activity guidelines, it is important to acknowledge that many typically developing children also do not meet physical activity guidelines, with similar patterns regarding activity decline with age, and differences in physical activity between males and females [115]. However, the difference between these populations is that children with Down syndrome face additional challenges related to their disability (and external barriers such as access issues, time limitations, transport issues etc.) that are not experienced by typically developing children.

The most obvious detrimental effect of low physical activity among children with Down syndrome relates to the impact on physical health. Low physical activity is the primary cause of a large proportion of chronic diseases [116]. It increases the risk of obesity, which is common in children with Down syndrome. Obesity increases the risk of serious chronic health conditions (including osteoarthritis [117], type 2 diabetes, hypertension, cardiovascular diseases and cancer [118]) and further exacerbates these conditions. This is problematic for children with Down syndrome as they are already predisposed to such conditions as a result of the effects of Down syndrome on the body. Further, low

physical activity may have a negative impact on skill development. For example, children with Down syndrome often engage in physical activity through play, which is an opportunity for children with Down syndrome to practice their physical, social, communication and verbal skills. Play time is also a significant means for social interaction as it provides an opportunity for children to engage with their peers. When this form of physical activity is reduced, this can have consequential effects on the development of these skills. It is evident that regular physical activity for children with Down syndrome is essential for their health and holds significant consequences when not performed. Despite this, children with Down syndrome face unique challenges that contribute to low physical activity levels. These factors are discussed in Section 1.3.3.

1.3.3 Barriers and facilitators to physical activity in children with Down syndrome

The causes for reduced physical activity in children with Down syndrome are complex and multifactorial [119]. Broadly, contributing factors are associated with characteristics of Down syndrome, family factors and the availability of opportunities for physical activity for children with Down syndrome. However, some of these factors are modifiable and can be considered as facilitators that can improve physical activity engagement and allow children with Down syndrome to experience the benefits of physical activity.

Physical activity can be affected by the characteristics of Down syndrome. Cardiovascular conditions (e.g. congenital heart disease and altered physiology [reduced heart rate peak]) [120] affect physical activity through reduced energy and endurance. Musculoskeletal characteristics (i.e. hypotonia and ligamentous laxity [43, 82, 121]) make physical activity more challenging for children with Down syndrome, particularly in the presence of a musculoskeletal condition (e.g. hip dysplasia). Reduced gross motor skills in children with Down syndrome also affect physical activity participation. Differences in cognitive and physical skills between children with Down syndrome and their typically developing peers worsen with increasing age and act as a barrier to participation [119]. Additionally, the presence of an intellectual disability and the reduced communication ability of some children with Down syndrome may make it more challenging for them to participate in mainstream activities. However, certain personal characteristics of children with Down syndrome can facilitate physical activity. Children with Down syndrome who are strong verbal communicators, have good physical skills or those who have the cognitive ability to comprehend rules are more likely to be involved in mainstream physical activities [122]. Children who are determined and driven by success are likely to persist with physical activity, particularly when an activity involves competition.

Family members play a major role in influencing physical activity. Parental concern can limit children with Down syndrome from engaging in certain types of activity. In some instances, parents may be overprotective of their child and discourage them from participating if they feel they are too vulnerable in mainstream settings [119, 123]. There may be additional family restrictions that can limit physical activity such as other family responsibilities and time (e.g. parents' work commitments or balancing the needs of other family members [119]). This is particularly true when children with Down syndrome require one-on-one supervision due to safety concerns (i.e. the risk of injury) or behavioural issues. On the other hand, family members can facilitate physical activity by providing support and initiating opportunities. Parents who value sports and recognise its importance are more likely to involve their children in physical activity; either by sourcing opportunities for their child or creating them [119]. Siblings act as role models and encourage physical activity through creating interest or opportunities [119, 122]. Children with Down syndrome are also more likely to emulate behaviours observed by siblings, which can further facilitate opportunities of physical activity.

Finally, a major barrier to physical activity involves the lack of suitable programs for children with Down syndrome. Mainstream physical activity programs may not have the capacity to be adapted to account for the needs of children with Down syndrome [119]. Adapting existing mainstream programs to suit children with Down syndrome is difficult due to issues relating to limited staff, lack of resources (which may include financial constraints), time restrictions and lack of education. To address this, greater efforts are required to create programs that cater to the needs of children with Down syndrome. For example, team-based activities can be appropriate for children who are motivated by winning or enjoy teamwork or peer interaction. Alternatively, activities that are not team-based may suit other children with Down syndrome, who may require direct attention or guidance. A physical activity program should consist of activities that are enjoyed by children with Down syndrome, which reduces the likelihood of children requiring external encouragement from parents to participate.

Given that poorly-fitting footwear is associated with reduced physical activity (Section 1.2.3), improving footwear fit offers potential as an intervention that may improve physical activity. At present, the effectiveness of many interventions with potential to improve physical activity in children with Down syndrome is unknown, as few randomised trials (required to determine intervention effectiveness) exist [124-126]. More broadly, no systematic reviews have yet been published that have evaluated the effectiveness of

interventions to increase physical activity in individuals with intellectual disabilities. Accordingly, a broader synthesis of the literature to investigate the effectiveness of physical activity interventions in those with intellectual disabilities may provide insight as to what may be effective and applicable to children with Down syndrome.

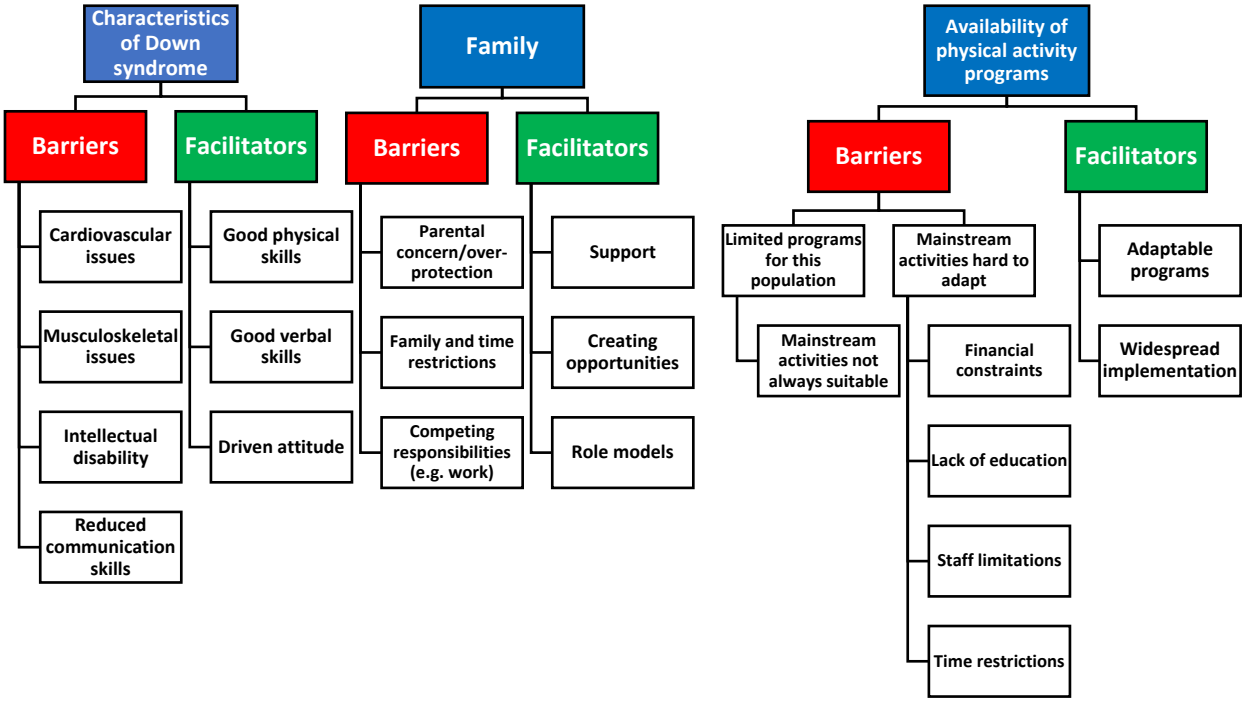


Figure 2. Barriers and facilitators to physical activity participation in children with Down syndrome.

1.4 Thesis outline

In the preceding sections, two linked problems were identified – these problems underpin the body of work presented in this thesis. The first is children with Down syndrome often do not participate in recommended levels of physical activity, and the second is they commonly wear poorly-fitting footwear. These problems also interact with each other; that is, wearing poorly-fitting footwear can make it more difficult for children with Down syndrome to be physically active. Therefore, improving footwear fit may have favourable effects on physical activity.

Children with Down syndrome also have unique foot dimensions that differ to their peers without Down syndrome, and this impacts footwear fit. Investigating the foot dimensions of children and Down syndrome is an important step to understanding how they differ to typically developed children, and how this affects footwear fit. Doing so will highlight the areas of fit that are most problematic to children with Down syndrome, which can then be used to design and manufacture more appropriate footwear for this population.

With this in mind, there are two primary aims to this thesis, which are stated below.

1. To determine the effectiveness of interventions (including custom-fitted footwear to improve fit) to increase physical activity in children with Down syndrome.
2. To determine the foot dimensions of children with Down syndrome, to better understand the issue of poorly-fitting footwear.

The following objectives address the primary aims of the thesis.

- Systematically synthesise the findings from studies that have evaluated the effectiveness of interventions to increase physical activity in individuals with intellectual disabilities.
- Determine the feasibility of conducting a definitive randomised trial to evaluate the efficacy of custom-fitted footwear for increasing physical activity in children with Down syndrome.
- Determine the reproducibility of measuring foot dimensions of children with Down syndrome using 3D-scanning.
- Compare the foot dimensions (obtained from 3D-scanning) of children with and without Down syndrome.

To address the objectives of this thesis, four related studies will be reported. These studies are presented across four chapters, as outlined below.

Chapter 2 presents a systematic review of the effectiveness of physical activity programs to increase physical activity in individuals with intellectual disabilities. The review

synthesised the findings of randomised controlled trials that have evaluated the effectiveness of physical activity interventions in individuals with intellectual disabilities. The findings from this review informed the study presented in Chapter 3.

Chapter 3 presents a randomised pilot study that aimed to determine the feasibility of conducting a definitive randomised trial to evaluate the efficacy of custom-fitted footwear for increasing physical activity in children with Down syndrome. Bowen's feasibility framework was used to guide this study. The results demonstrated that sizing of commercially available footwear may not accommodate the unique foot structure of children with Down syndrome. This limitation led to two further studies, presented in Chapters 4 and 5.

Chapter 4 presents a reproducibility study of measuring foot dimensions of children with Down syndrome using 3D foot scans. This study reported the intra- and inter-rater reproducibility of measuring foot dimensions that are used in the design of footwear. This study informed the measurements used in the study presented in Chapter 5.

Chapter 5 is a cross-sectional observational study that compared the differences in foot dimensions of a group of children with Down syndrome to an age and sex-matched group of typically developing children. This study reported the differences in foot dimensions and their implications for footwear fit.

Chapter 6 presents a discussion of the overall findings including the strengths and limitations of the research in this thesis and concludes with recommendations for future research in the field.

Chapter 2 – Systematic review

Preface

Children with Down syndrome are often less physically active than their typically developing peers and may not engage in adequate levels of physical activity. This places them at greater risk for many chronic health conditions. Therefore, increasing physical activity in this population is crucial. There is potential to modify recognised environmental barriers and/or enhance facilitators to physical activity to improve participation and health outcomes in this population.

Two previous systematic reviews reported physical activity interventions can have a positive effect on fitness (such as muscle strength), balance and psychological outcomes (such as improved self-confidence and self-esteem) in adults with intellectual disabilities [127, 128]. However, it is unclear whether physical activity interventions have a specific effect on physical activity. Accordingly, this chapter aims to address *Objective 1* of this thesis, which is to systematically synthesise the findings from randomised controlled trials that have evaluated the effectiveness of interventions to increase physical activity in individuals with intellectual disabilities. This study considers individuals with an intellectual disability originating from *any* condition, not specifically Down syndrome, as the body of literature involving children with Down syndrome was too small to perform a meaningful synthesis.

This chapter was published in the *Journal of Intellectual Disability Research* in 2019 with the associated online supplementary files presented in Appendix 1. The citations within this chapter relate to the reference list of the publication, not the reference list included at the end of this thesis.

2.1 Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials

The reference for this publication is:

Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. *Journal of Intellectual Disability Research;* 2019:63(2);168 – 191. DOI: https://doi.org/10.1111/jir.12562

This study was presented at two conferences:

Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. Australasian Academy of Cerebral Palsy and Developmental Medicine Conference. March 2018, Auckland, New Zealand.

Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. VIC State Podiatry Conference. April 2018, Melbourne, Australia.

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

Journal of Intellectual Disability Research doi: 10.1111/jir.12562

VOLUME 63 PART 2 pp 168–191 FEBRUARY 2019

Systematic Review

Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials

N. M. Hassan,[1,2] K. B. Landorf,[1,2,3] N. Shields[2,3,4] & S. E. Munteanu[1,2,3]

1 *Discipline of Podiatry, School of Allied Health, La Trobe University, Melbourne, Victoria, Australia*
2 *Living with Disability Research Centre, School of Allied Health, La Trobe University, Melbourne, Victoria, Australia*
3 *La Trobe Sport and Exercise Medicine Research Centre, School of Allied Health, La Trobe University, Melbourne, Victoria, Australia*
4 *Discipline of Physiotherapy, School of Allied Health, La Trobe University, Melbourne, Victoria, Australia*

Trial registration: This systematic review has been registered with PROSPERO, ID number CRD42016046948.

Abstract

Background People with intellectual disabilities (ID) often do not meet recommended guidelines for physical activity. The aim of this study was to systematically review available evidence that evaluated the effectiveness of interventions to increase physical activity in individuals with ID.

Method Five electronic databases (MEDLINE, CINAHL, EMBASE, SPORTDiscus and Cochrane Central Register of Controlled Trials) were searched from inception of the database to July 2017 to identify randomised controlled trials that evaluated the effectiveness of interventions to improve physical activity among people with ID. Trials were included if they measured at least one objective measure of physical activity. Quality appraisal was completed by two independent reviewers using the Cochrane Risk of Bias Tool. The magnitude of treatment effect was estimated for each intervention by calculating the standardised mean difference (SMD) and associated 95% confidence interval.

Results Nine randomised controlled trials (976 participants, 501 women, age range 9 months to 83 years) were included. Four trials evaluated unimodal interventions and five trials evaluated multimodal health promotion programmes based on using supportive environments to enable sustained behavioural changes in physical activity. None of the trials were rated as low risk of bias as all had at least one item on the Cochrane Risk of Bias Tool that was considered to be high risk. No trials were able to implement participant blinding. Three trials found statistically significant beneficial effects of interventions for increasing physical activity. Results showed that a 10-week progressive resistance training programme led to maintenance of physical activity levels at 24 weeks in adolescents with Down syndrome (SMD 0.78, 95% CI 0.17 to 1.40). Additionally, a 12- to 16-month multicomponent diet and physical activity programme produced improvement in physical activity at programme completion in adults with ID (reported effect size of 0.29). Finally, an 8-month physical activity and fitness programme

Correspondence: Ms. Nirmeen Hassan, Discipline of Podiatry, School of Allied Health, La Trobe University, Kingsbury Drive, Bundoora, Victoria, 3086, Australia (e-mail: n.hassan@latrobe.edu.au).

increased physical activity at 8 months in adults with ID (SMD 0.91, 95% CI 0.20 to 1.60). Findings regarding other interventions were inconclusive with small effects that were not statistically significant.
Conclusions There is inconsistent evidence of the effects of interventions for improving physical activity levels in individuals with ID. A progressive resistance training programme was found to maintain physical activity levels in adolescents with Down syndrome, while a multicomponent diet and physical activity programme and a physical activity and fitness programme were found to improve physical activity levels in adults with ID. Future trials using rigorous research designs are required to confirm these findings and establish whether other interventions designed to increase physical activity in people with ID are effective.

Keywords health promotion, intellectual disability, physical activity, randomised controlled trial, review, systematic

Introduction

Between 67% and 83% of children and adults with intellectual disabilities (ID) do not participate in the recommended amount of physical activity (Stanish *et al.* 2006). Physical activity is essential for cardiovascular fitness, bone health, psychological well-being and maintenance of a healthy body weight (Warburton *et al.* 2006). Recommended guidelines suggest children should partake in a minimum of 60 min of moderate to vigorous physical activity per day on most days of the week and adults between the ages of 18 to 64 years should perform 150 min of moderate to vigorous physical activity per week (Pitetti *et al.* 2009; World Health Organization 2010). However, people with ID often lead sedentary lifestyles (Robertson *et al.* 2000; Emerson 2005; Bartlo & Klein 2011; Tudor-Locke *et al.* 2011) and have significantly lower levels of physical activity levels compared with the general population (Robertson *et al.* 2000; Frey 2004; Emerson 2005; Stanish *et al.* 2006; Bartlo & Klein 2011). There are many personal, social and environmental barriers to physical activity for people with ID. For children with ID, reported barriers to physical activity include competing family responsibilities, parental overprotection, child factors (e.g. cognitive abilities, physical characteristics and behavioural problems), reduced physical or behavioural skills and lack of accessible physical activity programmes (Barr & Shields 2011; McGarty & Melville 2018). For adults with ID, barriers to physical activity include lack of support, not wanting to engage in physical activity, as well as medical and physiological factors (Mahy *et al.* 2010).

Low physical activity is associated with negative health outcomes and is the fourth leading risk factor for mortality worldwide (World Health Organization 2010). It increases the risk of non-communicable diseases, such as diabetes and cardiovascular disease, which account for approximately 50% of the overall global burden of disease (World Health Organization 2010). People with ID have poorer physical and mental health than the general population (Sutherland *et al.* 2002; Lennox *et al.* 2007). Further, this increased morbidity in people with ID increases the burden on caretakers and disability services (Krahn *et al.* 2006).

Regular participation in physical activity has the ability to reduce the burden of chronic disease by improving and maintaining physical and mental health and is therefore considered an important intervention for people with ID (Durstine *et al.* 2013). Two previous systematic reviews reported physical activity interventions can improve fitness (e.g. muscle strength), balance and psychological outcomes, such as enhanced self-confidence, in adults with ID (Bartlo & Klein 2011; Heller *et al.* 2011). However, these reviews did not evaluate whether the interventions were effective at increasing physical activity levels. More recently, a systematic review investigated the efficacy of interventions to increase physical activity in children and adolescents; however, adults were not included (McGarty *et al.* 2018).

Therefore, the aim of this study was to systematically review available evidence that has evaluated the effectiveness of interventions to increase physical activity in individuals with ID.

Method

The protocol for this systematic review was prospectively registered in PROSPERO (ID: CRD42016046948). This systematic review is reported using the PRISMA guidelines (Moher *et al.* 2009).

Eligibility criteria

Types of studies

Only randomised controlled trials were included as this is a rigorous research design. There were no restrictions on the language of trials, publication status or year of publication. However, only trials that were published in peer-reviewed journals were included. Grey literature, such as conference abstracts and theses, were not included.

Types of participants

We included trials that recruited participants of all ages who were described as having an ID that originated prior to the age of 18 years.

Types of interventions

Interventions were included if their primary or secondary aim was to increase physical activity in individuals with ID. Interventions could be unimodal or multimodal. For this review, *unimodal* interventions were single interventions that had no health promotion component, and *multimodal* interventions were programmes that combined education, health promotion and physical activity. Interventions included but were not limited to (1) structured physical activity (exercise), cycling or walking programmes and (2) behavioural change programmes that included education and nutrition components in addition to physical activity.

Types of outcome measures

The primary outcome measure of this review was physical activity, therefore included trials needed to have at least one objective measure of physical activity. We limited trials to those that measured physical activity using objective measures such as direct observation, accelerometers, pedometers and doubly labelled water. Trials that used subjective measures (such as questionnaires) to measure physical activity were not included as such measures have questionable validity (Matthews *et al.* 2011). Objective measures of physical activity are considered more valid than subjective measures (Hinckson & Curtis 2013; Sylvia *et al.* 2014). Physical activity was considered as any bodily movement that resulted in energy expenditure, which covers all forms of activity (Caspersen *et al.* 1985). We did not limit the duration of follow-up.

Search strategy

Electronic searches

Five electronic databases (MEDLINE (1946-), CINAHL, EMBASE, SPORTDiscus and Cochrane Central Register of Controlled Trials) were searched from inception of the database to July 2017. A search strategy adapted for each database was developed based on two key concepts: *intellectual disability* and *physical activity* (Data S1). Relevant MeSH and free text terms were incorporated into the search strategy; for example, mentally disabled persons for ID and exercise for physical activity. Terms were combined with appropriate Boolean operators. Valid search filters were used (Data S2) to identify randomised controlled trials (Wong *et al.* 2006; Lefebvre *et al.* 2011; SIGN 2017). Citation tracking of eligible trials was performed using Scopus, and reference lists of the identified trials were checked for potentially eligible trials that database searches may have missed.

Data extraction and analysis

Selection of studies

Retrieved searches were exported into Endnote version X7.4 (Thomson Reuters, New York, USA) and duplicates removed. Two reviewers (N. M. H. and S. E. M.) independently screened titles and abstracts of the articles retrieved (n = 3188) and excluded those that did not meet the eligibility criteria (n = 863) (Fig. 1). Where it was unclear from the title and abstract if an article should be included, the full text was retrieved and the eligibility criteria reapplied. Disagreements were resolved by consensus. There were three studies (Neman *et al.* 1975; Shields *et al.* 2008; Schijndel-Speet *et al.* 2012) that were excluded from being included in the review following a consensus discussion.

Data management

We used a data extraction form to obtain data on study characteristics (sample size, description of the interventions and participant characteristics) and outcome measures (Tables 1–3). Two reviewers (N. M. H. and S. E. M.) extracted data from the trials

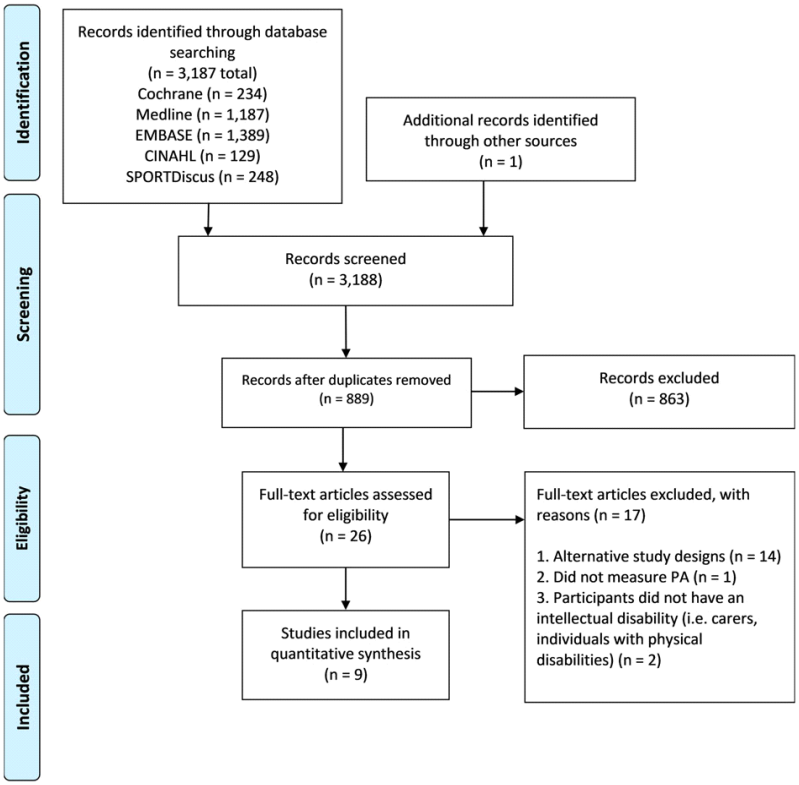


Figure 1 Study selection process (PRISMA). [Colour figure can be viewed at wileyonlinelibrary.com]

independently. Where possible, means and standard deviations were extracted for physical activity outcome measures at each reported time point. In cases where data were missing, the trial authors were contacted via email for further information.

Assessment of risk of bias in included studies

The internal validity of included studies was evaluated using the Cochrane Collaboration Risk of Bias Tool (Higgins *et al.* 2011). This tool evaluates the risk of selection bias, performance bias, detection bias, attrition bias and other potential biases in randomised controlled trials. The criteria considers the method of random sequence generation, allocation concealment, blinding of participants and personnel, blinding of outcome assessment, incomplete outcome data and selective reporting. Review Manager 5.3 (Review Manager 2014) was used to summarise the risk of bias for each trial. Each criterion was assigned a high, low or unclear risk rating, in which 'yes' indicates a low risk, 'no' indicates a high risk and 'unclear' indicates insufficient information was provided to enable a judgement of risk of bias (Higgins *et al.* 2011). A trial was considered to be at high risk of bias if at least one of the criterion was rated as high risk. In contrast, a trial was considered to be at low risk of bias if all criteria were rated low risk. Any trials not meeting these criteria were rated unclear (Higgins *et al.* 2011; Whittaker *et al.* 2017). However, importantly, because of the nature of the interventions used, it is very difficult to implement participant blinding, and thus, trial findings were interpreted in the context of this issue. Two reviewers (N. M. H. and K. B. L.) assessed the risk of bias independently, and any

Table 1 Study characteristics

Trial	Study design	Setting/country	Inclusion/exclusion criteria	Groups	Sample size at baseline	Participant characteristics	
Angulo-Barroso *et al.* (2008)	RCT	Community setting, Michigan	Inclusion Down syndrome (age not specified). Ability to take a minimum of six spontaneous steps on the treadmill in any given minute of a 5-min testing session. Exclusion NR.	Intervention Treadmill protocol (high-intensity). Control Treadmill protocol (low-intensity).	IG: 16 CG: 14	No. of men (women) Mean age (SD) in months‡ Mean BMI (SD)	IG: 12 (4) CG: 6 (8) IG: 9.7 (4.1) CG: 10.4 (2.2) IG: NR CG: NR
Bergstrom *et al.* (2013)	Cluster RCT	Community residence, Sweden	Inclusion Adults with mild to moderate ID. If 3 other residents chose to participate. Ability to understand basic English. Able to provide consent. Exclusion Require use of ambulatory device.	Intervention Multicomponent diet and PA programme. Control Wait-list control.	IG: 64 CG: 66	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 27 (37) CG: 29 (37) IG: 36.2 (10.1) CG: 39.4 (11.3) IG: 30 (7.6) CG: 28.5 (6.6)
Curtin *et al.* (2013)	Pilot RCT	Community setting, USA	Inclusion Aged 13 to 26 years. BMI $>$ 85th percentile. IQ 45 to70. Written physician approval. Parent willing to attend session. Exclusion Use of appetite altering medications. Chronic gastrointestinal illness. Untreated thyroid issues. Uncontrolled seizure disorders or orthopaedic cardiac conditions limiting PA.	Intervention Nutrition and physical activity programme. Control Nutrition and activity education and behavioural intervention.	IG: 10 CG: 11	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 1 (9) CG: 3 (8) IG: 20.5 (4.1) CG: 20.5 (2.4) IG: 36.5 (6.9) CG: 35.8 (5.4)

Table 1. *(Continued)*

Trial	Study design	Setting/country	Inclusion/exclusion criteria	Groups	Sample size at baseline	Participant characteristics	
McDermott *et al.* (2012)[†]	RCT	Local disability agency service facility, North and South Carolina	Inclusion Aged 18 to 65 years. Mild to moderate ID. Ambulatory and can communicate verbally. Able to give informed consent. Exclusion Underweight. Serious medical conditions. Medical conditions affecting ability to participate in PA.	Intervention Steps To Your Health Program. Control Hygiene and safety classes.	IG: 216 CG: 216	No. of men (women) Mean age (SD) in years Mean BMI (SD)	No group data presented for both groups IG: 38.8 (NR) CG: 38.8 (NR) Mean not reported for both groups
Melville *et al.* (2015)	Cluster RCT	Community based setting, Scotland	Inclusion >18 years and any level of ID. Exclusion Severe challenging behaviour. Requires constant 1:1 support from carers. Significant mobility issues.	Intervention Walking programme 'Walk Well'. Control Wait-list control.	IG: 54 CG: 48	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 29 (25) CG: 28 (20) IG: 44.9 (13.5) CG: 47.7 (12.3) Mean not reported for both groups
Shields *et al.* (2013)	RCT	Community setting, Melbourne, Australia	Inclusion Aged 14 to 22 years with Down syndrome. Mild to moderate ID according to parental report. Can follow simple instructions in English. Fit and well to follow a high intensity PA programme. Exclusion Partaken in a progressive resistance training programme 3 months prior.	Intervention Student-led progressive resistance training programme. Control Social programme.	IG: 34 CG: 34	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 19 (15) CG: 19 (15) IG: 17.7 (2.4) CG: 18.2 (2.8) IG: 27.3 (3.8) CG: 27.2 (5.6)

Table 1. *(Continued)*

Trial	Study design	Setting/country	Inclusion/exclusion criteria	Groups	Sample size at baseline	Participant characteristics	
			Concurrent medical condition in addition to Down syndrome. History of violent outburst, absconding, antisocial behaviour or aggressive behaviour.				
Shields and Taylor (2015)	Phase II RCT	Community setting, Melbourne, Australia	Inclusion Aged 18 to 35 years. Mild to moderate ID according to parental report. Can follow simple English instructions. And able to engage in PA. Exclusion Previous completion of a physical activity programme 3 months prior to the trial. Acute or concurrent medical condition that would render them as unfit to participate in a PA programme. Significant behavioural issues that may affect their ability to complete the programme.	Intervention Structured walking programme 'Walkabout'. Control Social programme.	IG: 8 CG: 8	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 3 (5) CG: 5 (3) IG: 21.6 (3.4) CG: 21.2 (3.2) IG: 95.6 (17.2) CG: 89.3 (8.8)
Ulrich *et al.* (2011)	RCT	Community setting, Michigan, Ohio	Inclusion Aged 8 to 15 years. Down syndrome. Exclusion Medical condition limiting PA or severe behavioural issues during screening.	Intervention Modified bicycle. Control Wait-list control.	IG: 19 CG: 27	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 9 (10) CG: 11 (16) IG: 12.4 (2.2) CG: 12.0 (1.9) IG: 24.3 (NR) CG: 23.0 (NR)
van Schijndel-Speet *et al.* (2017)	Cluster RCT	DACs, Netherlands	Inclusion >40 years of age. Mild to moderate ID. Exclusion	Intervention PA programme and education programme. Control	IG: 66 CG: 65	No. of men (women) Mean age (SD) in years Mean BMI (SD)	IG: 28 (38) CG: 31 (34) IG: 58.2 (NR) CG: 57.9 (NR) IG: 27.9 (NR)

Table 1. *(Continued)*

Trial	Study design	Setting/country	Inclusion/exclusion criteria	Groups	Sample size at baseline	Participant characteristics
			Use of an ambulatory device. Medical condition limiting PA (e.g. dementia). DACs with <15 older adults with mild to moderate ID and DACs with organisational issues.	Care as usual.		[*n* = 59] CG: 27.5 (NR) [*n* = 58]

BMI, body mass index; CG, control group; DACs, day activity centres; ID, intellectual disability; IG, intervention group; NR, not reported; PA, physical activity; RCT, randomised controlled trial.
†McDermott *et al.* (2012) did not provide group data, instead they provided a percentage of each characteristic.
‡Participants were infants in this trial.

Table 2 Description of interventions

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
Angulo-Barroso *et al.* (2008)	Unimodal	High-intensity interval training programme was compared with a low-intensity interval training programme using a custom-built treadmill.	9 months	5 days per week	5 min	Parents.	Participants' home.	The high-intensity protocol involved progressive increases to treadmill belt speed, time and ankle weights. The low-intensity protocol did not change during the longitudinal interval of the intervention.	Not reported.
Bergstrom *et al.* (2013)	Multimodal	A three component multic omponent diet and PA	12 to 16 months	(1) Six network meetings	(1) 3 h			The components of the intervention were	Median score was 65 out of a

Table 2. *(Continued)*

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
		programme targeting both carers and participants. The three components were (1) Appointment for a health ambassador in each community residence attending network meetings. Role was to provide health information to colleagues and organise health-promoting activities for residents; (2) A study circle (Focus Health) for caregivers to increase knowledge, discuss and plan health-promotion work in residence; (3) A health course for residents which composed of a course manual that was led by a course leader.		(2) Ten sessions (3) Ten sessions to be fitted into the routines of residents	(2) 90 min for the study circle for care givers (3) Duration to reported for health course for residents	Health ambassadors and care givers.	Common rooms in each residence.	developed in cooperation with managers, caregivers and the Swedish National Association for Persons with intellectual disability to achieve appropriateness for a real-life setting. The intervention allowed some tailoring to suit the needs of the individuals.	possible score of 90. Those in the higher fidelity group saw higher improvements in results.
Curtin *et al.* (2013)	Multimodal	Nutrition and physical activity programme with two active intervention groups. One group received a 6-month nutrition and activity education intervention, and the other group received a 6-month nutrition and activity education plus a behavioural intervention.	9 months	Ten weekly sessions in first 3 months followed 10, weekly sessions for 3 months with tapered intensity (4 biweekly sessions, 2 sessions every 3rd week). Total of 16 sessions	90 min	Parents to implement the intervention at home, while a dietician and therapeutic recreation specialist delivered the nutrition and physical activity sessions.	Participants' home. Session location not specified.	The education programme allocated to one group was a nutrition and activity education programme that taught participants simple nutritional concepts and exercises via verbal instruction, demonstration taste tests and activities. The other group received same nutrition and activity	The overall session attendance rates were reported at 93%. Fidelity checks revealed 100% adherence to the intervention protocol with the new treatment team at the end of the trial.

Table 2. *(Continued)*

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
McDermott *et al.* (2012)	Multimodal	8 participatory classes referred to as 'Steps To Your Health Program' that covered a variety of topics that encouraged moderate to vigorous physical activity and body mass reduction.	3 months	One session per week	90 min	Health educator.	Local room provided by the disability service agency in a local area to where participants resided.	education programme, plus the addition of a behavioural intervention that involved sessions with a behavioural specialist who provided instructions on behavioural strategies (i.e. monitoring diet and activity). Exercises were modified if a participant was unable to perform them. Each participant received a tailored diet plan from a registered dietician. Classes covered topics including nutrition, exercise, thinking patterns and behaviour management. The control group covered topics relating to hygiene and safety. The health educator ensured teaching style suited participants.	Not reported.

Table 2. *(Continued)*

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
Melville *et al.* (2015)	Multimodal	Walking programme referred to as 'Walk Well'. The intervention involved physical activity consultations to implement a behaviour change model (encompassing goal-setting, self-efficacy, self-monitoring and mobilising social support for change) and involved carers.	3 months	5 days per week	30 min	Carers.	Day centres.	The walking intervention was simplified as much as possible and adapted to account for the differences in communication and cognitive abilities of the participants. Apart from the structured walking programme, there were three face to face meetings between participants, carers and a walking advisor, where educational resources were provided, progress reviewed towards set goals and the final meeting focused on encouraging participants to maintain changes by reviewing goal attainment and perceived benefits of the programme.	Not reported.
Shields *et al.* (2013)	Unimodal	Student-led progressive resistance training programme (using readily available resistance machines). It involved pairing a physiotherapy student as a mentor with a	10 weeks	Twice per week	45 to 60 min	Physiotherapy student as a mentor.	Local gymnasium.	There were 7 exercises in total, 3 upper body exercises, 3 lower body exercises and 1 trunk exercise. The	Participants attended 92% of their sessions and data obtained in log books suggests

Table 2. *(Continued)*

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
		participant to assist them complete the training programme and provide support. The physiotherapy student also completed the exercises with the participant.						programme prescribed 3 sets of 12 repetitions (or until muscle fatigue). The sessions were documented regarding exercises completed, weights lifted and number of repetitions and sets completed. The intensity progressed once participants were able to perform all sets and repetitions prescribed.	high fidelity to the intervention.
Shields and Taylor (2015)	Unimodal	Structured walking programme referred to as 'Walkabout'. Participants in the intervention group worked with their student mentor to complete walking sessions together and to plan for an additional session of walking without the student mentor while participants in the control group engaged in social activities with their student mentors. Mentors were contacted every fortnight by a member of the research team to ensure that the programme was going to plan and participants were adhering	2 months	Once per week	Two 45-min mentored sessions and 60 min individually tailored	Student mentor.	Local community.	Student mentors were physiotherapy students who attended a 2-h training session prior to the commencement of the trial. The training session covered information about the intervention, Down syndrome, strategies when interacting with young adolescents with Down syndrome and motivational and teaching strategies.	Participants attended 96% of the scheduled sessions.

Table 2. *(Continued)*

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
		to the 90 min of supported walking.							
Ulrich *et al.* (2011)	Unimodal	Participants were taught how to ride a bicycle (intervention). The rollers tapered as the rider progressed in skill, which eventually led to riding a standard two-wheel bicycle.	1 week	5 days a week	75 min per day	Trained staff from the 'Lose the Training Wheels Organisation'.	Local community setting.	The bicycle used was a specially designed, adapted bicycle that provided stability while learning. The bicycle was designed to allow for incremental progress to a two-wheel bicycle and could be altered to suit the needs of the rider. Special rollers could be fitted in place of the rear wheel to facilitate movement that was similar to a two-wheel bicycle. The rollers included a series of eight different sizes that increased in difficulty level.	Not reported.
van Schijndel-Speet *et al.* (2017)	Multimodal	PA programme and education programme With two components: 1. Education programme (to improve participants' knowledge on physical activity and its health benefits). 2. Physical activity programme (based on guidelines set by the American College of Sports	8 months	PA programme: 3 days per week Education programme: 2 days per week	PA programme: 15 to 20 min- increased to 45 min Education program: 44-min pre/post PA	Physical activity instructors and day activity centre staff.	Day activity centres.	The education component was inspired by another health promotion programme (known as 'Health Matters'), which is developed with experts in educating people with intellectual disabilities and	Participants took part in 78% of sessions.

Table 2. *(Continued)*

Trial	Intervention type	Intervention	Length of programme	Frequency	Duration	Intervention provider	Location	Tailoring/specifics	Intervention fidelity
		Medicine and the American Heart Association.						developing appropriate educational content.). The physical activity programme was set to address fitness components, including strength, endurance, balance and flexibility.	

Table 3 Physical activity outcomes

Trial	PA outcome measure	Measure of PA	Time points	Sample size at baseline	PA results T1Mean (SD)		PA results T2 Mean (SD)		PA results T3 Mean (SD)	
Angulo-Barroso *et al.* (2008)	Magnitude of leg activity (average counts/15 s)	ActiWatch	T1: End of intervention phase (approximately 9 months) T2: 3 months post walking onset T3: 6 months post walking onset	IG: 16 CG: 14	IG (*n* = 16) 171 (20.0)	CG (*n* = 14) 160 (18.7)	IG (*n* = 16) NR	CG (*n* = 14) NR	IG (*n* = 16) NR	CG (*n* = 14) NR
Bergstrom *et al.* (2013)	Steps/day	Keep Walking LS2000	T1: 12 to 16 months post baseline date	IG: 64 CG: 66	IG (*n* = 32) NR	CG (*n* = 37) NR				
Curtin *et al.* (2013)	MVPA min/day	Actical accelerometer	T1: 10 weeks T2: 6 monthsT3: 12 months	IG: 10 CG: 11	IG (*n* = 11) 9.8 (17.6)	CG (*n* = 8) −13.3 (19.0)	IG (*n* = 11) 17.6 (19.2)	CG (*n* = 8) −7.1 (21.2)	IG (*n* = 11) 9.3 (19.6)	CG (*n* = 8) −6.3 (21.5)
				IG: 216	IG (*n* = NR)	CG (*n* = NR)	IG (*n* = NR)	CG (*n* = NR)	IG (*n* = 61)	CG (*n* = 57)

Table 3. *(Continued)*

Trial	PA outcome measure	Measure of PA	Time points	Sample size at baseline	PA results T1 Mean (SD)		PA results T2 Mean (SD)		PA results T3 Mean (SD)	
McDermott *et al.* (2012)	MVPA min/min of wear time	ActiGraph accelerometer	T1: week 9T2: 6 monthsT3: 12 months	CG: 216	NR	NR	NR	NR	0.023 (0.025)	0.020 (0.019)
Melville *et al.* (2015)	% time per day MVPA	ActiGraph GT3X	T1: 12 weeksT2: 24 weeks	IG: 54 CG: 48	IG (n = 42) 3.0 (2.6)	CG (n = 40) 3.1 (2.1)	IG (n = NR) NR	CG (n = NR) NR		
Shields *et al.* (2013)	Average vector magnitude activity per minute	RT3 activity monitor	T1: week 11T2: week 24	IG: 34 CG: 34	IG (n = 22) 326 (87)	CG (n = 22) 304 (123)	IG (n = 22) 337 (86)	CG (n = 22) 265 (95)		
Shields and Taylor (2015)	Average vector magnitude activity per minute	RT3 activity monitor	T1: week 9	IG: 8 CG: 8	IG (n = 7) 344.2 (158.8)	CG (n = 5) 291.0 (99.4)				
Ulrich *et al.* (2011)	Time spent in MVPA	Actical accelerometer	T1: 8 weeksT2: 52 weeks post baseline	IG: 36 CG: 36	IG (n = 19) 36.5 (25.8)	CG (n = 17) 34.8 (18.0)	IG (n = 19) 48.7 (26.2)	CG (n = 17) 39.7 (23.8)		
van Schijndel-Speet *et al.* (2017)	Steps per day	NL-1000 (higher walking speed) StepWatch (lower walking speed)	T1: 4 months post-intervention T2: 8 months post-intervention T3: 6 months after trial	IG: 66 CG: 65	IG (n = NR) NR	CG (n = NR) NR	IG (n = 14) 836 (2386)[§]	CG (n = 24) −368 (1135)[§]	IG (n = 16) 78 (1924)[§]	CG (n = 19) −859 (1689)[§]

CG, control group; IG, intervention group; MVPA, moderate to vigorous physical activity; NR, not reported; PA, physical activity.
[§]PA results reported as mean difference from baseline.

disagreements were resolved by consensus. There was excellent agreement between reviewers for the ratings, with agreement ranging from 78% to 100% across items.

Data synthesis

Data were synthesised and analysed using Review Manager 5.3 (Review Manager 2014). The magnitude of effect of each intervention was estimated by calculating the standardised mean difference (SMD) and associated 95% confidence interval. SMDs were used to allow comparison of results across studies because the unit of measurement of physical activity varied. Where the SMD was significant (i.e. its 95% confidence intervals did not include zero), the size of the effect was interpreted as follows: a small effect if the SMD was approximately 0.2, a moderate effect if approximately 0.5 and a large effect if approximately 0.8 or greater (Faraone 2008). Data from the included trials were not pooled for meta-analysis because of large variability in interventions and time points across trials (i.e. clinical heterogeneity).

Results

Description of studies

The search strategy yielded 3188 records for assessment of eligibility (Data S1). The yield was reduced to 26 articles that were reviewed in full text. After applying the eligibility criteria, nine trials were included. Reasons for exclusion included non-randomised trials, implementing the intervention on individuals without ID (i.e. carers only) and not including a measure of physical activity (Fig. 1). One author was contacted to determine whether their study protocol (van Schijndel-Speet *et al.* 2013) had led to any publications, and the trial was subsequently added to the review (van Schijndel-Speet *et al.* 2017). Three authors (McDermott *et al.* 2012; Bergstrom *et al.* 2013; Melville *et al.* 2015) were contacted to request missing data but we were unable to obtain this information.

Tables 1 and 2 present a summary of the study characteristics and interventions of the nine trials included in the review. Three trials (Bergstrom *et al.* 2013; Melville *et al.* 2015; van Schijndel-Speet *et al.* 2017) were cluster randomised controlled trials, and six (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; McDermott *et al.* 2012; Shields *et al.* 2013) were randomised controlled trials, of which two (Curtin *et al.* 2013; Shields & Taylor 2015) were pilot randomised controlled trials.

The age of participants varied across trials from 9 months to 83 years. Five trials included participants who were young children and adolescents (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; Curtin *et al.* 2013; Shields *et al.* 2013; Shields & Taylor 2015), while the remaining four trials included adult participants (McDermott *et al.* 2012; Bergstrom *et al.* 2013; Melville *et al.* 2015; van Schijndel-Speet *et al.* 2017). The number of men and women in each group was relatively balanced, except for one trial (Curtin *et al.* 2013) that included very few men. Sample sizes ranged from 16 to 432 participants. The severity of ID of the participants was recorded as mild to moderate in five trials (McDermott *et al.* 2012; Bergstrom *et al.* 2013; Curtin *et al.* 2013; Shields *et al.* 2013; Shields & Taylor 2015; van Schijndel-Speet *et al.* 2017), and three trials (Ulrich *et al.* 2011; Curtin *et al.* 2013; Melville *et al.* 2015) included participants with any level of ID. Two trials (Shields *et al.* 2013; Shields & Taylor 2015) relied on parental report to determine the level of ID, and the remaining trials (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; McDermott *et al.* 2012; Bergstrom *et al.* 2013; Curtin *et al.* 2013; Melville *et al.* 2015; van Schijndel-Speet *et al.* 2017) did not describe how the level of ID was measured. Participants resided in group homes, homes with supported living or with family (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; Curtin *et al.* 2013; Shields *et al.* 2013; Shields & Taylor 2015).

Physical activity interventions were broadly categorised across trials as unimodal or multimodal interventions. Four trials (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; Shields *et al.* 2013; Shields & Taylor 2015) used unimodal interventions including progressive resistance training, cycling, treadmill training and walking programmes (Tables 1,2). Five trials (McDermott *et al.* 2012; Bergstrom *et al.* 2013; Curtin *et al.* 2013; Melville *et al.* 2015; van Schijndel-Speet *et al.* 2017) used multimodal interventions that included a physical activity programme, education and nutrition advice (Tables 1,2). Interventions were all implemented in a community setting, with one exception, which was delivered to participants living at a day activity centre (van Schijndel-Speet *et al.* 2017). The frequency and duration of each

intervention varied in each trial. The frequency ranged from one weekly session (McDermott *et al.* 2012; Curtin *et al.* 2013; Shields & Taylor 2015) to five sessions per week (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2008; Melville *et al.* 2015). The duration of each session ranged from 5 min (Angulo-Barroso *et al.* 2008) to 90 min (McDermott *et al.* 2012; Curtin *et al.* 2013). The duration of the interventions varied from 1 week (Ulrich *et al.* 2011) to 16 months (Bergstrom *et al.* 2013), with a median duration of 3 months. Where required, interventions were adapted to suit the abilities of the participants by simplifying the task or modifying equipment required for the intervention. Multimodal interventions involved other professionals (i.e. dietitian, health ambassador or individuals with experience in teaching individuals with ID) to deliver the educational components to participants. When a trial included children, their parents were involved in applying the intervention, and in the other remaining trials with adult participants, carers or staff members of the place of residence were also involved. For adolescents, two trials used student mentors to support the participants to exercise (Shields *et al.* 2013; Shields & Taylor 2015). Four interventions were based on a theoretical framework (i.e. social cognitive theory) that simplified the intervention to account for differences in communication and cognitive levels and to improve its applicability in a real-life setting (McDermott *et al.* 2012; Bergstrom *et al.* 2013; Melville *et al.* 2015; van Schijndel-Speet *et al.* 2017).

Control groups were either wait-listed (Ulrich *et al.* 2011; Bergstrom *et al.* 2013; Melville *et al.* 2015), usual care (van Schijndel-Speet *et al.* 2017) or inert interventions that were not expected to have an effect on physical activity (e.g. social activities not involving physical activities) (McDermott *et al.* 2012; Shields *et al.* 2013; Shields & Taylor 2015). In two trials (Angulo-Barroso *et al.* 2008; Curtin *et al.* 2013), both groups received active interventions. In the first trial by Curtin *et al.* (2013), one group received nutrition and physical activity education, and the other received a behavioural intervention in addition to nutrition and physical activity education. In the second trial by Angulo-Barroso *et al.* (2008), two treadmill training protocols were compared (one group received high-intensity training while the other group received low-intensity training). All included trials measured physical activity as the primary outcome measure except for one trial (Shields *et al.* 2013) where physical activity was a secondary outcome. All trials measured physical activity using accelerometers or pedometers (Table 3). The definition of adherence to wearing the physical activity monitor varied across trials (Data S3). Eight trials (Ulrich *et al.* 2008; McDermott *et al.* 2012; Bergstrom *et al.* 2013; Curtin *et al.* 2013; Shields *et al.* 2013; Melville *et al.* 2015; Shields & Taylor 2015; van Schijndel-Speet *et al.* 2017) reported participants wore the activity monitor for at least 6 h a day for 3 days (Penpraze *et al.* 2006; Temple & Stanish 2009). Only one trial (Angulo-Barroso *et al.* 2008) did not specify how adherence to wearing an activity monitor was defined.

Risk of bias assessment

Risk of bias is displayed in Figure 2. None of the trials were rated as low risk of bias as all had at least one

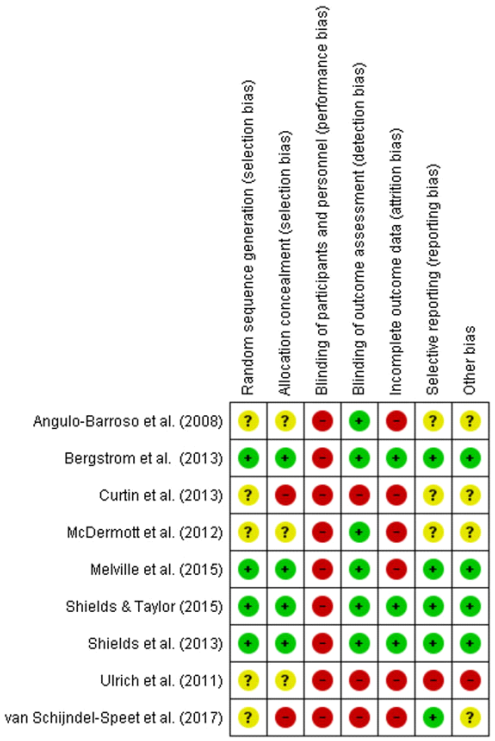


Figure 2 Risk of bias summary: review authors' judgements about each risk of bias item for each included trial. [Colour figure can be viewed at wileyonlinelibrary.com]

item on the Cochrane Risk of Bias Tool that was considered to be high risk. No trials were able to implement participant blinding. Two trials (Curtin *et al.* 2013; van Schijndel-Speet *et al.* 2017) were rated as high risk because of inappropriate methods of allocation concealment, three trials (Ulrich *et al.* 2011; Curtin *et al.* 2013; van Schijndel-Speet *et al.* 2017) did not include blinded outcome assessment and six trials (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; McDermott *et al.* 2012; Curtin *et al.* 2013; Melville *et al.* 2015; van Schijndel-Speet *et al.* 2017) had significant amounts of incomplete outcome data because of poor adherence in the use of equipment that measured physical activity and high rates of participant attrition.

Effects of interventions

Unimodal interventions

Four trials (Angulo-Barroso *et al.* 2008; Ulrich *et al.* 2011; Shields *et al.* 2013; Shields & Taylor 2015) evaluated the effectiveness of unimodal interventions (Tables 3,4). One trial (Ulrich *et al.* 2011) compared a 1-week cycle training intervention with a wait-list control group in 46 participants. There was no difference between groups for physical activity at 12 months (SMD 0.35, 95% CI −0.31 to 1.01).

In the second trial (Shields *et al.* 2013), a 10-week progressive resistance training programme was compared with a social programme of recreational activities not related to physical activity (e.g. crafts, baking or watching movies) in 68 participants. A large significant difference in physical activity in favour of the progressive resistance training programme was found at 24 weeks (SMD 0.78, 95% CI 0.17 to 1.40), but not immediately after the programme at 11 weeks (SMD 0.20, 95% CI −0.39 to 0.80).

The third trial (Shields & Taylor 2015) compared an 8-week walking programme with a social programme in 16 participants using student mentors in both groups. No difference was found in physical activity between the groups at 9 weeks (SMD 0.36, 95% CI −0.81 to 1.52).

The final trial (Angulo-Barroso *et al.* 2008) compared a 9-month high-intensity treadmill protocol with a low-intensity treadmill protocol in 30 infant participants. The intervention ended for each participant when they were able to independently walk three steps (walking onset). There was no difference in physical activity levels between groups during the intervention phase (SMD 0.55, 95% CI −0.18 to 1.28).

Multimodal interventions

Five trials examined the effectiveness of multimodal interventions (Tables 3,4). One trial (Melville *et al.* 2007) compared a 12-week multimodal intervention of a physical activity programme and behavioural

Table 4 Effects of the interventions on physical activity

Trial	Intervention	Time point	SMD (95% CI)
Angulo-Barroso *et al.* (2008)	Treadmill protocol	9 months	0.55 (−0.18 to 1.28)
Bergstrom *et al.* (2013)	Multicomponent diet and PA programme	12 to 16 months	0.29[†]
Curtin *et al.* (2013)	Nutrition and physical activity programme	12 months	0.73 (−0.22 to 1.68)
McDermott *et al.* (2012)	'Steps to your Health' programme	12 months	0.14 (−0.23 to 0.50)
Melville *et al.* (2015)	Walking programme 'Walk Well'	3 months	−0.04 (−0.47 to 0.39)
Shields *et al.* (2013)	Progressive resistance training programme	11 weeks	0.20 (−0.39 to 0.80)
		24 weeks	0.78 (0.17 to 1.40)
Shields and Taylor (2015)	Structured walking programme	9 weeks	0.36 (−0.81 to 1.52)
Ulrich *et al.* (2011)	Modified bicycle	12 months	0.35 (−0.31 to 1.01)
van Schijndel-Speet *et al.* (2017)	PA programme and education programme	6 months	Fast walkers: 0.51 (−0.17 to 1.19) Slow walkers: 0.03 (−1.16 to 1.21)
		8 months	Fast walkers: 0.91 (0.20 to 1.60) Slow walkers: Not reported

PA, physical activity.

[†]Bergstrom *et al.* (2013) did not report means and standard deviations to allow for effect size calculation, however an effect size was reported.

change techniques with a wait-list control group in 102 participants. There was no difference between the groups for physical activity immediately post-intervention (SMD −0.04, 95% CI −0.47 to 0.39).

Three trials (McDermott *et al.* 2012; Bergstrom *et al.* 2013; Curtin *et al.* 2013) used multimodal interventions that included physical activity and nutrition education components. One of these trials (Bergstrom *et al.* 2013) compared a 12- to 16-month intervention with a wait-list control group in 130 participants. Data (i.e. means and standard deviations) were not reported to allow calculation of SMDs. However, the authors reported a positive intervention effect on physical activity, which was statistically significant ($P = 0.045$), with an effect size of 0.29. The second of these trials (McDermott *et al.* 2012) compared a 12-week health promotion programme with a hygiene and safety programme in 432 participants. There was no difference in moderate to vigorous physical activity between the groups at 12 months (SMD 0.14, 95% CI −0.23 to 0.50). The third of these trials (Curtin *et al.* 2013) compared a 9-month education programme with the same education programme that also included a behavioural intervention programme in 21 participants. There was no difference in physical activity levels between the groups at 12 months (SMD 0.73, 95% CI −0.22 to 1.68).

Lastly, one trial (van Schijndel-Speet *et al.* 2017) compared an 8-month physical activity framework and education programme with usual care in 131 participants. Participants in both groups were stratified according to their walking speed; fast and slow walkers. At 6 months, differences between groups were not statistically significant for both subgroups (fast walkers SMD 0.51, 95% CI −0.17 to 1.19; slow walkers SMD 0.03, 95% CI −1.16 to 1.21). However, for fast walkers, there was a large statistically significant improvement in physical activity levels at 8 months (SMD 0.91, 95% CI 0.20 to 1.60). No data were reported for the 8-month time point for the slow walkers.

Discussion

The aim of this study was to systematically review available evidence that evaluated the effectiveness of interventions to increase physical activity in individuals with ID. Two previous systematic reviews reported physical activity interventions can improve fitness (e.g. muscle strength), balance and psychological outcomes, in adults with ID (Bartlo & Klein 2011; Heller *et al.* 2011). However, these reviews did not evaluate whether the interventions were effective at increasing physical activity levels. Further, a recent systematic review investigated the efficacy of interventions to increase physical activity in children and adolescents. However, unlike the present study, adults were not included. Our study showed that the body of evidence was small, with only nine randomised controlled trials having investigated the effectiveness of interventions to increase physical activity for individuals with ID. The majority of the trials (six trials) found no differences between groups that suggests the experimental interventions that have been evaluated are no more effective than the control interventions. Only three trials (Bergstrom *et al.* 2013; Shields *et al.* 2013; van Schijndel-Speet *et al.* 2017) found positive effects on physical activity, and these will be discussed in more detail in the succeeding text.

A 10-week supervised progressive resistance training programme appears to be an effective intervention to maintain physical activity levels in young people with Down syndrome (Shields *et al.* 2013) for at least 3 months post-intervention. This effect occurred as a result of participants of the programme maintaining their physical activity levels after the intervention ceased, whereas physical activity levels of the control group participants declined. These findings suggest that a key benefit of the training programme is that it assists people in establishing a routine that involves daily physical activity while also preventing overall physical activity levels from declining over time. Other benefits to a progressive resistance training programme includes being simple to perform and relatively simple to resource (improving applicability to the intended population). However, while the intervention has shown to be beneficial for young adults with Down syndrome, additional evaluation is required to confirm if this intervention is effective in different populations with ID.

Of the multimodal interventions, a 12- to 16-month multicomponent diet and physical activity health promotion programme involving carers and participants was found to significantly improve physical activity levels post-intervention (Bergstrom *et al.* 2013). The involvement of a caregiver as part of

the intervention is likely to have been an important contribution to its effectiveness, as caregivers play a key role in encouraging physical activity (Heller *et al.* 2004; Mahy *et al.* 2010) and can assist in reducing the complexity of a multicomponent intervention. Further, adaptability of a programme to the routines of carers and residents was also favourable as it made the programme easier to implement and supported changes to physical activity. Although the intervention resulted in improvements in physical activity, this was only found at the completion of the trial (16 months), so the long-term effect of the programme on physical activity is unclear.

Another multimodal intervention using a physical activity and fitness programme also showed a large effect in improving physical activity at 8 months in people with ID that were 'fast walkers' (van Schijndel-Speet *et al.* 2017). The features of this intervention were advantageous, as, first, the programme used staff members from the day activity centres to conduct the programme at the same facility, so the participants were familiar with the setting and the individuals implementing the intervention. Second, the programme was adapted to the needs of the participants, which helped to incorporate physical activity into their daily routine. This programme highlights the importance of designing interventions that use established facilitators to physical activity; these include support from others, familiarity and routine (Mahy *et al.* 2010).

For the remaining trials, particularly those with an intervention that involved a health promotion programme, it is possible that the lack of effect of the interventions is related to the difficulty in adapting complex behaviour change interventions for people with ID. For example, despite interventions adhering to guidelines for developing physical activity interventions for disadvantaged groups (Michie *et al.* 2009) – by simplifying several components of the programme and avoiding complex behaviour change techniques – it was reported that participants still expressed difficulty in completing these tasks (Melville *et al.* 2015). Additionally, some interventions were based on social cognitive theory where the main principle is the concept of self-efficacy, which is a belief in one's capabilities in performing a behaviour. It also identifies that the outcomes of the behaviour must be valued by the individual (Bandura 1997). Individuals with ID may not perceive the outcomes of the interventions as rewarding or valuable. Further, another theory that may explain these findings is self-determination theory, which highlights the importance of both intrinsic and extrinsic motivation as precursors to behaviour change (Deci & Ryan 2008). It is possible that a lack of motivation (either intrinsic or extrinsic) may have influenced the outcomes of the intervention and participants' intentions to continue performing physical activity in the long term. The use of a mentor, however, has the ability to overcome these barriers and improve physical activity by supporting the individual. This is supported by three of the studies reviewed (Bergstrom *et al.* 2013; Shields *et al.* 2013; van Schijndel-Speet *et al.* 2017) – where a mentor to support the interventions was utilised – all of which reported a positive effect on physical activity.

An important consideration when interpreting the findings of this review relates to the quality of the included trials. None of the included trials were assessed to be at low risk of bias – all of the trials were rated as high risk of bias in at least one, and usually across multiple, domains of the Cochrane Risk of Bias Tool. Specifically, trials were often at risk of selection bias (from lack of allocation concealment), detection bias (from lack of assessor blinding) and attrition bias (from incomplete outcome data). Although we acknowledge the inherent difficulty in blinding participants to non-pharmacologic (e.g. physical) interventions of any kind in clinical trials (Boutron *et al.* 2004), performance bias is a risk across the included trials. Future trials need to implement greater methodological rigour (i.e. allocation concealment and assessor blinding) to confirm the true effectiveness of interventions. An additional limitation is that not all included trials were based on a theoretical framework, which is considered an important prerequisite for designing an effective physical activity intervention (Temple & Walkley 2007). Suboptimal reporting of adverse events was also an issue. Only one trial (van Schijndel-Speet *et al.* 2017) reported mild adverse events that occurred during the intervention. However, none of the trials that measured adverse events reported any (Bergstrom *et al.* 2013; Curtin *et al.* 2013; Shields *et al.* 2013; Melville *et al.* 2015; Shields & Taylor 2015), which suggests that thoughtfully designed physical activity interventions are safe and feasible in this population.

There are several strengths and potential limitations of this review. We used a robust search strategy and used two reviewers to independently screen studies for inclusion, perform data extraction and risk of bias assessment. This review included only randomised controlled trials, as this is a rigorous research design that minimises confounding, and if methodologically rigorous, reduces bias. The inclusion of randomised controlled trials could include any type of randomised controlled trial, including pilot trials that randomised at allocation, which included two pilot trials in this review (Curtin *et al.* 2013; Shields & Taylor 2015). Although an issue associated with pilot randomised controlled trials is their small sample size, we included these types of studies as they are important in providing information on the feasibility of an idea or intervention that may help guide a larger scaled trial (Leon *et al.* 2011; Spieth *et al.* 2016). Furthermore, if the samples and methods of any included pilot randomised controlled trials were homogenous with any other trials that were included in this review, we could have included these in a planned meta-analyses. However, we found that following the final inclusion of trials, we were unable to conduct meta-analyses.

Conclusions

There is inconsistent evidence of the effects of interventions on improving physical activity in individuals with ID. A progressive resistance training programme was found to maintain physical activity levels in adolescents with Down syndrome, while a multicomponent diet and physical activity programme and a physical activity and fitness programme were found to improve physical activity levels in adults with ID. However, future trials using rigorous research designs are required to confirm these effects and establish whether other interventions designed to increase physical activity in people with ID are effective.

Source of Funding

This work was supported by an Australian Government Research Training Program Scholarship.

Conflict of interest

The authors declare that they have no competing interests.

Authors' contributions

N. M. H., S. E. M. and K. B. L. conceived the study. N. M. H. conducted the systematic search and N. M. H. and S. E. M. performed the study selection. N. M. H. and K. B. L. completed the risk of bias assessment. N. M. H. and S. E. M. performed the data extraction. N. S. provided her expertise in the field and assisted in guiding data extraction. This manuscript was drafted by N. M. H. with the assistance of S. E. M., K. B. L. and N. S. who made amendments where required. N. S. also authored two studies used in this review (Shields *et al.* 2013; Shields & Taylor 2015). N. S. was not involved in the selection of articles, appraisal of methodological quality or the calculations of effects. All authors have read and approved the final manuscript.

References

Angulo-Barroso R., Burghardt A. R., Lloyd M. & Ulrich D. A. (2008) Physical activity in infants with Down syndrome receiving a treadmill intervention. *Infant Behavior & Development* **31**, 255–69.

Bandura A. (1997) *Self-efficacy: The Exercise of Control*. Freeman, NY, New York.

Barr M. & Shields N. (2011) Identifying the barriers and facilitators to participation in physical activity for children with Down syndrome. *Journal of Intellectual Disability Research* **55**, 1020–33.

Bartlo P. & Klein P. J. (2011) Physical activity benefits and needs in adults with intellectual disabilities: systematic review of the literature. *American Journal on Intellectual and Developmental Disabilities* **116**, 220–32.

Bergstrom H., Hagstromer M., Hagberg J. & Elinder L. S. (2013) A multi-component universal intervention to improve diet and physical activity among adults with intellectual disabilities in community residences: a cluster randomised controlled trial. *Research in Developmental Disabilities* **34**, 3847–57.

Boutron I., Tubach F., Giraudeau B. & Ravaud P. (2004) Blinding was judged more difficult to achieve and maintain in nonpharmacologic than pharmacologic trials. *Journal of Clinical Epidemiology* **57**, 543–50.

Caspersen C. J., Powell K. E. & Christenson G. M. (1985) Physical activity, exercise, and physical fitness: definitions

and distinctions for health-related research. *Public Health Reports* **100**, 126–31.

Curtin C., Bandini L. G., Must A., Gleason J., Lividini K., Phillips S. *et al.* (2013) Parent support improves weight loss in adolescents and young adults with Down syndrome. *Journal of Pediatrics* **163**, 1402–8 e1.

Deci E. L. & Ryan R. M. (2008) Self-determination theory: a macrotheory of human motivation, development, and health. *Canadian Psychology/Psychologie Canadienne* **49**, 182–5.

Durstine J. L., Gordon B., Wang Z. & Luo X. (2013) Chronic disease and the link to physical activity. *Journal of Sport and Health Science* **2**, 3–11.

Emerson E. (2005) Underweight, obesity and exercise among adults with intellectual disabilities in supported accommodation in Northern England. *Journal of Intellectual Disability Research* **49**, 134–43.

Faraone S. V. (2008) Interpreting estimates of treatment effects: implications for managed care. *Pharmacy and Therapeutics* **33**, 700–11.

Frey G. C. (2004) Comparison of physical activity levels between adults with and without mental retardation. *Journal of Physical Activity and Health* **1**, 235–45.

Heller T., Hsieh K. & Rimmer J. H. (2004) Attitudinal and psychosocial outcomes of a fitness and health education program on adults with down syndrome. *American Journal of Mental Retardation* **109**, 175–85.

Heller T., McCubbin J. A., Drum C. & Peterson J. (2011) Physical activity and nutrition health promotion interventions: what is working for people with intellectual disabilities? *Intellectual and Developmental Disabilities* **49**, 26–36.

Higgins J. P. T., Altman D. G. & Sterne J. A. C. (2011) Chapter 8: assessing risk of bias in included studies. In: *Cochrane Handbook for Systematic Reviews of Interventions* (eds J. P. Higgins & S. Green), Version 5.1.0 (updated March 2011). The Cochrane Collaboration.

Hinckson E. A. & Curtis A. (2013) Measuring physical activity in children and youth living with intellectual disabilities: a systematic review. *Research in Developmental Disabilities* **34**, 72–86.

Krahn G. L., Hammond L. & Turner A. (2006) A cascade of disparities: health and health care access for people with intellectual disabilities. *Mental Retardation and Developmental Disabilities Research Reviews* **12**, 70–82.

Lefebvre C., Manheimer E. & Glanville J. (2011) Chapter 6: searching for studies. In: *Cochrane Handbook for Systematic Reviews of Interventions* (eds J. P. Higgins & S. Green), Version 5.1.0 (updated March 2011). The Cochrane Collaboration.

Lennox N., Bain C., Rey-Conde T., Purdie D., Bush R. & Pandeya N. (2007) Effects of a comprehensive health assessment programme for Australian adults with intellectual disability: a cluster randomized trial. *International Journal of Epidemiology* **36**, 139–46.

Leon A. C., Davis L. L. & Kraemer H. C. (2011) The role and interpretation of pilot studies in clinical research. *Journal of Psychiatric Research* **45**, 626–9.

Mahy J., Shields N., Taylor N. F. & Dodd K. J. (2010) Identifying facilitators and barriers to physical activity for adults with Down syndrome. *Journal of Intellectual Disability Research* **54**, 795–805.

Matthews L., Hankey C., Penpraze V., Boyle S., Macmillan S., Miller S. *et al.* (2011) Agreement of accelerometer and a physical activity questionnaire in adults with intellectual disabilities. *Preventive Medicine* **52**, 361–4.

McDermott S., Whitner W., Thomas-Koger M., Mann J. R., Clarkson J., Barnes T. L. *et al.* (2012) An efficacy trial of 'Steps to Your Health', a health promotion programme for adults with intellectual disability. *Health Education Journal* **71**, 278–90.

McGarty A. M., Downs S. J., Melville C. A. & Harris L. (2018) A systematic review and meta-analysis of interventions to increase physical activity in children and adolescents with intellectual disabilities. *Journal of Intellectual Disability Research* **62**, 312–29.

McGarty A. M. & Melville C. A. (2018) Parental perceptions of facilitators and barriers to physical activity for children with intellectual disabilities: a mixed methods systematic review. *Research in Developmental Disabilities* **73**, 40–57.

Melville C. A., Hamilton S., Hankey C. R., Miller S. & Boyle S. (2007) The prevalence and determinants of obesity in adults with intellectual disabilities. *Obesity Reviews* **8**, 223–30.

Melville C. A., Mitchell F., Stalker K., Matthews L., McConnachie A., Murray H. M. *et al.* (2015) Effectiveness of a walking programme to support adults with intellectual disabilities to increase physical activity: walk well cluster-randomised controlled trial. *International Journal of Behavioral Nutrition and Physical Activity* **12**, 125.

Michie S., Jochelson K., Markham W. A. & Bridle C. (2009) Low-income groups and behaviour change interventions: a review of intervention content, effectiveness and theoretical frameworks. *Journal of Epidemiology and Community Health* **63**, 610–22.

Moher D., Liberati A., Tetzlaff J. & Altman D. G. (2009) Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *BMJ [British Medical Journal]* **339**, b2535.

Neman R., Roos P., McCann R. M., Menolascino F. J. & Heal L. W. (1975) Experimental evaluation of sensorimotor patterning used with mentally retarded children. *American Journal of Mental Deficiency* **79**, 372–84.

Penpraze V., Reilly J. J., MacLean C. M., Montgomery C., Kelly L. A., Paton J. Y. *et al.* (2006) Monitoring of physical activity in young children: how much is enough? *Pediatric Exercise Science* **18**, 483–91.

Pitetti K. H., Beets M. W. & Combs C. (2009) Physical activity levels of children with intellectual disabilities during school. *Medicine and Science in Sports and Exercise* **41**, 1580–6.

Review Manager (2014) (RevMan) [Computer program]. Version [5.3]. Copenhagen: The Nordic Cochrane Centre.

Robertson J., Emerson E., Gregory N., Hatto C., Turner S., Kessissoglou S. *et al.* (2000) Lifestyle related risk factors for poor health in residential settings for people with intellectual disabilities. *Research in Developmental Disabilities* **21**, 469–86.

Schijndel-Speet M., Wijck R., Evenhuis H. M. & Echteld M. A. (2012) Effectiveness of a physical activity program for seniors with intellectual disabilities: results of a RCT. *Journal of Intellectual Disability Research* **56**, 757.

Shields N. & Taylor N. F. (2015) The feasibility of a physical activity program for young adults with Down syndrome: a phase II randomised controlled trial. *Journal of Intellectual and Developmental Disability* **40**, 115–25.

Shields N., Taylor N. F. & Dodd K. J. (2008) Effects of a community-based progressive resistance training program on muscle performance and physical function in adults with Down syndrome: a randomized controlled trial. *Archives of Physical Medicine and Rehabilitation* **89**, 1215–20.

Shields N., Taylor N. F., Wee E., Wollersheim D., O'Shea S. D. & Fernhall B. (2013) A community-based strength training programme increases muscle strength and physical activity in young people with Down syndrome: a randomised controlled trial. *Research in Developmental Disabilities* **34**, 4385–94.

SIGN (2017) Search filters. *SIGN*. Available at: www.sign.ac.uk/search-filters.html (retrieved 11 October 2016).

Spieth P. M., Kubasch A. S., Penzlin A. I., Illigens B. M.-W., Barlinn K. & Siepmann T. (2016) Randomized controlled trials – a matter of design. *Neuropsychiatric Disease and Treatment* **12**, 1341–9.

Stanish H. I., Temple V. A. & Frey G. C. (2006) Health-promoting physical activity of adults with mental retardation. *Mental Retardation and Developmental Disabilities Research Reviews* **12**, 13–21.

Sutherland G., Couch M. A. & Iacono T. (2002) Health issues for adults with developmental disability. *Research in Developmental Disabilities* **23**, 422–45.

Sylvia L. G., Bernstein E. E., Hubbard J. L., Keating L. & Anderson E. J. (2014) A practical guide to measuring physical activity. *Journal of the Academy of Nutrition and Dietetics* **114**, 199–208.

Temple V. A. & Stanish H. I. (2009) Pedometer-measured physical activity of adults with intellectual disability: predicting weekly step counts. *American Journal on Intellectual and Developmental Disabilities* **114**, 15–22.

Temple V. A. & Walkley J. W. (2007) Perspectives of constraining and enabling factors for health-promoting physical activity by adults with intellectual disability. *Journal of Intellectual and Developmental Disability* **32**, 28–38.

Tudor-Locke C., Craig C. L., Aoyagi Y., Bell R. C., Croteau K. A., De Bourdeaudhuij I. *et al.* (2011) How many steps/day are enough? For older adults and special populations. *The International Journal of Behavioral Nutrition and Physical Activity* **8**, 80.

Ulrich D. A., Burghardt A. R., Lloyd M., Tiernan C. & Hornyak J. E. (2011) Physical activity benefits of learning to ride a two-wheel bicycle for children with Down syndrome: a randomized trial. *Physical Therapy* **91**, 1463–77.

Ulrich D. A., Lloyd M. C., Tiernan C. W., Looper J. E. & Angulo-Barroso R. M. (2008) Effects of intensity of treadmill training on developmental outcomes and stepping in infants with Down syndrome: a randomized trial. *Physical Therapy* **88**, 114–22.

van Schijndel-Speet M., Evenhuis H. M., van Empelen P., van Wijck R. & Echteld M. A. (2013) Development and evaluation of a structured programme for promoting physical activity among seniors with intellectual disabilities: a study protocol for a cluster randomized trial. *BMC Public Health* **13**, 746.

van Schijndel-Speet M., Evenhuis H. M., van Wijck R., van Montfort K. C. & Echteld M. A. (2017) A structured physical activity and fitness programme for older adults with intellectual disabilities: results of a cluster-randomised clinical trial. *Journal of Intellectual Disability Research* **61**, 16–29.

Warburton D. E., Nicol C. W. & Bredin S. S. (2006) Health benefits of physical activity: the evidence. *Canadian Medical Association Journal* **174**, 801–9.

Whittaker G. A., Munteanu S. E., Menz H. B., Tan J. M., Rabusin C. L. & Landorf K. B. (2017) Foot orthoses for plantar heel pain: a systematic review and meta-analysis. *British Journal of Sports Medicine* **52**, 322–8.

Wong S. S. L., Wilczynski N. L. & Haynes R. B. (2006) Developing optimal search strategies for detecting clinically sound treatment studies in EMBASE. *Journal of the Medical Library Association* **94**, 41–7.

World Health Organization (2010) *Global recommendations on physical activity for health*. World Health Organization. Available at: http://apps.who.int/iris/bitstream/10665/44399/1/9789241599979_eng.pdf (retrieved 10 October 2016).

Accepted 1 October 2018

Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

Data S1. Supporting information
Data S2. Supporting information
Data S3. Supporting information

Chapter 3 – Randomised pilot study

Preface

The findings of the systematic review (Chapter 2) showed only nine studies have evaluated the effectiveness of interventions to increase physical activity in individuals with intellectual disabilities, and only three of interventions were effective in improving physical activity. No study had evaluated footwear as an intervention to improve physical activity.

As raised in the introduction to this thesis (Chapter 1, page 26), poorly-fitting footwear can interfere with walking patterns, thus making physical activity more challenging for children with Down syndrome. Poorly-fitting footwear may also contribute to foot pain, which is a risk factor for reduced physical activity. Accordingly, improving footwear fit may have a positive effect on physical activity levels through reduction of foot pain and improved walking patterns.

This chapter aims to address *Objective 2* of this thesis, to determine the feasibility of conducting a definitive randomised trial to evaluate the efficacy of custom-fitted footwear for increasing physical activity in children with Down syndrome. In this study*, custom-fitted footwear* refers to footwear that was fitted according to the manufacturer's protocol using a foot gauge (provided by the manufacturer) to measure foot length and width.

This chapter was published in the journal of *Disability and Rehabilitation* in 2019 with the associated online supplementary files presented in Appendix 2. The citations within this chapter relate to the reference list of the publication, not the reference list included at the end of this thesis.

3.1 Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study

The reference for this publication is:

Hassan NM, Shields N, Landorf KB, Buldt AK, Taylor NF, Evans AM, Williams CM, Menz HB, Munteanu SE. Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study. *Disability and Rehabilitation*; 2019:22;1 – 10. DOI: https://doi.org/10.1080/09638288.2019.1692380

Disability and Rehabilitation

ISSN: 0963-8288 (Print) 1464-5165 (Online) Journal homepage: https://www.tandfonline.com/loi/idre20

Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study

Nirmeen M. Hassan, Nora Shields, Karl B. Landorf, Andrew K. Buldt, Nicholas F. Taylor, Angela M. Evans, Cylie M. Williams, Hylton B. Menz & Shannon E. Munteanu

To cite this article: Nirmeen M. Hassan, Nora Shields, Karl B. Landorf, Andrew K. Buldt, Nicholas F. Taylor, Angela M. Evans, Cylie M. Williams, Hylton B. Menz & Shannon E. Munteanu (2019): Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study, Disability and Rehabilitation, DOI: 10.1080/09638288.2019.1692380

To link to this article: https://doi.org/10.1080/09638288.2019.1692380

DISABILITY AND REHABILITATION
https://doi.org/10.1080/09638288.2019.1692380

Taylor & Francis
Taylor & Francis Group

ORIGINAL ARTICLE

Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study

Nirmeen M. Hassan[a,b], Nora Shields[b,c,d], Karl B. Landorf[a,c], Andrew K. Buldt[a,c], Nicholas F. Taylor[c,d], Angela M. Evans[a], Cylie M. Williams[e,f], Hylton B. Menz[a,c] and Shannon E. Munteanu[a,b,c]

[a]Discipline of Podiatry, School of Allied Health, Human Services and Sport, La Trobe University, Melbourne, Victoria, Australia; [b]Living with Disability Research Centre, School of Allied Health, Human Services and Sport, La Trobe University, Melbourne, Victoria, Australia; [c]La Trobe Sport and Exercise Medicine Research Centre, School of Allied Health, Human Services and Sport, La Trobe University, Melbourne, Victoria, Australia; [d]Discipline of Physiotherapy, School of Allied Health, Human Services and Sport, La Trobe University, Melbourne, Victoria, Australia; [e]Physiotherapy Department, School of Primary and Allied Health Care, Monash University, Melbourne, Victoria, Australia; [f]Allied Health, Peninsula Health, Melbourne, Victoria, Australia

ABSTRACT

Purpose: To determine the feasibility of conducting a definitive randomised trial to evaluate the efficacy of custom-fitted footwear for increasing physical activity in children and adolescents with Down syndrome.

Methods: Assessor-blinded, parallel-group randomised pilot study. Thirty-three children and adolescents with Down syndrome were randomly allocated to a custom-fitted footwear group (Clarks® footwear) or a wait-list control group. Six feasibility domains were evaluated at baseline, 6 and 12 weeks; demand (recruitment), implementation (co-interventions and adherence), acceptability, practicality (adverse events), limited efficacy testing (physical activity, disability associated with foot and ankle problems, and gait parameters), and adaptation (shoe-fit).

Results: Three participants were recruited per month. The use of co-interventions was common with six control group participants purchasing new footwear during the study. Mean adherence was 35 h/week in the custom-fitted footwear group, and there were few minor adverse events. There were trends for differences in physical activity favouring the custom-fitted footwear, but no trends for differences in disability associated with foot and ankle problems or gait parameters. The fit of the custom-fitted footwear was no better than participants' regular footwear.

Conclusions: A definitive randomised trial is feasible. However, recruitment, use of co-interventions and footwear fit need further consideration.

➤ IMPLICATIONS FOR REHABILITATION

- Conducting a definitive randomised trial to determine the efficacy of custom-fitted footwear in increasing physical activity in children and adolescents with Down syndrome is feasible.
- Custom-fitted footwear may improve physical activity in children and adolescents with Down syndrome.
- Commercially available footwear may not be suitable for children and adolescents with Down syndrome due to their unique foot shape.

ARTICLE HISTORY
Received 10 June 2019
Revised 7 November 2019
Accepted 10 November 2019

KEYWORDS
Down syndrome; exercise; feasibility studies; randomised controlled trial; shoes

Introduction

Guidelines recommend all children perform at least 60 min of moderate to vigorous physical activity per day to obtain health benefits, such as physical and psychological wellbeing, and improved self-esteem [1–3]. Children with Down syndrome often do not meet physical activity recommendations and are less active compared to their typically developing peers [4]. There are many personal, social, and environmental reasons why children with Down syndrome have low physical activity levels, including competing family responsibilities, lack of accessible programs, and reduced physical skills [5]. Physiological characteristics of children with Down syndrome, such as congenital heart conditions (e.g., atrioventricular septal defect), hypotonia [6], and reduced muscular strength and cardiovascular fitness [7], can also make it harder to participate in physical activity.

An additional barrier to participation in physical activity is foot problems, including foot pain, which is common in children and young adolescents with Down syndrome [8,9]. A population-based study of 197 adolescents and young adults with Down syndrome in Australia found nearly two-thirds experienced foot problems and reported foot pain that negatively impacts their life [8]. Structural anomalies such as hallux valgus (bunions), toe deformities and pes planus (flat feet) can also cause foot pain and are prevalent among children with Down syndrome [10,11]. These anomalies can negatively affect walking and participation in daily activities in children with Down syndrome [12–15].

CONTACT Nirmeen M. Hassan n.hassan@latrobe.edu.au Discipline of Podiatry, School of Allied Health, Human Services and Sport, La Trobe University, Corner Plenty Road and Kingsbury Drive, Melbourne, Victoria 3086, Australia

Supplemental data for this article can be accessed here.

Ill-fitting footwear is common in children and adolescents with Down syndrome and may also exert a negative influence on physical activity [16,17]. A cross-sectional study involving 50 children and adolescents with Down syndrome in Australia found that 8% of participants wore appropriately fitting footwear [17]. Specifically, 60% of participants wore footwear that was too narrow and 54% wore footwear that was too long, which is most likely due to the unique foot shape of children with Down syndrome. Children with Down syndrome often have a wider forefoot [10], and as a consequence, they are more likely to wear footwear that is either too long to allow for the extra width requirement, or that is an appropriate length but too narrow [16].

Given ill-fitting footwear is common in children and adolescents with Down syndrome and is associated with reduced physical activity, it is possible that wearing appropriately fitting footwear may improve their physical activity levels. However, we are not aware of any trials that have evaluated the efficacy of custom-fitted footwear to increase physical activity levels in children and adolescents with Down syndrome. Therefore, the aim of this randomised pilot study was to determine the feasibility of conducting a definitive randomised trial to determine the efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome.

Methods

This study is reported in accordance with the Consolidated Standards of Reporting Trials (CONSORT) extension for randomised pilot and feasibility trials [18], and was registered prospectively with the Australian New Zealand Clinical Trials Registry (ACTRN12616001118493).

Study design

This was an assessor-blinded, parallel-group randomised pilot study. Six domains of Bowen's framework for feasibility studies [19] were evaluated; *demand* (recruiting participants with Down syndrome), *implementation* (intervention adherence and co-interventions), *acceptability* (participants' experiences of wearing the footwear), *practicality* (adverse events), *limited efficacy testing* (differences between groups for physical activity, disability associated with foot and ankle problems and gait parameters), and adaptation (footwear fit).

Ethics approval

La Trobe University Human Ethics Committee (HEC16-027) approved this study. Written informed consent (including the use of anonymised data) was obtained from parents or guardians, and where appropriate, children and adolescents provided written assent for participation.

Participant recruitment and eligibility criteria

Participants were recruited through a community, member-based disability organisation for people with Down syndrome (Down Syndrome Victoria). Potential participants were sent a flyer with information outlining the trial. Interested parents or guardians contacted the research team directly.

Eligibility criteria

Participants were eligible to participate if they: (i) were aged 5–17 years, (ii) had Down syndrome, and (iii) were able to understand simple verbal instructions in English. Participants were excluded if they (i) had another health condition with the potential to affect their physical activity levels, (ii) significant joint laxity that resulted in subluxation, or (iii) used an assistive ambulatory device. Participants were required to be adherent with wearing an accelerometer for a minimum of 10 h for a minimum period of four days, including one weekend day [20]. Participants were also excluded if the available footwear did not adequately accommodate their foot size (greater than UK children's size 6).

Randomisation

Participants were randomly allocated to either a custom-fitted footwear group or a wait-list control group after the baseline assessment using a computer-generated random number sequence with permuted block sizes of four. The allocation sequence was generated and held by a researcher (NFT) not directly involved in recruitment, allocation, data collection, data processing, or statistical analysis. Each participant's allocation was contained in sequentially numbered sealed, opaque envelopes.

Custom-fitted footwear group

Participants in the custom-fitted footwear group were provided with two-pairs of custom-fitted footwear (Clarks®, Brand Collective, Melbourne, Australia) via post: a pair of casual shoes and a pair of school shoes, or two pairs of casual shoes if school shoes were not necessary. Participants were given the option of Velcro® or lace style fixation (Figure 1). The casual shoes were

Figure 1. Models of the Clarks® footwear used as the intervention. Top panel, school shoes, from left to right: Lochie™, Laura™, and Daytona™. Second panel, casual shoes, from left to right: Ventura™ and Vancouver™.

Ventura™ and Vancouver™ models and the school shoes were the Laura™, Daytona™, and Lochie™ models. Half sizes and a range of width sizes (D to H fittings) were available for the school shoes. Only full-sized length fittings and a fixed width fitting of E+ (with a removable insole for increased space) were available for the casual shoes.

The shoes were fitted using a foot measuring gauge, as per the manufacturer's protocol. Prior to the study, the researcher fitting the shoes (NMH), a podiatrist, attended a one-hour training session with staff from Clarks® on footwear fitting. Where measurements were between two readings, the next size up was selected. Where there were differences in dimensions of the measured feet (i.e., left and right feet), the larger of the measures was used for sizing. Once participants received the intervention, they were instructed to wear the footwear for the next 12 weeks.

The intervention commenced immediately after the participants were allocated the footwear. Previous work suggests that 4 weeks is considered a reasonable time to allow for habituation [21,22]. This allowed sufficient time for participants to have habituated to the footwear by the first outcome assessment time (6 weeks).

Control group

The control group was a wait-list control. Participants were fitted for custom-fitted footwear using the above procedures; however, they received their footwear after their final assessment (i.e., at the end of the study). Participants were informed to continue wearing their existing footwear for the duration of their involvement in the study.

Outcome measures

Outcomes were assessed at baseline prior to randomisation, at 6 weeks (via post), and at 12 weeks. Outcome assessment was conducted at baseline and 12 weeks by an assessor who was blind to group allocation (the assessment at 6 weeks did not require an assessor as it was posted to participants to complete).

Demand was calculated as the rate of participant recruitment (number randomised per month).

Implementation was assessed at 6 and 12 weeks by recording adherence and co-interventions. Adherence to wearing the custom-fitted footwear was reported via a parent-reported questionnaire, which asked for the number of hours per day and number of days their child wore the custom-fitted footwear during the previous 6 weeks [23]. To minimise burden, parents documented adherence over the previous 6 weeks, rather than by daily diary entries. Co-intervention use was evaluated in the custom-fitted footwear group participants, with a parent-completed questionnaire to elicit use of any other intervention that may have affected physical activity (e.g., visits to health care practitioners, illness, new activities, or purchase and use of other footwear) during the previous 6 weeks.

Acceptability of the footwear was assessed in the custom-fitted footwear group participants through brief semi-structured interviews involving children and their parents at 12 weeks. Interviews were conducted by a member of the research team (AKB). Participants and their parents were asked about their perceptions of the footwear including comfort and fit, and any perceived changes to physical activity levels.

Practicality was evaluated in the custom-fitted footwear group participants at 6 and 12 weeks using a questionnaire that recorded adverse events, including type, location, severity, and duration. In cases where severe adverse events occurred, parents or guardians were advised to contact the investigators and seek medical attention [24,25].

Limited efficacy testing compared differences between groups in: (i) physical activity levels (primary outcome) at baseline, 6 and 12 weeks, (ii) disability associated foot and ankle problems at 6 and 12 weeks, and (iii) gait parameters at 12 weeks. Physical activity was assessed using an ActiGraph® wGT3x-BT accelerometer (ActiGraph, Pensacola, FL). The accelerometer is a small, lightweight device worn at the hip using an elastic, removable belt, which was placed underneath clothing. Participants and parents were instructed on how to wear the accelerometer and were advised to keep it on for all waking hours for seven consecutive days, removing it only for water-based activities and sleep. The ActiGraph® wGT3x-BT accelerometer has previously been used to measure physical activity in children and adolescents with Down syndrome and has demonstrated validity and reliability [26,27]. The accelerometer was returned after wear via post. Data were included in the analysis if there was a recording of at least 10 h for a minimum period of four days, including one weekend day [20]. Wear time was classified using the Choi automated algorithm [28], and the accelerometers were programmed to capture physical activity in 15 s epochs. Physical activity was classified as sedentary, light, moderate, and vigorous using recommended cut-points [29]. We measured average number of days and hours the accelerometer was worn, steps per day, vector magnitude, sedentary activity, light activity, and moderate to vigorous physical activity.

Disability associated with foot and ankle problems was assessed at baseline, 6 weeks and 12 weeks using the Oxford Ankle Foot Questionnaire for Children (OxAFQ-C) parent version [30] because of the young age of some participants and the presence of intellectual disability [16]. The OxAFQ-C has demonstrated validity and reliability [30] and consists of three domains of disability: Physical (six items), School and play (four items), and Emotional (four items). The final domain (Footwear) reflects the issue of whether the participant's foot or ankle prevents them from wearing their preferred footwear. Each question uses a five-point Likert scale rated as never (4), rarely (3), sometimes (2), very often (1), or always (0). Domain scores are presented as a percentage, where 100% represents perfect health for the domain [28]. To calculate domain scores, the total of the scale item score was divided by the maximum possible score and multiplied by 100 [30].

Gait parameters were assessed at baseline and 12 weeks using the GaitRite® electronic walkway (CIR Systems, Inc, Franklin, NJ) for both groups. The GaitRite® records the location and timing of footprints as the participant walks over the mat, which has an active sensor area of 366 cm long and 61 cm wide, containing 13 824 pressure sensors with a spatial resolution of 1.27 cm and a sampling frequency of 80 Hz [31]. The following variables were measured: walking velocity, cadence, stride length, step length, base of support, toe-in/out angle, and step width. The test–retest reliability of spatio-temporal measurements recorded using the GaitRite® is good to excellent [32]. Gait was assessed under four conditions: the (i) custom-fitted school shoe, (ii) custom-fitted casual shoe, (iii) participant's usual casual shoe, and (iv) participant's usual school shoe. Participants familiarised themselves with walking on the GaitRite® walkway and completed three trials per condition while walking at a self-selected speed (the three trials were later averaged for each participant prior to data analysis). The order of the conditions were randomised for each participant to minimise ordering effects. To exclude the effects of acceleration and deceleration, the first and last steps were removed from the analysis [33].

Adaptation was assessed by estimating footwear fit. Footwear fit measurements were collected at baseline but were analysed at the end of the study; that is, these measures were not used to

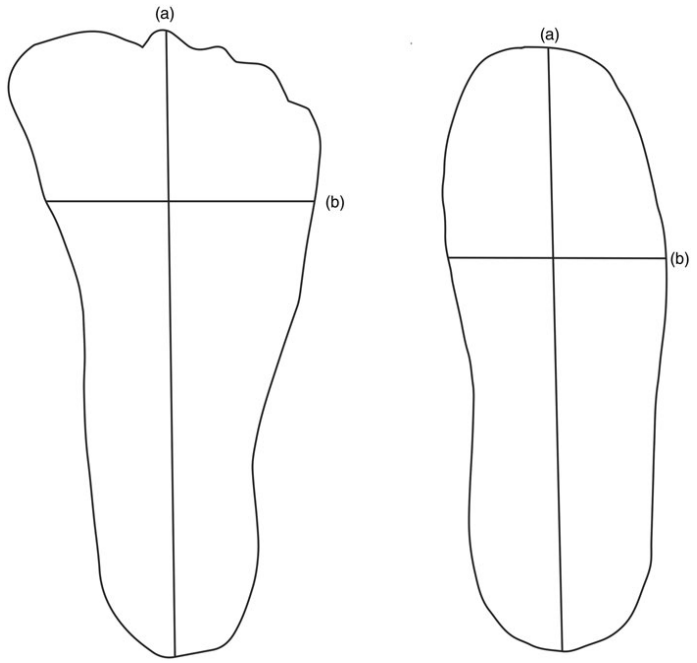


Figure 2. Foot and shoe measurements: (a) length measurement and (b) width measurement.

determine which shoes the participants received since this was not part of usual practice in custom-fitting shoes. The purpose was to understand whether an existing fitting process used in typically developing children was suitable for children with foot structural changes associated with Down syndrome. Footwear fit was estimated by comparing footwear length and width of the participant's right shoe to measurements of their right foot (Figure 2). Shoe dimensions were obtained by tracing the outline of the sole of the right shoe (or the inner sole of the shoe if removable) onto paper. The participant's foot dimensions were obtained by tracing the outline of the participant's right foot onto carbon paper imprint material in relaxed, bipedal standing. Foot length and shoe length were measured by drawing a longitudinal line from the bisection of the heel to the bisection of the second digit to obtain the length measurement [34]. Foot width and shoe width were measured by measuring width at the widest point of the forefoot region. To achieve this, a line perpendicular to the aforementioned longitudinal line was constructed at the point where the line representing the widest point of the forefoot intersected with the longitudinal line. Appropriate footwear fit was defined as a difference between foot length and shoe length of 0–20 mm, and a difference between foot width and shoe width of 0–10 mm. Differences in length less than 0 were considered too short and greater than 20 mm were considered too long [17]. Differences in width less than 0 were considered too narrow and greater than 10 mm were considered too wide [17].

Sample size and data analysis

Sample size was not formally determined *a priori* as we aimed to determine feasibility. We deemed a sample size of 30 participants was reasonable to allow estimation of key feasibility parameters. Statistical analysis was completed using IBM SPSS Statistics version 25.0 (IBM Corp, Armonk, NY).

Quantitative data were analysed according to the intention-to-treat principle [35]. Missing data were handled using multiple imputation [36]. For multiple imputation, five iterations were used, with age, group allocation, and baseline scores used as predictors. There was no data substitution for adverse events, adherence, and use of co-interventions. Descriptive statistics were used to report participant characteristics. Continuous data were explored for normality using standard tests (skewness and kurtosis) [37] and all variables were found to be normally distributed. Analysis of covariance (ANCOVA) was performed for between-group comparisons at 6 and 12 weeks, using baseline scores as covariates [38]. Effect sizes (standardised mean differences (SMDs)) and associated 95% confidence intervals were calculated for between-group differences.

Qualitative semi-structured interviews were audio-recorded and transcribed verbatim. Two researchers (SC and NMH) independently coded each transcript using an inductive method. The codes were then grouped into emerging themes and subthemes. Final themes were agreed on by consensus.

Results

Demand (including participant flow through the trial)

Over 11 months, 58 parents completed a telephone screening questionnaire to determine their child's eligibility for participation. Thirty-three children were eligible to take part and randomised. The recruitment rate was three participants per month. Reasons for exclusion are itemised in Figure 3. Seventeen participants

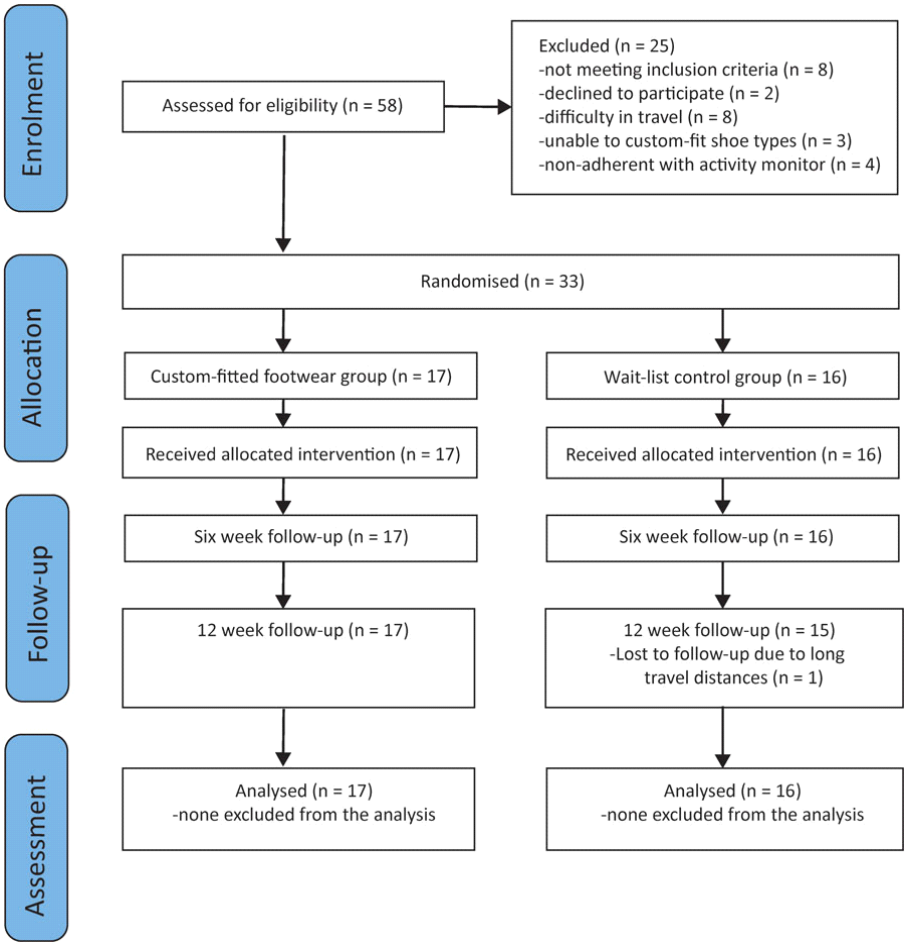


Figure 3. Flow of participants through study.

were allocated to the custom-fitted footwear group and 16 participants to the control group. Participants in both groups had similar baseline characteristics (Table 1). One participant was lost to follow-up.

Implementation (adherence and co-interventions)

Mid-way through the trial, one model of the allocated school shoes (Laura™ model, a Mary-Jane style shoe for girls) was discontinued by the manufacturer. This resulted in four female participants receiving the male equivalent school shoe (Lochie™ model) instead. Both models were made on the same shoe last and the only differences were in the appearance of the shoe and the amount of coverage of the dorsal aspect of the foot provided by the shoe. Although this replacement shoe did not change the fitting process, it may have affected participants' perceptions of the appearance of the shoe, which was an issue raised by one parent.

Data on adherence show that at 6 weeks, participants in the custom-fitted footwear group reported wearing the casual shoes for an average of 32.3 (SD 30.0) hours per week and the school shoes for an average of 33.1 (SD 12.9) hours per week. At 12 weeks, participants in the custom-fitted footwear group wore their casual shoes for an average of 35.0 (SD 43.1) hours and their school shoes for an average of 35.0 (SD 21.8) hours per week.

Some participants reported using co-interventions during the trial. At 6 weeks, one participant in the control group received a new pair of foot orthoses due to out-growing their existing foot orthoses. At 6 weeks, three participants in the custom-fitted footwear group and six participants in the control group commenced a new sport. At 12 weeks, four participants in the custom-fitted footwear group and control group commenced a new sport over the last 6 weeks. Six participants in the control group reported they purchased and wore new footwear during the trial.

Acceptability

Three themes emerged from the interviews: (i) footwear fit and comfort, (ii) perceived changes to physical activity, and (iii) perceptions of the footwear. Parents and participants indicated the

Table 1. Participant characteristics at baseline.

Characteristic	Custom-fitted footwear group ($n = 17$)	Control group ($n = 16$)
General		
Age (years)	9.7 (3.6)	9.6 (4.0)
Females/males, *n*	12/5	9/7
Height (m)	1.26 (0.2)	1.24 (0.2)
Weight (kg)	33.6 (14.3)	35.4 (17.7)
Body mass index (kg/m^2)	20.0 (4.3)	21.4 (5.0)
Type of DS[a], *n* (trisomy 21/translocation)	15/2	15/1
Level of perceived ID[b], *n* (unclear/mild/moderate/severe)	5/8/4/0	5/6/5/0
Current use of foot orthoses, n	4	4
Co-morbidities		
Hearing impairment, *n*	3	4
Vision impairment, *n*	2	4
Heart disease, *n*	2	0
Incontinence, *n*	1	3
Hypothyroidism, *n*	2	2
Foot health assessment		
Hallux valgus, *n*	1	2
Lesser toe deformity, *n*	10	6
FPI[c] (R[d])	8.5 (2.0)	8.5 (1.5)
Arch index (R)	0.30 (0.05)	0.29 (0.04)
Physical activity		
Days accelerometer worn (days)	6.0 (0.9)	6.0 (0.8)
Hours accelerometer worn (hours/day)	12.7 (1.2)	12.6 (1.4)
Vector magnitude per day (counts)	745 516 (239 783)	783 212 (303 109)
Steps per day	6981 (1280)	6942 (2218)
Time spent in sedentary activities per day (min)	555 (56)	564 (82)
Time spent in light activities per day (min)	64 (20)	65 (27)
Time spent in MVPA[e] per day (min)	93 (36)	96 (52)
OxAFQ-C[f] domain scores		
Physical (%)	74 (18)	72 (18)
School and play (%)	85 (18)	88 (15)
Emotional (%)	94 (6)	96 (7)
Concern (%)	62 (38)	70 (28)

Values are mean (SD) unless otherwise stated.
[a]Down syndrome.
[b]Intellectual disability.
[c]Foot Posture Index-6.
[d]Right foot.
[e]Moderate to vigorous physical activity.
[f]Oxford Ankle Foot Questionnaire for Children – higher domain scores indicate better health for the respective domain.

custom-fitted footwear fitted well and were comfortable. Parents reported their children had no issues wearing the shoes and did not show signs of refusal to wear the shoes. One parent reported:

> He has not taken them off. So generally he is one that would take his shoes on and off … once they're on in the morning, they're on all day, so they're obviously comfortable.

Another parent reported:

> When we first got these to him, he didn't object. Normally with new shoes he objects, but because we took the other ones away and he really liked these (intervention), there was no objection.

Participants reported the shoes helped them "run faster" or "play for longer." Many parents indicated they perceived an increase in their child's physical activity and improvement in the quality of their child's walking. One parent reported:

> I would say that he would do the same things but better. In a sense, with a lesser pair of shoes his legs possibly get tired more quickly … but with these shoes, his (foot) pronation seems to disappear.

All participants indicated they liked the shoes they received and described their favourite shoe features to be the colour, fit, and fixation style. Most participants preferred the casual shoe over the school shoe, describing the school shoes as "too hard" or they were difficult to break in, which at times caused them pain and discomfort. Participants indicated they preferred the shoes with Velcro® fixation over laces because of difficultly tying laces. Having Velcro® meant they were in control of wearing their own shoes.

Practicality (adverse events)

In the custom-fitted footwear group, one participant experienced a minor adverse event (blister) at 6 and 12 weeks. One participant reported the style of shoe was uncomfortable due to the closed-in upper of the school shoe (Lochie™ model) and another participant reported mild rubbing from their shoes at 12 weeks. There were no serious adverse events.

Limited efficacy testing (physical activity, disability associated with foot and ankle problems and gait parameters)

One participant did not return their physical activity monitor at 6 weeks, and two participants did not return their activity monitor at 12 weeks. At 6 weeks, the average accelerometer wear times per day were 12.9 h (SD 1.2) in the custom-fitted footwear group and 12.3 h (SD 1.1) in the control group. At 12 weeks, the average accelerometer wear times were 13.0 h (SD 1.2) in the custom-fitted footwear group and 12.3 h (SD 1.5) in the control group. There were trends for between-group differences in moderate to vigorous physical activity per day at 6 and 12 weeks, with greater differences observed at 6 weeks. Participants allocated to the custom-fitted footwear group demonstrated an increase in moderate to vigorous physical activity per day at 6 weeks (adjusted mean difference= 20 min; 95% CI −9 to 50, p= 0.170) and 12 weeks (adjusted mean difference= 8 min; 95% CI −21 to 37; p= 0.554). These effects were associated with trends for an increase in steps per day, reduced time spent in sedentary activity, as well as an increase in time spent in light activity in those allocated to the custom-fitted footwear group (Table 2).

There were no trends for between-group differences across domain scores of the OxAFQ-C (disability associated with foot and ankle problems) at 6 and 12 weeks (Table 2). There were also no trends for between-group differences for any gait parameters at 12 weeks (see online Supplementary File 1).

Adaptation (footwear fit)

Table 3 presents the estimated footwear fit. Overall ($n = 33$), the usual casual shoes and usual school shoes were considered to be an appropriate length in 20 (61%) and 17 (52%) participants, respectively, and an appropriate width in seven (21%) and six (18%) of participants, respectively. In those allocated to receive footwear as part of the custom-fitted footwear group ($n = 17$), the casual shoes and the school shoes were considered to be an appropriate length in seven (41%) and 10 (59%) participants, respectively, and an appropriate width in one (10%) and one (10%) participant, respectively. Those allocated to the wait-list control group received their footwear at the end of the study. The footwear fit measures for these participants showed that the casual shoes and the school shoes were considered to be an appropriate length in nine (56%) and eight (50%) participants, respectively, and an appropriate width in one (6%) and two (13%) participants, respectively.

Table 2. Physical activity and Oxford Ankle Foot Questionnaire for Children outcome data at 6 and 12 weeks.

	Custom-fitted footwear group ($n = 17$)	Control group ($n = 16$)	Adjusted mean difference (95% CI) (footwear − control)	SMD[a] (95% CI)
Physical activity (6 weeks)				
Average vector magnitude per day (counts)	788 695 (232 774)	698 591 (259 432)	90 104 (−78 597 to 258 804)	0.37 (−0.32 to 1.06)
Average steps per day	7259 (1573)	6480 (1757)	779 (−368 to 1926)	0.47 (−0.22 to 1.16)
Average time spent in sedentary activities per day (min)	558 (79)	620 (163)	−62 (−149 to 25)	−0.49 (−1.18 to 0.20)
Average time spent in light activities per day (min)	64 (21)	57 (25)	7 (−10 to 23)	0.31 (−0.38 to 0.99)
Average time spent in MVPA[b] per day (min)	103 (43)	83 (43)	20 (−9 to 50)	0.48 (−0.21 to 1.17)
OxAFQ-C[c] (6 weeks)				
Physical domain (%)	78 (18)	77 (15)	0.2 (−9 to 10)	0.07 (−0.61 to 0.76)
School and play (%)	85 (18)	88 (15)	−2 (−12 to 8)	−0.22 (−0.90 to 0.47)
Emotional (%)	94 (6)	96 (7)	−1 (−5 to 3)	−0.22 (−0.91 to 0.46)
Concern (%)	76 (32)	74 (31)	8 (−8 to 25)	0.08 (−0.60 to 0.76)
Physical activity (12 weeks)				
Average vector magnitude (counts)	728 079 (218 028)	737 483 (266 748)	−9404 (−174 777 to 155 970)	−0.04 (−0.72 to 0.64)
Average steps per day	7119 (2094)	6981 (1847)	138 (−1254 to 1530)	0.07 (−0.61 to 0.75)
Average time spent in sedentary activities per day (min)	567 (59)	572 (65)	−5 (−48 to 37)	−0.08 (−0.76 to 0.60)
Average time spent in light activities per day (min)	60 (21)	64 (26)	−5 (−20 to 11)	−0.17 (−0.85 to 0.51)
Average time spent in MVPA per day (min)	93 (42)	84 (39)	8 (−21 to 37)	0.20 (−0.48 to 0.89)
OxAFQ-C (12 weeks)				
Physical domain (%)	80 (19)	75 (19)	4 (−8 to 15)	0.24 (−0.45 to 0.92)
School and play (%)	89 (14)	89 (13)	1 (−7 to 10)	−0.01 (−0.69 to 0.68)
Emotional (%)	98 (5)	96 (8)	3 (−2 to 7)	0.27 (−0.42 to 0.95)
Concern (%)	74 (29)	65 (34)	12 (−7 to 32)	0.27 (−0.42 to 0.95)

Values are mean (SD) unless otherwise stated.
[a]Standardised mean differences are based on mean data at the specified time point.
[b]Moderate to vigorous physical activity.
[c]Oxford Ankle Foot Questionnaire for Children – higher domain scores indicate better health for the respective domain.

Table 3. Footwear fit for participants' usual and custom-fitted footwear.

	Usual casual shoe		Usual school shoe		Clarks casual shoe		Clarks school shoe	
	Custom-fitted footwear group ($n = 17$)	Control group ($n = 16$)	Custom-fitted footwear group ($n = 17$)	Control group ($n = 16$)	Custom-fitted footwear group ($n = 17$)	Control group ($n = 16$)	Custom-fitted footwear group ($n = 17$)	Control group ($n = 16$)
Length assessment								
Too short, *n*	0	2	0	1	0	0	1	0
Too long, *n*	6	5	8	6	10	7	6	8
Acceptable, *n*	11	9	9	8	7	9	10	8
Mean difference (SD) between shoe length and foot length (mm)	16.9 (8.0)	15.6 (12.4)	18.2 (8.3)	16.4 (10.5)	20.9 (5.0)	18.31 (8.3)	17.7 (8.8)	18.4 (8.2)
Width assessment								
Too narrow, *n*	13	13	14	13	16	16	16	14
Too wide, *n*	0	0	0	0	0	0	0	0
Acceptable, *n*	4	3	3	3	1	1	1	2
Mean difference (SD) between shoe width and foot width (mm)	−5.82 (8.8)	−6.25 (6.9)	−6.6 (7.4)	−7.6 (7.4)	−6.9 (6.9)	−8.5 (8.3)	−7.1 (7.4)	−8 (8.4)
Acceptable fit (length and width), n (%)	2 (12)	0 (0)	0 (0)	0 (0)	0 (0)	0 (0)	1 (10)	0 (0)

Footwear fit defined as follows: acceptable shoe length = difference in length between shoe and foot was 0–20 mm; too short = difference in length between shoe and foot was <0 mm; too long = difference in length between shoe and foot was >20 mm. Acceptable shoe width = difference in width between shoe and foot was 0–10 mm; too narrow = difference in width between shoe and foot was <0 mm; too wide = difference in width between shoe and foot was >10 mm.

Discussion

Our findings demonstrate the feasibility of several aspects of the study. Participants found the shoes acceptable, they adhered to wearing the shoes, and only a small number of minor adverse events were reported. However, participant recruitment was slower than anticipated, the relatively high rate of use of co-interventions was not anticipated by the researchers, and the method used to fit the shoes (which is based on typically developing children) may not be suitable for children with Down syndrome.

Although meeting the eligibility criteria, many parents indicated long travel distance to the assessment location was a barrier to study participation. Parents reported this caused difficulty in coordinating appointment times, which may adversely affect their child's mood and subsequently their level of cooperation during assessments. Holidays and missing school to attend assessment sessions were additional barriers to participation, which delayed enrolment. Future trials would need to allow greater flexibility in assessment location and timing to reduce the travel burden on families. It is important to note that, once enrolled in the study, there was good participant retention, with only one participant lost to follow-up.

The use of co-interventions was higher than expected, which may affect the internal validity of a future definitive trial. The high rate of co-intervention use, particularly the purchase and use of new footwear by control group participants, may be explained by resentful demoralisation [39] as a result of being allocated into the less preferred group [40]. Parents were not instructed to modify shoe purchases and therefore were not aware of any potential

effects of co-interventions on the outcome measures or may have experienced issues such as requiring new footwear during the study. Prospectively requesting parents about refraining from using co-interventions may reduce their use during any future trials.

Another important finding of this study was that footwear fit was not ideal for the majority of participants in the custom-fitted footwear group. Although the shoes provided to participants were fitted according to the manufacturer's protocol, our estimation of fit demonstrated the footwear provided was excessively long and not wide enough for most participants. A likely reason for this is that the rounded toe box of the shoes appeared to not match the forefoot shape of children with Down syndrome, which is typically broad and relatively square [10]. These findings suggest that the manufacturer's shoe fitting protocol may not be suitable for children with Down syndrome. These issues need further consideration in the planning of any future trials.

Limited efficacy testing showed that there was a trend for increased physical activity in participants that received custom-fitted footwear. At 6 weeks, there was a 20 min per day difference between groups in moderate to vigorous physical activity in favour of the custom-fitted footwear. At 12 weeks, there was an 8 min per day difference in favour of the custom-fitted footwear. These effects were associated with trends for an increase in steps per day, reduced time spent in sedentary activity, as well as an increase in time spent in light activity, particularly at 6 weeks, in participants that received custom-fitted footwear. Statistically significant differences between the groups were not anticipated as this was a relatively small feasibility study. However, the differences in moderate to vigorous physical activity at 6 and 12 weeks equate to an increase in moderate to vigorous physical activity of 140 and 56 min per week, respectively. This is a potentially clinically worthwhile increase because it can assist children with Down syndrome meet physical activity guidelines, which suggest that children should engage in at least 60 min of moderate to vigorous physical activity per day [3]. There were no between-group differences, or trends for between group differences, for disability associated with foot and ankle problems (assessed using the OxAFQ-C) or gait parameters, so the inclusion of these measures in future trials may be unnecessary.

There are three major strengths to this pilot study. First, we evaluated several domains of an evidence-based framework to determine feasibility [19], which supports the primary aim of the study. Second, we used commercially available footwear and a recommended shoe fitting technique, which adds external validity to the findings, since this is what children with Down syndrome and their families usually experience in everyday life. Third, the study design aimed to minimise bias through design features and data analysis, such as using random sequence generation, concealed allocation of interventions, as well as assessor blinding. Further, there was minimal attrition and an intention-to-treat analysis was performed.

This study has five limitations that need to be acknowledged. First, we did not investigate all domains of feasibility (integration, expansion) because these domains were not relevant to the outcomes measured, so it is unclear what the feasibility of the intervention is in relation to these domains. Second, due to the use of commercially available footwear, there were only limited size combinations available (shoe length and widths), which constrained our ability to truly custom-fit the footwear to the dimensions of participants' feet. In most cases, the allocated footwear was too long and/or too narrow. Future trials would need to ensure the custom-fitted footwear being evaluated is appropriately fitting. Third, we did not exclude participants who were already wearing appropriately fitting footwear prior to enrolment, so a future trials could consider this an exclusion criteria. Fourth, we did not normalise gait parameters (stride length, step length and walking velocity) for leg length. This has been recommended when participants of different age and leg length are included in a study [41]. However, as shown in Table 1, the mean and standard deviations values for age and height suggest a similar distribution between the groups, hence the data were not normalised. Finally, it was not possible to blind participants, so the estimation of intervention effects may have been overinflated due to ascertainment bias [42]. This is an inherent problem faced when conducting trials using physical interventions [43].

Conclusions

This study has shown that a definitive randomised trial to evaluate the efficacy of custom-fitted shoes to increase physical activity in children with Down syndrome is feasible. However, future trials would need to ensure that the footwear intervention used fits participants' feet appropriately. This requires the custom-fitted footwear being evaluated to have a wider variety of fitting options than those used in this study. Additionally, greater flexibility with respect to location of assessments would improve recruitment rate in this population.

Acknowledgements

The authors thank Clarks® who provided the footwear as an in-kind donation and Down Syndrome Victoria who assisted in recruitment. The authors also thank Maria Auhl for assisting in data entry and Dr Stacey Cleary for assisting in coding the qualitative data. All authors have read and approved the final manuscript.

Author contributions

Conception and design: NS, SEM, NMH, NFT, AME, CMW, KBL, and AKB; Obtaining of funding: NS, SEM, AME and CMW; Collection and assembly of data: NMH, SEM, AKB, MA and SC; Analysis and interpretation of data: NMH, SEM, NS and KBL; Drafting of the article: NMH, SEM, NS, KBL, AKB, NFT, AME and CMW; Final approval of the article: NMH, NS, KBL, AKB, NFT, AME, CMW and SEM.

Disclosure statement

The authors declare that they have no competing interests. NMH is a recipient of an Australian Government Research Training Scholarship 2015. HBM is currently an NHMRC Senior Research Fellow (ID 1135995). CMW is currently an NHMRC Early Career Health Professional Research Fellow (ID APP1126960).

Funding

This work was supported by the La Trobe University Living with Disability Research Centre and La Trobe University Sport, Exercise and Rehabilitation Research Focus Area.

ORCID

Nirmeen M. Hassan http://orcid.org/0000-0003-3472-8502
Nora Shields http://orcid.org/0000-0002-6840-2378
Karl B. Landorf http://orcid.org/0000-0002-3882-5206

Andrew K. Buldt http://orcid.org/0000-0002-2020-2780
Cylie M. Williams http://orcid.org/0000-0002-0223-9141
Hylton B. Menz http://orcid.org/0000-0002-2045-3846
Shannon E. Munteanu http://orcid.org/0000-0001-6780-2743

Data availability statement

Data available on request from the authors.

References

[1] King G, Law M, King S, et al. A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities. Phys Occup Ther Pediatr. 2003;23:63–90.
[2] Rimmer JH, Riley B, Wang E, et al. Physical activity participation among persons with disabilities: barriers and facilitators. Am J Prev Med. 2004;26:419–425.
[3] The Department of Health. Australia's physical activity and sedentary behaviour guidelines. The Department of Health; 2014. Available from: http://www.health.gov.au/internet/main/publishing.nsf/content/health-pubhlth-strateg-phys-act-guidelines#apa512
[4] Whitt-Glover MC, O'Neill KL, Stettler N. Physical activity patterns in children with and without Down syndrome. Pediatr Rehabil. 2006;9:158–164.
[5] Barr M, Shields N. Identifying the barriers and facilitators to participation in physical activity for children with Down syndrome. J Intellect Disabil Res. 2011;55:1020–1033.
[6] Roizen NJ, Patterson D. Down's syndrome. Lancet. 2003;361:1281–1289.
[7] Horvat M, Pitetti KH, Croce R. Isokinetic torque, average power, and flexion/extension ratios in nondisabled adults and adults with mental retardation. J Orthop Sports Phys Ther. 1997;25:395–399.
[8] Pikora TJ, Bourke J, Bathgate K, et al. Health conditions and their impact among adolescents and young adults with Down syndrome. PLoS One. 2014;9:e96868.
[9] Mahy J, Shields N, Taylor NF, et al. Identifying facilitators and barriers to physical activity for adults with Down syndrome. J Intellect Disabil Res. 2010;54:795–805.
[10] Perotti LR, Abousamra O, Del Pilar Duque Orozco M, et al. Foot and ankle deformities in children with Down syndrome. J Child Orthop. 2018;12:218–226.
[11] Caird MS, Wills BP, Dormans JP. Down syndrome in children: the role of the orthopaedic surgeon. J Am Acad Orthop Surg. 2006;14:610–619.
[12] Concolino D, Pasquzzi A, Capalbo G, et al. Early detection of podiatric anomalies in children with Down syndrome. Acta Paediatr. 2006;95:17–20.
[13] Galli M, Cimolin V, Pau M, et al. Relationship between flat foot condition and gait pattern alterations in children with Down syndrome. J Intellect Disabil Res. 2014;58:269–276.
[14] Pau M, Galli M, Crivellini M, et al. Foot-ground interaction during upright standing in children with Down syndrome. Res Dev Disabil. 2012;33:1881–1887.
[15] Prasher VP, Robinson L, Krishnan VH, et al. Podiatric disorders among children with Down syndrome and learning disability. Dev Med Child Neurol. 2008;37:131–134.
[16] Lim PQ, Shields N, Nikolopoulos N, et al. The association of foot structure and footwear fit with disability in children and adolescents with Down syndrome. J Foot Ankle Res. 2015;8:4.
[17] Shields N, Lim P, Wollersheim D, et al. Do foot posture, deformity, and footwear fit influence physical activity levels in children with Down syndrome? A prospective cohort study. J Intellect Dev Disabil. 2017;42:332–338.
[18] Eldridge SM, Chan CL, Campbell MJ, et al. CONSORT 2010 statement: extension to randomised pilot and feasibility trials. BMJ. 2016;355:i5239.
[19] Bowen DJ, Kreuter M, Spring B, et al. How we design feasibility studies. Am J Prev Med. 2009;36:452–457.
[20] Cain KL, Sallis JF, Conway TL, et al. Using accelerometers in youth physical activity studies: a review of methods. J Phys Act Health. 2013;10:437–450.
[21] McCormick CJ, Bonanno DR, Landorf KB. The effect of customised and sham foot orthoses on plantar pressures. J Foot Ankle Res. 2013;6:19.
[22] Warne JP, Warrington GD. Four-week habituation to simulated barefoot running improves running economy when compared with shod running. Scand J Med Sci Sports. 2014;24:563–568.
[23] Menz HB, Auhl M, Tan JM, et al. Effectiveness of foot orthoses versus rocker-sole footwear for first metatarsophalangeal joint osteoarthritis: randomized trial. Arthritis Care Res (Hoboken). 2016;68:581–589.
[24] Menz HB, Auhl M, Ristevski S, et al. Effectiveness of off-the-shelf, extra-depth footwear in reducing foot pain in older people: a randomized controlled trial. J Gerontol A Biol Sci Med Sci. 2015;70:511–517.
[25] Munteanu SE, Scott LA, Bonanno DR, et al. Effectiveness of customised foot orthoses for Achilles tendinopathy: a randomised controlled trial. Br J Sports Med. 2015;49:989–994.
[26] O'Neil ME, Fragala-Pinkham MA, Forman JL, et al. Measuring reliability and validity of the ActiGraph GT3X accelerometer for children with cerebral palsy: a feasibility study. J Pediatr Rehabil Med. 2014;7:233–240.
[27] Pulakka A, Cheung YB, Ashorn U, et al. Feasibility and validity of the ActiGraph GT3X accelerometer in measuring physical activity of Malawian toddlers. Acta Paediatr. 2013;102:1192–1198.
[28] Choi L, Liu Z, Matthews CE, et al. Validation of accelerometer wear and nonwear time classification algorithm. Med Sci Sports Exerc. 2011;43:357–364.
[29] McGarty AM, Penpraze V, Melville CA. Calibration and cross-validation of the ActiGraph wGT3X + accelerometer for the estimation of physical activity intensity in children with intellectual disabilities. PLoS One. 2016;11:e0164928.
[30] Morris C, Doll HA, Wainwright A, et al. The Oxford ankle foot questionnaire for children: scaling, reliability and validity. J Bone Joint Surg Br. 2008;90:1451–1456.
[31] Menz HB, Latt MD, Tiedemann A, et al. Reliability of the GAITRite walkway system for the quantification of temporo-spatial parameters of gait in young and older people. Gait Posture. 2004;20:20–25.
[32] van Uden CJ, Besser MP. Test–retest reliability of temporal and spatial gait characteristics measured with an instrumented walkway system (GAITRite®). BMC Muskuloskelet Disord. 2004;5:13.
[33] Byun S, Han JW, Kim TH, et al. Test–retest reliability and concurrent validity of a single tri-axial accelerometer-based gait analysis in older adults with normal cognition. PLoS One. 2016;11:e0158956.

[34] Menz HB, Morris ME. Footwear characteristics and foot problems in older people. Gerontology. 2005;51:346–351.
[35] Altman DG. Missing outcomes in randomized trials: addressing the dilemma. Open Med. 2009;3:e51–e53.
[36] Sterne JAC, White IR, Carlin JB, et al. Multiple imputation for missing data in epidemiological and clinical research: potential and pitfalls. BMJ. 2009;338:b2393.
[37] Peat J, Barton B. Medical statistics: a guide to data analysis and critical appraisal. Oxford (UK): Blackwell Publishing; 2005.
[38] Twisk J, Proper K. Evaluation of the results of a randomized controlled trial: how to define changes between baseline and follow-up. J Clin Epidemiol. 2004;57:223–228.
[39] Onghena P. Resentful demoralization. In: Encyclopedia of statistics in behavioral science. 2nd ed. Hoboken (NJ): John Wiley and Sons; 2005.
[40] Preference Collaborative Review Group. Patients' preferences within randomised trials: systematic review and patient level meta-analysis. BMJ. 2008;337:a1864.
[41] Hof A. Scaling data to body size. Gait Posture. 1996;4:222–223.
[42] Schulz KF, Grimes DA. Blinding in randomised trials: hiding who got what. Lancet. 2002;359:696–700.
[43] Boutron I, Tubach F, Giraudeau B, et al. Blinding was judged more difficult to achieve and maintain in nonpharmacologic than pharmacologic trials. J Clin Epidemiol. 2004;57:543–550.

Chapter 4 – Reproducibility study

Preface

A randomised pilot study (Chapter 3) suggested a definitive randomised trial is feasible but highlighted that commercially available footwear – despite being custom-fitted – may not be able to accommodate the unique foot structure of children with Down syndrome.

To be able to supply appropriately fitting footwear to children with Down syndrome, a detailed understanding of the foot dimensions of children with Down syndrome is required. This requires reproducible methods of obtaining the foot dimensions of children with Down syndrome. 3D foot scanning technology has the potential to improve our knowledge on the foot dimensions of children with Down syndrome; however, its reproducibility has not been evaluated in children with Down syndrome. Therefore, the study presented in this chapter aims to address *Objective 3* of this thesis*,* to determine the reproducibility of measuring foot dimensions of children with Down syndrome using 3D foot scanning. The protocol for obtaining foot measurements from 3D foot scans described in this study was used in a subsequent study (Chapter 5).

This chapter has been published in the *Journal of Foot and Ankle Research* in 2020 with the associated online supplementary files presented in Appendix 3. The citations within this chapter relate to the reference list of the publication, not the reference list included at the end of this thesis. Following publication of this journal article, a correction (i.e. corrigendum) was published that detailed minor amendments to the wording of the article. The correction is included at the end of this chapter immediately after the journal article.

4.1 Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome

The reference for this publication is:

Hassan NM, Buldt AK, Shields N, Landorf KB, Menz HB, Munteanu SE. Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome. *Journal of Foot and Ankle Research.* 2020;13:31. DOI: https://doi.org/10.1186/s13047-020-00403-1

The reference for the correction is:

Hassan NM, Buldt AK, Shields N, Landorf KB, Menz HB, Munteanu SE. Correction to: Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome. *Journal of Foot and Ankle Research.* 2020;13:41. DOI: https://doi.org/10.1186/s13047-020-00409-9

Hassan *et al. Journal of Foot and Ankle Research* (2020) 13:31
https://doi.org/10.1186/s13047-020-00403-1

Journal of
Foot and Ankle Research

RESEARCH Open Access

Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome

Nirmeen M. Hassan[1,2*], Andrew K. Buldt[1,3], Nora Shields[2,3,4], Karl B. Landorf[1,2,3], Hylton B. Menz[1,3] and Shannon E. Munteanu[1,3]

Abstract

Background: Children and adolescents with Down syndrome have a distinctive foot shape (such as wide and flat feet) that often leads to difficulty with footwear fitting. 3-dimensional (3D) scanning can accurately measure the foot dimensions of individuals with Down syndrome, which may assist shoe fit. However, the reproducibility of measuring foot dimensions using 3D scans in children and adolescents with Down syndrome is unknown. The aim of this study was to determine the intra- and inter-rater reproducibility of measuring foot dimensions of children and adolescents with Down syndrome using 3D scanning.

Methods: 3D foot scans of 30 participants with Down syndrome aged 5 to 17 years were obtained using the FotoScan 3D scanner. Foot dimensions assessed were foot length, ball of foot length, outside ball of foot length, diagonal foot width, horizontal foot width, heel width, ball girth, instep girth, first and fifth toe height, and instep height. Additionally, the Wesjflog Index and forefoot shape were determined. Measurements were completed by two raters independently on two separate occasions, 2 weeks apart. Intra- and inter-rater reliability were assessed using intra-class coefficients (ICCs) and Gwet's AC1 statistics with 95% confidence intervals. Agreement was determined by calculating limits of agreement (LOA) and percentage agreement.

Results: Eighteen participants were female and 12 were male (mean age 10.6 [3.9] years). Intra-rater reproducibility (ICCs ranged from 0.74 to 0.99, 95% LOA from − 13.7 mm to 16.3 mm) and inter-rater reproducibility (ICCs ranging from 0.73 to 0.99, 95% LOA from − 18.8 mm to 12.7 mm) was good to excellent, although some measurements (ball of foot length, outside ball of foot length, heel width and girth measurements) displayed wider LOAs indicating relatively poorer agreement. Forefoot shape displayed substantial to almost perfect reliability (Gwet's AC1 0.68 to 0.85) and percentage agreement ranged from 73 to 87%, indicating acceptable agreement.

(Continued on next page)

* Correspondence: n.hassan@latrobe.edu.au
[1]Discipline of Podiatry, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia
[2]Living with Disability Research Centre, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia
Full list of author information is available at the end of the article

(Continued from previous page)
Conclusions: The measurement of specific foot dimensions of children and adolescents with Down syndrome using 3D scans is reproducible. Findings of this study may be used to support future research measuring specific foot dimensions of children and adolescents with Down syndrome using 3D foot scans.

Keywords: Foot, Shoes, Down syndrome, Child, Adolescent, Foot deformities, 3-D image, Reproducibility of results

Background

Down syndrome is the most common chromosomal disorder [1], occurring in 1 in every 650 to 1000 live births [2]. Down syndrome affects multiple body systems including the nervous, cardiovascular and the musculoskeletal systems [3], resulting in intellectual and physical disability. Individuals with Down syndrome can have reduced physical fitness [4], ligamentous laxity, hypotonia, reduced lower limb muscle strength [5], less functional gait patterns [6] and gait instability [7].

Children and adolescents with Down syndrome commonly experience conditions associated with the foot that may impact their physical function. A population-based study involving 197 young individuals with Down syndrome reported 63% of individuals with Down syndrome were affected by a musculoskeletal condition of the foot [8]. At present, the exact cause of many of these conditions is not clearly understood. However, a contributing factor may be the unique foot shape of this population leading to difficulties in finding appropriately-fitting footwear. Children and adolescents with Down syndrome often have a flatter, shorter and broader foot [9], and are more likely to have foot deformities that includes lesser toe deformities and hallux valgus [9]. These deviations in foot shape are likely to contribute to the number of children and adolescents with Down syndrome who wear poorly-fitting footwear; 60 to 88% of children and adolescents with Down syndrome [10, 11] compared to 16% of typically developing children [11]. Poorly-fitting footwear can have adverse outcomes including the development of foot pain, which may lead to impaired health-related quality of life and altered gait patterns [12, 13]. Additionally, poorly-fitting footwear may contribute to reduced physical activity in children and adolescents with Down syndrome [13]. This is an issue for children and adolescents with Down syndrome because they have low levels of physical activity and are at greater risk of developing chronic health conditions as a result. It may also contribute to their reduced social participation. Further, having an intellectual disability can be an additional complicating factor as children and adolescents with Down syndrome may not always verbally communicate pain experienced to their caregivers despite being more sensitive to pain [14].

Given the high prevalence of poorly-fitting footwear and the potentially detrimental effects on health, improving footwear-fit for children and adolescents with Down syndrome is important. Improving footwear fit may result in improved health-related quality of life and participation in physical activities. Additionally, improving the health of children and adolescents with Down syndrome may reduce burden to the health care system. Improving footwear fit can be achieved by designing footwear that can accommodate the unique foot shape of this population [12]. An essential initial step in designing footwear for children and adolescents with Down syndrome would be to capture the detailed foot dimensions of this population using reliable and valid methods. Although there is general consensus the foot shape of children and adolescents with Down syndrome differs to typically developing children, only one study [15] demonstrated the feet of young males with Down syndrome were shorter and narrower than age-matched peers, as measured using a podoscope. However, a limitation of this study is that only 2 foot dimensions (foot length and width) were measured using a 2-dimensional technique [8], which may not fully represent the complex shape of the foot. Three-dimensional (3D) scanning technology is a valid and reproducible means of obtaining detailed data on foot shape [16] and has been used to study the variations in foot shape in a number of different populations [17–19]. However, no studies have used 3D scanning to evaluate the foot dimensions of children and adolescents with Down syndrome, and the reproducibility of performing these measurements is unknown. Therefore, the aim of this study is to determine the reproducibility of measuring foot dimensions of children and adolescents with Down syndrome using 3D foot scanning.

Methods

This study is reported in accordance with Guidelines for Reporting Reliability and Agreement Studies [20].

Study design

Data were obtained from a previous feasibility study investigating the efficacy of custom-fitted footwear to increase physical activity levels in children and adolescents with Down syndrome [21]. The study was approved by the La Trobe University Human Ethics Committee (HEC16–027) and written informed consent was obtained from parents or guardians. Where appropriate,

children and adolescents also provided written assent for participation prior to enrolment [21].

Participants

Participants were children and adolescents aged 5 to 17 years with Down syndrome. Participants were excluded if they had any health condition that may affect physical activity (e.g. inflammatory arthritis, subluxation etc.) as reported by their parents, or required the use of an ambulatory device (e.g. cane, crutches or walker). Participants were recruited through a member-based disability organisation for individuals with Down syndrome that was based in the community [21].

Raters

Two raters (NMH and AKB) performed the measurements for all foot scans, and measurements were repeated twice within 4 weeks to assess intra- and inter-rater reproducibility. Both raters were registered podiatrists with four and 14 years of clinical experience, respectively. Rater 1 had 3 months and rater 2 had 5 years of experience using 3D scans to measure foot dimensions, respectively.

Measurements of participant characteristics

Participants had their height and weight measured to calculate body mass index (BMI). Measurements were for the right foot only. Foot posture was assessed using two indices; the Foot Posture Index [22] and the Arch Index [23]. For both indices, higher positive scores indicate a flatter foot posture. The presence of lesser digital deformities (i.e. hammer, mallet and claw toes) was documented [10, 24]. The presence and severity of hallux valgus deformity was assessed using the Manchester scale [25]. The degree of deformity was graded on a scale of 0 to 3 (no deformity, mild, moderate and severe). Scores for hallux valgus were dichotomised, where scores of 0 and 1 were graded as absent and scores of 2 and 3 were graded as present [26].

Scanning procedure

Participants stood in a relaxed, full weight-bearing position and a 3D scan was taken of their right foot using the FotoScan 3D scanner (Precision 3D, Weston-supermare, UK). The FotoScan 3D device uses a fixed system of cameras and projectors to obtain images of the foot, that are automatically converted into a 3D model [27]. According to the manufacturer, the scans obtained with this system are accurate to within less than half a millimetre. The 3D foot scans were then exported as stereolithography (STL) files (Fig. 1). The 3D-Tool© Version 13 (3D-Tool GmbH, Weinheim, Germany) was used to obtain all length, width and height measurements. For girth measurements, a cross-section of the foot at the relevant landmarks was created and exported as a drawing exchange format file. The perimeter of the cross-section was determined using Canvas© 11 software (ACD Systems International, Seattle, WA, USA).

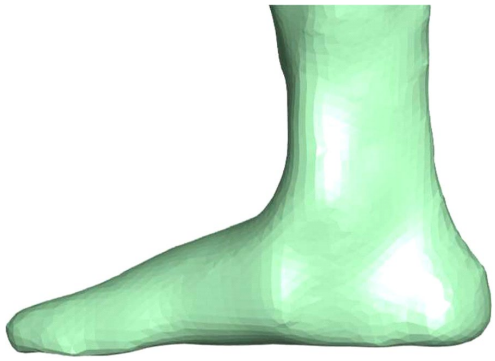


Fig. 1 3D model of right foot viewed from the medial aspect

Measurement of foot dimensions

Prior to data collection, pilot testing of the foot measurements was completed. A measurement technique protocol was developed (see Additional file 1). The protocol was piloted on the scans of five participants and the results were compared. Where there was substantial variability in results, the measurement technique was clarified between raters until consensus was reached. Each rater worked independently during data collection and compiled measurements on separate Excel spreadsheets (Microsoft® Office 365, Microsoft, Redmond, WA, USA) on two separate occasions that were two weeks apart.

We measured 13 foot dimensions that are relevant to footwear manufacturing [17, 28, 29]. These were (Figs. 2 and 3):

1. Foot length: distance between foot end (pternion) and foot tip (anterior point of most protruding toe).
2. Ball of foot length: distance between foot end (pternion) and the first metatarsophalangeal protrusion.
3. Outside ball of foot length: distance between foot end (pternion) and the fifth metatarsophalangeal protrusion.
4. Diagonal foot width: connecting line between the first metatarsophalangeal joint and the fifth metatarsophalangeal joint.
5. Horizontal (orthogonal) foot width: orthogonal connection line starting at the first metatarsophalangeal joint to the outside curvature of the foot.

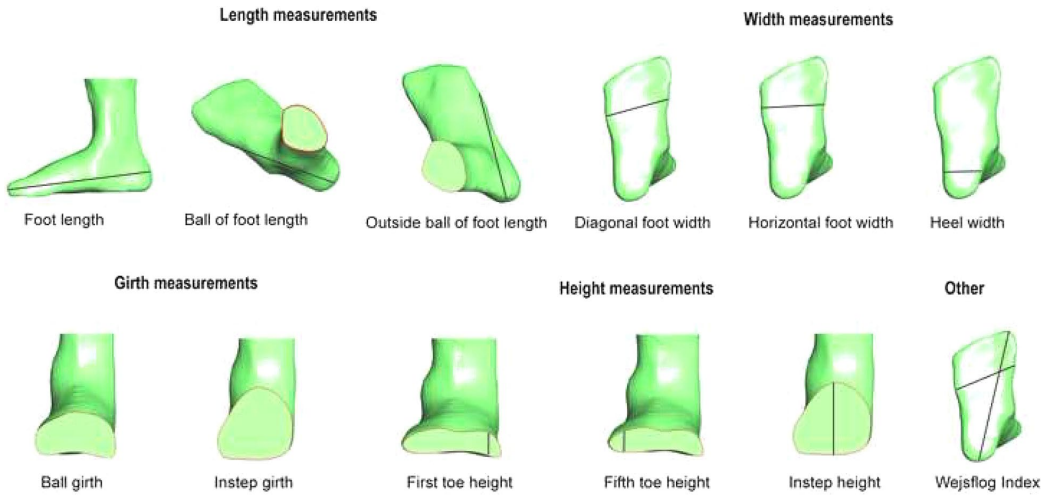


Fig. 2 Foot measurements

6. Heel width: maximum, orthogonal connection line starting at the medial side of the heel to the outside curvature of the heel.
7. Wejsflog Index: a ratio measurement of foot length to the diagonal forefoot width.
8. Ball girth: maximum circumference at the level of the first and the fifth metatarsophalangeal joint protrusion.
9. Instep girth: maximum circumference measured from the most plantar aspect of the foot to the most dorsal aspect of the foot, at the level of the navicular.
10. First toe height: maximum height of the hallux measured from the most plantar aspect of the hallux to the most dorsal aspect of the hallux.
11. Fifth toe height: maximum height of the fifth toe measured from the most plantar aspect of the fifth toe to the most dorsal aspect of the fifth toe.
12. Instep height: measured from the most plantar aspect of the foot to the most dorsal aspect of soft tissue (plantar foot end to the junction of shank and foot dorsum).
13. Forefoot shape: determined by categorising forefoot shape into three categories using the length of the digits. The categories were (i) first toe longest; (ii) second toe longest; and (iii) first and second toe length equal length.

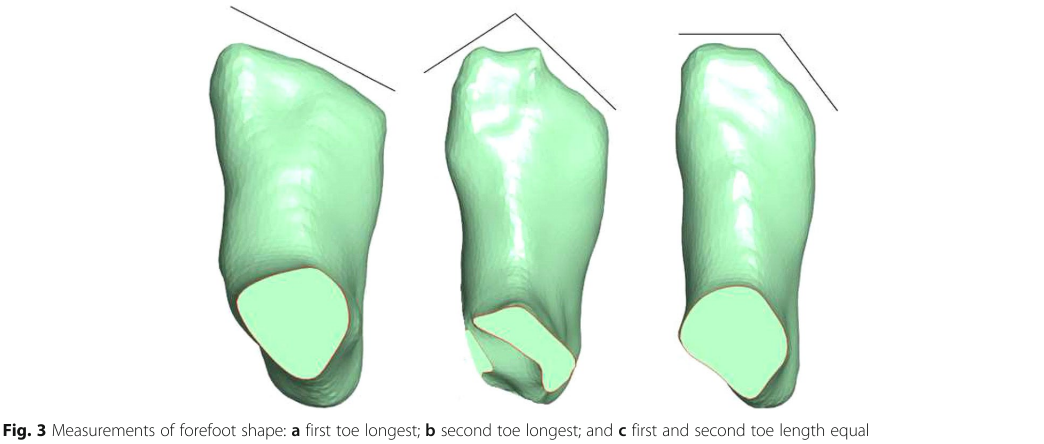


Fig. 3 Measurements of forefoot shape: **a** first toe longest; **b** second toe longest; and **c** first and second toe length equal

Statistical analysis

An a priori sample size estimation using the Power Analysis and Sample Size software (PASS 15 software, NCSS, LLC, Kaysville, UT, USA) for the ICC test, alpha of 0.05, power 0.80 and ICC of 0.60 determined that a minimum sample size of 19 participants (foot scans) was necessary. However, as we had access to foot scans of 30 children and adolescents with Down syndrome, all scans were measured. Analyses were performed using IBM SPSS Statistics version 25.0 (IBM Corp, NY, USA) and STATA SE Version 14.2 (StataCorp, College Station, TX, USA), using the *kappaetc* module. To satisfy the independence assumption of statistical analysis, measurements from the right foot only were analysed [30]. For continuous data, data were assessed for normality using skewness and kurtosis tests, and data were found to be normally distributed. For continuous data, reliability was calculated using ICCs with 95% confidence intervals (ICC [1, 2], consistency) [31]. Interpretation of ICCs were based on definitions provided by Portney and Watkins [32], where ICC values less than 0.5 was considered as poor, values between 0.5 to 0.75 was considered as moderate, values between 0.75 to 0.90 was considered as good and any value above 0.90 was considered as excellent reliability. For nominal data, reliability was determined by calculating Gwet's AC1 statistic [33] and results were interpreted according to Landis and Koch cut-offs, which are: less than zero was considered poor, 0 to 0.20 slight, 0.21 to 0.40 fair, 0.40 to 0.60 moderate, 0.60 to 0.80 substantial, 0.81 to 1.00 almost perfect reliability [34]. For continuous data, agreement was determined by calculating limits of agreement (LOA) [35]. For the calculation of LOAs, the presence of heteroscedasticity was assessed, and when present, ratio LOAs were calculated by taking the antilog of the calculated LOA values [36]. For nominal data, agreement was determined by using percentage agreement.

Results

Table 1 summarises the participant characteristics. There were 18 females and 12 males, with a mean (SD) age of 10.6 (3.9) years. The mean Foot Posture Index was + 9 (1.8) and the mean Arch Index was 0.30 (0.05), indicating that on average, participants had a flat foot type. Hallux valgus was present in 4 participants (13%), and 14 participants (46%) had some degree of deformity of one of the lesser toes. No participants were excluded on the basis of requiring an ambulatory device.

Intra-rater reproducibility

Table 2 summarises the intra-rater reproducibility of all 13 foot measurements. The intra-rater reliability of rater 1 was good to excellent for 11 out of 13 measurements, with ICCs ranging from 0.76 to 0.99. Fifth toe height and forefoot shape measurements had moderate reliability only (ICC = 0.74, Gwet's AC1 = 0.68 respectively). For rater 2, intra-rater reliability was good to excellent for all 13 measurements (ICCs/Gwet's AC1 ≥ 0.83).

There were 6 measurements (foot length, diagonal foot width, Wejsflog Index, first toe height, fifth toe height and instep height) for both raters and 1 measurement for Rater 2 (horizontal foot width) that exhibited narrow LOAs ranging from − 5.7 mm to 8.1 mm. However, 5 measurements (ball of foot length, outside ball of foot length, heel width, ball girth, and instep girth) demonstrated wider LOAs ranging from − 13.5 mm to 16.3 mm. For forefoot shape, agreement ranged from 73 to 87%.

Table 1 Participant characteristics – values are mean (SD) unless otherwise stated

Characteristic	Mean (SD)
Age, years	10.6 (3.9)
Females/males, n	18/12
Height (m)	1.30 (0.19)
Weight (kg)	40.1 (21.4)
BMI[1] (kg/m^2)	21.9 (6.7)
Type of Down syndrome, n (Trisomy 21 / Translocation)	27/3
Level of intellectual disability (unclear/mild/moderate/severe)	9/12/9/0
Presence of hallux valgus, n (%)	4 (13.0)
Presence of digital deformity, n (%)	14 (46.0)
Foot Posture Index[2]	8.8 (1.8)
Arch Index[3]	0.30 (0.05)

[1]Body mass index; [2]Foot Posture Index; [3]Arch Index. The Foot Posture Index scores range from −12 to + 12. Scores less than 1 indicate a supinated foot posture, scores between 1 and 7 indicate a normal foot posture, and scores greater than 7 indicate a pronated foot posture. Arch Index represents the ratio of the area of the middle third of a footprint to the entire footprint area, excluding the digits. Normal: 0.21 to 0.28, high: < 0.21 and low: > 0.28

Table 2 Intra-rater reproducibility of foot measurements

	Rater 1				Rater 2			
	Trial 1	Trial 2			Trial 1	Trial 2		
Foot measurement	**Mean (SD)**	**Mean (SD)**	**ICC (95% CI) [Interpretation]**	**95% LOA[1]**	**Mean (SD)**	**Mean (SD)**	**ICC (95% CI) [Interpretation]**	**95% LOA[1]**
Foot length	193.3 (27.9)	192.7 (28.1)	0.99 (0.99 to 1.00) [Excellent]	−2.0 to 3.2	193.2 (27.6)	192.5 (28.0)	0.99 (0.99 to 1.00) [Excellent]	−1.6 to 3.0
Ball of foot length	146.0 (21.1)	145.6 (22.2)	0.97 (0.94 to 0.99) [Excellent]	−9.7 to 10.6	147.3 (21.0)	149.2 (21.9)	0.98 (0.97 to 0.99) [Excellent]	−8.7 to 4.8
Outside ball of foot length	127.5 (18.4)	124.9 (17.7)	0.97 (0.94 to 0.99) [Excellent]	−5.7 to 10.7	130.5 (15.8)	129.3 (18.0)	0.89 (0.80 to 0.95) [Good]	−13.7 to 16.3
Diagonal foot width	79.4 (13.1)	79.0 (13.0)	0.99 (0.98 to 1.00) [Excellent]	−3.2 to 3.9	78.4 (12.0)	78.4 (12.3)	0.98 (0.98 to 0.99) [Excellent]	−3.8 to 3.7
Horizontal foot width	77.3 (12.7)	77.1 (12.2)	0.97 (0.94 to 0.99) [Excellent]	−5.8 to 6.1 / 0.934 to 1.06[2]	76.8 (12.0)	76.9 (11.7)	0.98 (0.98 to 1.00) [Excellent]	−3.6 to 3.3
Heel width	48.4 (8.5)	49.4 (7.9)	0.76 (0.60 to 0.90) [Good]	−12.1 to 10.1	47.1 (8.6)	45.5 (7.6)	0.83 (0.70 to 0.91) [Good]	−7.7 to 10.8 / 0.86 to 1.23[2]
Wejsflog Index	2.5 (0.2)	2.5 (0.2)	0.96 (0.92 to 0.98) [Excellent]	−0.1 to 0.1	2.5 (0.21)	2.5 (0.21)	0.95 (0.90 to 0.98) [Excellent]	−0.1 to 0.1
Ball girth	190.3 (29.1)	190.6 (31.5)	0.98 (0.96 to 0.99) [Excellent]	−11.4 to 10.7	189.4 (29.6)	189.2 (30.7)	0.98 (0.97 to 0.99) [Excellent]	−9.8 to 10.3
Instep girth	202.5 (26.5)	204.4 (27.5)	0.97 (0.95 to 0.99) [Excellent]	−13.5 to 3.7	200.0 (27.5)	198.7 (26.9)	0.99 (0.98 to 1.00) [Excellent]	−5.8 to 8.3
First toe height	20.1 (4.5)	20.7 (3.6)	0.88 (0.80 to 0.94) [Good]	−4.3 to 3.3	20.7 (4.1)	21.2 (3.8)	0.83 (0.70 to 0.92) [Good]	−4.9 to 3.9
Fifth toe height[3]	16.93 (2.6)	17.6 (2.7)	0.74 (0.52 to 0.87) [Moderate]	−4.3 to 3.0	17.5 (2.8)	17.49 (2.6)	0.84 (0.70 to 0.92) [Good]	−2.9 to 2.9
Instep height	57.1 (7.7)	54.4 (8.0)	0.93 (0.90 to 0.97) [Excellent]	2.8 to 8.1	53.6 (7.5)	55.3 (6.7)	0.95 (0.91 to 0.98) [Excellent]	−5.7 to 2.5 / 0.89 to 1.05[2]
Forefoot shape	–	–	0.68 (0.45 to 0.90)[4] [Substantial]	73 (60 to 90)[5]	–	–	0.85 (0.70 to 1.0)[4] [Almost perfect]	87 (70 to 100)[5]

[1]Limits of agreement. [2]Ratio limits of agreement also presented as measurement displays heteroscedasticity. [3]29 scans were used due to the presence of artefacts. [4]Gwet's AC1. [5]Percentage agreement. Foot dimensions are measured in millimetres

Inter-rater reproducibility

Table 3 summarises the inter-rater reproducibility of all 13 foot measurements. The inter-rater reliability was good to excellent for all 11 measurements, with ICCs ranging from 0.89 to 0.99. Fifth toe height and forefoot shape measurements had moderate reliability only (ICC = 0.73 and Gwet's AC1 = 0.77, respectively).

Six measurements (foot length, diagonal foot width, horizontal foot width, Wejsflog Index, first toe height and fifth toe height) demonstrated narrow LOAs, ranging from − 4.4 mm to 5.8 mm. However, the remaining 5 measurements (ball of foot length, outside ball of foot length, ball girth, instep girth and instep height) displayed relatively wider LOAs ranging from − 18.8 mm to 12.7 mm. Percentage agreement for forefoot shape was 80%.

Discussion

We found that the foot dimensions of children and adolescents with Down syndrome can be measured reliably from 3D foot scans. Our results indicate moderate to excellent reliability for all foot dimension measurements as demonstrated by high inter-rater ICC values or Gwet's AC1 values. However, the measurement of fifth toe height displayed poorer reliability. We observed some measurements (foot length, diagonal foot width, horizontal foot width, Wejsflog Index, first toe height and fifth toe height) had narrow LOAs indicating good agreement between raters, whereas others (ball of foot length, outside ball of foot length, ball girth, instep girth and instep height) displayed wider LOAs, suggesting relatively poorer agreement.

We observed differences in the performance of the two raters. The reliability for rater 1 was poorer than rater 2 for the measurements of heel width, fifth toe height and forefoot shape. The reason for this is unclear but could be that these measurements are more challenging to measure due to difficulty in locating the boundaries of these regions, particularly the toe region [16]. It is also possible that varying experience of the raters influenced the findings, since the reproducibility of rater 2, who had 5 years of experience, was greater than rater 1, who had 3 months' experience. This speculation is supported by previous

Table 3 Inter-rater reproducibility of foot measurements

Foot measurement	Rater 1 Mean (SD)	Rater 2 Mean (SD)	ICC (95% CI) [Interpretation]	95% LOA[1]
Foot length	193.3 (27.9)	193.2 (27.6)	0.99 (0.99 to 1.00) [Excellent]	−2.8 to 2.9 / 0.99 to 1.01[2]
Ball of foot length	146.0 (21.1)	147.3 (21.0)	0.97 (0.94 to 1.00) [Excellent]	−10.8 to 8.3
Outside ball of foot length	127.5 (18.4)	130.5 (15.8)	0.89 (0.80 to 0.95) [Good]	−18.8 to 12.7
Diagonal foot width	79.4 (13.1)	78.4 (12.0)	0.98 (0.96 to 0.99) [Excellent]	−3.8 to 5.8
Horizontal foot width	77.3 (12.7)	76.8 (12.0)	0.98 (0.96 to 0.99) [Excellent]	−4.3 to 5.4
Heel width	48.4 (8.5)	47.1 (8.6)	0.91 (0.81 to 0.95) [Excellent]	−5.9 to 8.6 / 0.89 to 1.19[2]
Wejsflog Index	2.4 (0.2)	2.5 (0.2)	0.90 (0.82 to 0.96) [Good]	−0.2 to 0.2
Ball girth	190.3 (29.1)	189.4 (29.6)	0.99 (0.97 to 0.99) [Excellent]	−8.5 to 10.2
Instep girth	202.5 (26.5)	200.0 (27.5)	0.98 (0.97 to 0.99) [Excellent]	−7.3 to 12.2
First toe height	20.1 (4.5)	20.7 (4.1)	0.92 (0.84 to 0.96) [Excellent]	−3.9 to 2.9
Fifth toe height[3]	16.9 (2.6)	17.5 (2.8)	0.73 (0.50 to 0.86) [Moderate]	−4.4 to 3.3
Instep height	57.1 (7.7)	53.6 (7.4)	0.90 (0.81 to 0.95) [Good]	−3.0 to 9.9
Forefoot shape	–	–	0.77 (0.58 to 0.96)[4] [Substantial]	80 (65 to 95)[5]

[1]Limits of agreement. [2]Ratio limits of agreement also presented as measurement displays heteroscedasticity. [3]29 scans were used due to the presence of artefacts. [4]Gwet's AC1. [5]Percentage agreement. Foot dimensions are measured in millimetres

work that has shown that rater experience and training is an important consideration in the reliability of measuring foot dimensions, particularly when it involves manual allocation of landmarks for calculating dimensions [16].

As this is the first study to investigate the reproducibility of the measurement of foot dimensions of children and adolescents using 3D scans, it is not possible to directly compare our findings. However, our findings are in general agreement with studies that investigated the reliability of measuring foot dimensions using adult foot scans where ICCs ranged from 0.82 to 0.99 [16, 27, 37–40].

Our findings relating to agreement allows for the interpretation of the acceptability of the reproducibility of the measurements. Where the LOA value is less than the minimally important difference for a measurement, the agreement could be considered acceptable. However, the value for the minimally important difference may vary depending on the context of the measurement. For example, for footwear fitting, it is common practice that foot length and foot width are measured in order to select an appropriately sized shoe. The International Organization for Standardization (2015) for footwear reports whole sizes (US and UK) differ in length by 8 mm [41], which indicates the reproducibility of the measurement of foot length is likely to be acceptable, as the range for the inter-rater LOA value for this measurement was 3.9 mm. That is, it is less than the value that would necessitate a difference in shoe size (8 mm). In contrast, the acceptability of the measurement of horizontal foot width is questionable, as the range of the inter-rater LOA value was 9.7 mm, which exceeds the standard 4.8 mm that defines a difference in shoe width [42]. The acceptability of the reproducibility of the remaining measurements (if used to guide the manufacture of footwear) for children and adolescents with Down syndrome is unclear, as shoe last dimensions for different shoe sizes is commercially sensitive information and not readily available.

Footwear-fit can be difficult in children and adolescents with Down syndrome because of variations to the shape of their feet. This study has shown measuring the foot dimensions from 3D foot scans of children and adolescents with Down syndrome can be done reproducibly. As they are reproducible, these measurements can be used with confidence in several settings. In a clinical setting, clinicians could perform measurements of foot dimensions to monitor and inform parents of their child's foot shape, and to provide guidance on appropriate footwear selection. In an applied research setting, the measurements may be used to determine detailed differences in foot dimensions of children and adolescents with Down syndrome, which may then be considered when manufacturing footwear for this population.

Our findings need to be interpreted in the context of the strengths and limitations of this study. We used two raters who worked independently throughout the data collection process in order to reduce the risk of bias, and we assessed both intra- and inter-rater reproducibility for completeness of results. In addition, both raters were podiatrists, who were experienced in foot and ankle measurements. However, the generalisability of the findings to raters from other professional backgrounds and/or with different experience in measuring 3D foot scans needs further investigation. Finally, the measurements in this study were manually calculated according to definitions that were outlined in our methods, so findings may differ if dimensions are calculated by scanning software with pre-populated definitions of measurements that are different to the ones that we used.

Conclusions

The measurement of specific foot dimensions of children and adolescents with Down syndrome using 3D scans is reproducible. The measurement described has multiple applications. In a clinical setting, clinicians can perform measurements of foot dimensions to monitor and inform parents of their child's foot shape, and to provide guidance on appropriate footwear selection. In an applied research setting, the measurements may be used to determine detailed differences in foot dimensions of children and adolescents with Down syndrome, which may then be considered when manufacturing footwear for this population.

Supplementary information

Supplementary information accompanies this paper at https://doi.org/10.1186/s13047-020-00403-1.

Additional file 1. 3D foot scan measurement protocol

Abbreviations

2D: 2-dimensional; 3D: 3-dimensional; BMI: Body mass index; ICC: Intra-class coefficient; LOA: Limits of agreement

Acknowledgements

We acknowledge all participants and their parents for being involved in the original feasibility study in which this data was derived from. NMH is a recipient of an Australian Government Research Training Scholarship 2015. This work was supported by the La Trobe University Living with Disability Research Centre, as well as the La Trobe University Sport, Exercise and Rehabilitation Research Focus Area.

Authors' contributions

NMH, SEM, KBL and NS conceived the idea and design of the study. Collection and assembly of data was completed by NMH and AKB. Data analysis and interpretation was completed by NMH, SEM, KBL, HBM and NS. The manuscript was drafted by NMH, SEM, KBL, HBM and NS. All authors contributed substantially to the review and editing of the final manuscript. All author(s) read and approved the final manuscript.

Funding

NMH is a recipient of an Australian Government Research Training Scholarship 2015. This work was supported by the La Trobe University Living with Disability Research Centre.

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author upon reasonable request.

Ethics approval and consent to participate

Ethical approval was obtained from the La Trobe University Human Ethics Committee (ethics approval number HEC16–027).

Consent for publication

Consent for publication was obtained from parents of participants. All authors have read and approved the final manuscript.

Competing interests

The authors declare that they have no competing interests.

Author details

[1]Discipline of Podiatry, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia. [2]Living with Disability Research Centre, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia. [3]La Trobe Sport and Exercise Medicine Research Centre, School of Allied Health, Human Services and Sport, Victoria 3086, Australia. [4]Discipline of Physiotherapy, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia.

Received: 2 March 2020 Accepted: 26 May 2020
Published online: 04 June 2020

References

1. Oliver TR, Feingold E, Yu K, Cheung V, Tinker S, Yadav-Shah M, et al. New insights into human nondisjunction of chromosome 21 in oocytes. PLoS Genet. 2008;4(3):e1000033.
2. Bittles AH, Glasson EJ. Clinical, social, and ethical implications of changing life expectancy in Down syndrome. Dev Med Child Neurol. 2004;46(4):282–6.
3. Bianca S. Non congenital heart disease aspects of Down's syndrome. Images Paediatr Cardiol. 2002;4(4):3–11.
4. Mendonca GV, Pereira FD, Fernhall B. Reduced exercise capacity in persons with Down syndrome: cause, effect, and management. Ther Clin Risk Manag. 2010;6:601–10.
5. Foley C, MacDermott E, Killeen O. Musculoskeletal anomalies in a national cohort of children and adolescents with trisomy 21. Pediatr Rheumatol Online J. 2014;12:P160.
6. Galli M, Rigoldi C, Brunner R, Virji-Babul N, Giorgio A. Joint stiffness and gait pattern evaluation in children with Down syndrome. Gait Posture. 2008;28(3):502–6.
7. Cimolin V, Galli M, Grugni G, Vismara L, Albertini G, Rigoldi C, et al. Gait patterns in Prader-Willi and Down syndrome patients. J Neuroeng Rehabil. 2010;7:28.
8. Pikora TJ, Bourke J, Bathgate K, Foley K-R, Lennox N, Leonard H. Health conditions and their impact among adolescents and Young adults with Down syndrome. PLoS One. 2014;9(5):e96868.
9. Concolino D, Pasquzzi A, Capalbo G, Sinopoli S, Strisciuglio P. Early detection of podiatric anomalies in children with Down syndrome. Acta Paediatr. 2006;95(1):17–20.
10. Lim PQ, Shields N, Nikolopoulos N, Barrett J, Evans A, Taylor N, et al. The association of foot structure and footwear fit with disability in children and adolescents with Down syndrome. J Foot Ankle Res. 2015;8:4.
11. Calvo-Lobo C, Ramos Garcia A, Losa Iglesias ME, Lopez-Lopez D, Rodriguez-Sanz D, Romero-Morales C, et al. The relationship between shoe fitting and foot health of persons with Down syndrome: a case control study. Int J Environ Res Public Health. 2018;15(5):983.
12. Buldt AK, Menz HB. Incorrectly fitted footwear, foot pain and foot disorders: a systematic search and narrative review of the literature. J Foot Ankle Res. 2018;11:43.
13. Shields N, Lim P, Wollersheim D, Nikolopoulos N, Barrett J, Evans A, et al. Do foot posture, deformity, and footwear fit influence physical activity levels in children with Down syndrome? A prospective cohort study. J Intellect Disabil Res. 2017;42(4):332–8.
14. McGuire BE, Defrin R. Pain perception in people with Down syndrome: a synthesis of clinical and experimental research. Front Behav Neurosci. 2015;9:194.
15. Puszczałowska-Lizis E, Nowak K, Omorczyk J, Ambroży T, Bujas P, Nosiadek L. Foot structure in boys with Down syndrome. Biomed Res Int. 2017;7047468.
16. Lee Y-C, Lin G, Wang M-JJ. Comparing 3D foot scanning with conventional measurement methods. J Foot Ankle Res. 2014;7:44.
17. Krauss I, Grau S, Mauch M, Maiwald C, Horstmann T. Sex-related differences in foot shape. Ergonomics. 2008;51(11):1693–709.
18. Lee YC, Kouchi M, Mochimaru M, Wang MJ. Comparing 3D foot shape models between Taiwanese and Japanese females. J Hum Ergol (Tokyo). 2015;44(1):11–20.
19. Wunderlich RE, Cavanagh PR. Gender differences in adult foot shape: implications for shoe design. Med Sci Sports Exerc. 2001;33(4):605–11.
20. Kottner J, Audigé L, Brorson S, Donner A, Gajewski BJ, Hróbjartsson A, et al. Guidelines for reporting reliability and agreement studies (GRRAS) were proposed. J Clin Epidemiol. 2011;64(1):96–106.
21. Hassan NM, Shields N, Landorf KB, Buldt AK, Taylor NF, Evans AM, et al. Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study. Disabil Rehabil. 2019:1–10 [Epub ahead of print].

22. Redmond AC, Crosbie J, Ouvrier RA. Development and validation of a novel rating system for scoring standing foot posture: The Foot Posture Index. Clin Biomech (Bristol, Avon). 2006;21(1):89–98.
23. Cavanagh PR, Rodgers MM. The arch index: a useful measure from footprints. J Biomech. 1987;20(5):547–51.
24. Menz HB, Lord SR. Foot pain impairs balance and functional ability in community-dwelling older people. J Am Podiatr Med Assoc. 2001;91(5):222–9.
25. Garrow AP, Papageorgiou A, Silman AJ, Thomas E, Jayson MI, Macfarlane GJ. The grading of hallux valgus. The Manchester scale. J Am Podiatr Med Assoc. 2001;91(2):74–8.
26. Menz HB, Munteanu SE. Radiographic validation of the Manchester scale for the classification of hallux valgus deformity. Rheumatology (Oxford). 2005; 44(8):1061–6.
27. Menz HB, Auhl M, Ristevski S, Frescos N, Munteanu SE. Evaluation of the accuracy of shoe fitting in older people using three-dimensional foot scanning. J Foot Ankle Res. 2014;7:3.
28. Goonetilleke R. The science of footwear. 1st ed ed. Boca Raton: CRC Press; 2013.
29. Lee YC, Wang MJ. Taiwanese adult foot shape classification using 3D scanning data. Ergonomics. 2015;58(3):513–23.
30. Menz HB. Two feet, or one person? Problems associated with statistical analysis of paired data in foot and ankle medicine. Foot (Edinb). 2004;14(1):2–5.
31. Koo TK, Li MY. A guideline of selecting and reporting Intraclass correlation coefficients for reliability research. J Chiropr Med. 2016;15(2):155–63.
32. Portney L, Watkins M. Foundations of clinical research. Applications to practice. 2 ed. Upper Saddle River, NJ. : Prentice Hall Health; 2000.
33. Wongpakaran N, Wongpakaran T, Wedding D, Gwet KL. A comparison of Cohen's kappa and Gwet's AC1 when calculating inter-rater reliability coefficients: a study conducted with personality disorder samples. BMC Med Res Methodol. 2013;13:61.
34. Landis JR, Koch GG. The measurement of observer agreement for categorical data. Biometrics. 1977;33(1):159–74.
35. Bland JM, Altman DG. Measuring agreement in method comparison studies. Stat Methods Med Res. 1999;8(2):135–60.
36. Nevill A, Atkinson G. Assessing agreement between measurements recorded on a ratio scale in sports medicine and sports science. Br J Sports Med. 1997;31:314–8.
37. De Mits S, Coorevits P, De Clercq D, Elewaut D, Woodburn J, Roosen P. Validity and reliability of the Infoot 3D foot digitizer for rheumatoid arthritis patients. J Am Podiatr Med Assoc. 2009;101(3):198–207.
38. Robinette KM, Daanen HA. Precision of the CAESAR scan-extracted measurements. Appl Ergon. 2006;37(3):259–65.
39. Seminati E, Canepa Talamas D, Young M, Twiste M, Dhokia V, Bilzon JLJ. Validity and reliability of a novel 3D scanner for assessment of the shape and volume of amputees' residual limb models. PLoS One. 2017;12(9): e0184498.
40. De Mits S, Coorevits P, De Clercq D, Elewaut D, Woodburn J, Roosen P. Reliability and validity of the Infoot 3D foot digitizer for normal healthy adults. Footwear Sci. 2010;2(2):65–75.
41. International Organization for Standardization. ISO/TS 19407. Footwear — Sizing — Conversion of sizing systems. 2015.
42. Brannock Device. 2019. [Available from: http://www.brannock.com].

Publisher's Note

Journal of
Foot and Ankle Research

CORRECTION Open Access

Correction to: Reproducibility offoot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome

Nirmeen M. Hassan[1,2*], Andrew K. Buldt[1,3], Nora Shields[2,3,4], Karl B. Landorf[2,3], Hylton B. Menz[1,3] and Shannon E. Munteanu[1,3]

Correction to: J Foot Ankle Res 13, 31 (2020)
https://doi.org/10.1186/s13047-020-00403-1

Following publication of the original article [1], we have been notified of the below corrections:

1. On Page 1, Abstract, 'Methods' section, line 6: 'intra-class coefficients' should read 'intra-class correlation coefficients'. This error is also present on page 8 in the 'Abbreviations' section.
2. In Table 1 on page 5, within the final sentence of the notes section at the bottom of the table: the cut-off values used for the Arch Index 'Normal: 0.21 to 0.28, high: < 0.21 and low: > 0.28' should read 'Normal: 0.21 to 0.26, high: < 0.21 and low: > 0.26 [23]'

Author details
[1]Discipline of Podiatry, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia. [2]Living with Disability Research Centre, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia. [3]La Trobe Sport and Exercise Medicine Research Centre, School of Allied Health, Human Services and Sport, Victoria 3086, Australia. [4]Discipline of Physiotherapy, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia.

Published online: 02 July 2020

Reference

1. Hassan, et al. Reproducibility offoot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome. J Foot Ankle Res. 2020;13:31.https://doi.org/10.1186/s13047-020-00403-1

The original article can be found online at https://doi.org/10.1186/s13047-020-00403-1

* Correspondence: n.hassan@latrobe.edu.au
[1]Discipline of Podiatry, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia
[2]Living with Disability Research Centre, School of Allied Health, Human Services and Sport, La Trobe University, Victoria 3086, Australia
Full list of author information is available at the end of the article

Chapter 5 – Cross-sectional observational study

Preface

Chapter 4 highlighted the measurement of foot dimensions using 3D foot scans was reproducible. Subsequently, this measurement was used for the study described in this chapter.

The study presented in this chapter is a cross-sectional observational study that aimed to address *Objective 5* of this thesis, which was to determine differences in foot dimensions of children with and without Down syndrome using 3D foot scans. This study was submitted to *Disability and Rehabilitation* in June 2020 and is under review. The study is presented in this chapter as it was submitted to *Disability and Rehabilitation* (i.e. formatted for this journal), and the citations within this chapter relate to the reference list of the manuscript, not the reference list included at the end of this thesis. Likewise, the figures and tables presented in this chapter relate to this chapter and are not part of the numbering system used in the rest of this thesis.

5.1 Differences in foot dimensions between children and adolescents with and without Down syndrome

Abstract

Children with Down syndrome frequently wear poorly-fitting footwear, and this has been attributed to their unique foot shape. This study compared the differences in foot dimensions obtained from three-dimensional (3D) foot scans between 51 children with Down syndrome (mean age 10.5 years; 28 female) and an age and sex-matched cohort of 51 typically developing children. Twelve-foot dimensions were measured. Absolute and normalised (for height or foot length) measurements were compared between groups. Absolute differences suggest children with Down syndrome have smaller feet than typically developing children. When normalised for height, foot length remained shorter in children with Down syndrome. When normalised for foot length, ball of foot length, foot width, girth and fifth toe height were significantly greater in children with Down syndrome. This indicates substantial variations in foot shape of children with Down syndrome and should be considered when fitting and manufacturing footwear for this group.

Key words

Foot anthropometry, Shoes, Down syndrome, Child, Adolescent, Foot deformities; 3-D image

Practitioner summary

Children with Down syndrome wear poorly-fitting footwear due to their unique foot shape. The 3D foot dimensions were compared between children with and without Down syndrome. There are substantial variations in foot shape in children with Down syndrome which should be considered when fitting and manufacturing footwear for this population.

Background

Children with Down syndrome often do not wear appropriately fitting footwear. One study involving 50 children with Down syndrome found 60% of participants wore footwear that was too narrow and 54% of participants wore footwear that was too long [1]. Another study with 105 participants (50 with Down syndrome) found only 12% of individuals with Down syndrome (which included adolescents and adults) wore appropriately fitting footwear, compared with 84% of individuals without Down syndrome [2]. Poorly-fitting footwear may lead to inefficient gait, and this can be further compounded in those with Down syndrome who also have hypotonia and reduced muscle strength [3]. Further, poorly-fitting footwear is a recognised contributor to foot pain and deformity (i.e. hallux valgus and lesser toe deformity) [4].

The characteristic foot shape of children with Down syndrome is speculated to be a factor in contributing to their increased prevalence of poorly-fitting footwear. Four studies comparing the foot shape of children with and without Down syndrome have demonstrated that pes planus foot type, hallux valgus deformity and lesser digital deformities are more common in children with Down syndrome [2, 5-7]. Only one study [8] has compared the foot dimensions of children with and without Down syndrome. Puszczałowska-Lizis et al. [8] demonstrated the feet of males with Down syndrome aged 14 to 15 years were shorter and narrower than age-matched peers, as measured using a podoscope. However, a limitation of this study is that only boys of a limited age range were included (14 to 15), so the generalisibilty of these findings to females with Down syndrome or across a broader age range is limited. Further, only two foot dimensions were measured (foot length and width) using a 2-dimensional technique [8], which may not fully represent the complex shape of the foot.

Three-dimensional (3D) foot scanning is an accurate and reliable approach for capturing the dimensions of the foot [9]. Several studies have used 3D foot scanning to identify variations in foot structure between populations, as population-specific foot dimension data is important for footwear design and manufacture [10-12]. However, to our knowledge, no reported study has used 3D foot scanning technology to study the foot dimensions of children with Down syndrome. Therefore, the aim of this study was to compare the 3D foot dimensions in children with Down syndrome to typically developing children.

Methods

This study is reported in accordance with the Strengthening the Reporting of Observational Studies in Epidemiology Statement (STROBE) [13].

Study design

This was a cross-sectional, observational study comparing children with Down syndrome (which included adolescents) to typically developing children, who were matched according to age (±2 years) and sex.

Ethics approval

Ethics approval was obtained from the La Trobe University Human Ethics Committee (HEC13-035, HEC16-027, HEC19-290). Prior to participation, written informed consent was obtained from parents. Where possible, the children who participated were also invited to provide written assent.

Participant recruitment and eligibility

Participant data came from three studies. Data for children with Down syndrome was obtained from two previous studies in which 3D foot scans were obtained. The first study was a cross-sectional study investigating the association of foot structure and footwear-fit with foot-specific disability in children with Down syndrome [1]. The second study was a randomised pilot study investigating the efficacy of custom-fitted footwear to increase physical activity levels in children with Down syndrome [14]. Both studies shared similar recruitment methods and eligibility criteria [1, 14]. Participants were recruited through a community, member-based disability organisation (Down Syndrome Victoria). Participants were excluded if they had a medical condition that may have affected physical activity levels; had lower limb surgery in the previous 12 months; had significant joint laxity that may result in subluxation; or required the use of an ambulatory device (such as a cane or walker).

Typically developing children were recruited in a third study from the community using promotional flyers. Eligibility criteria were the same as for the two studies involving children with Down syndrome outlined above, with the exception that these children were typically developing. The typically developing children were matched to the children with Down syndrome according to age (±2 years) and sex. Recruitment occurred over a period of five months (August to December 2019). All data were collected at La Trobe University, Melbourne campus.

Data collected

Participant characteristics

Age (in years), sex, height (in metres), weight (in kilograms) and body mass index (BMI) were documented for all participants.

Foot posture

Foot Posture Index classified foot type into normal, supinated foot or pronated foot types. This is a six-criterion tool used to classify foot posture according to the alignment of the

rearfoot and forefoot. Scores range from -12 to +12, and normal foot posture in children lies within the range of +2 to +9. Scores greater than +9 indicate a pes planus (i.e. low-arched) foot posture, while scores less than +2 indicate a pes cavus (i.e. high-arched) foot posture [15]. Previous research has shown the Foot Posture Index is a reliable and valid measurement tool [16].

Foot scanning process

A FotoScan 3D scanner (Precision 3D, Weston-super-mare, UK) was used to obtain 3D foot scans to measure the foot dimensions of children with Down syndrome (n = 51). According to the manufacturer, the scanned model has an accuracy to within 0.5 mm. Standardised verbal instructions and a demonstration of the scanning process was provided to each child prior to the scan. The same order of assessments was performed for each child. Timing of 3D scanning was not standardised per day due to the need to accommodate participant availability, which varied between children. Both scanners were calibrated where required and used in accordance to the manufacturer's guidelines. Participants placed their right foot on the scanner and stood in a relaxed, bipedal stance. The FotoScan 3D device uses a fixed system of high resolution cameras and projectors to obtain images of the foot, which are then automatically converted into a 3D model [17]. At the time of data collection for typically developing children, an unforeseen technical issue occurred with the FotoScan 3D scanner that resulted in an alternative foot scanner (INFOOT 3D scanner) being used (n = 51). The INFOOT scanner is an optical laser scanning system that captures a 3D image of the foot, with the same accuracy as the previous scanner (0.5 mm). The scanning protocol was the same across both scanners. A stereolithography (STL) file was created from both foot scanners (Figure 1). The 3D-Tool© Version 13 (3D-Tool GmbH, Weinheim, Germany) software was used to calculate all length, width and height measurements. Although the software of the INFOOT scanner is capable of automatically calculating a range of foot dimensions based on identified landmarks, we used the same software (3D-Tool© Version 13) for all STL files when measuring foot dimensions to maintain consistency.

Measurement of foot dimensions

All dimensions were measured by the same researcher (NMH). The following dimensions were assessed: foot length, ball of foot length, outside ball of foot length, diagonal foot width, horizontal foot width, heel width, ball girth, instep girth, first toe height, fifth toe height, instep height and forefoot shape [18, 19]. Each foot measurement is defined in Table 1 and illustrated in Figure 2. The intra- and inter-rater reliability of these measurements is moderate to excellent (ICC ranging from 0.70 to 0.90) [17, 20]. These measurements have been used previously in other anthropometric studies of the feet and are used in footwear design and manufacture [19].

Sample size and statistical analysis

No formal sample size calculation was performed. The sample size (n = 51 per group) was based on available data and was considered feasible [21]. All analyses were performed using IBM SPSS Statistics version 25.0 (IBM Corp, NY, USA). Data were assessed for normality (using skewness and kurtosis tests) and were found to be normally distributed. Only measurements of the right foot were analysed so that the independence assumption of statistical analysis was fulfilled [22]. Independent *t*-tests were performed for continuous variables and chi-square analyses were performed for nominal data (e.g. forefoot shape). Differences in foot dimensions were compared using two approaches. First, we analysed differences between groups using the absolute value of measurements. Second, to account for differences in foot sizes between participants (e.g. due to age differences), we analysed differences in foot dimensions normalised for foot length [23]. Foot length was normalised for differences in height. Where significant differences were identified, Cohen's *d* effect sizes and 95% confidence intervals were calculated to obtain a measure of the magnitude of differences. Cohen's *d* values were classified as small (0.20), moderate (0.50) and large (>0.80) [24].

Results

Participant characteristics

One hundred and two participants took part, with 51 participants (28 females and 23 males) in each group. The groups of children with Down syndrome and typically developing children were of similar mean age (Table 2) and both groups had a mean BMI within the normal range. Children with Down syndrome had a more pronated foot type, as indicated by a higher mean Foot Posture Index score than the typically developing children (Table 2).

Differences between groups in absolute foot dimensions

Table 3 shows the absolute differences in foot dimensions between groups. Differences between the groups were found for 10 of the 12 variables measured. Children with Down syndrome had a shorter foot length and also a shorter outside ball of foot length compared to typically developing children. Children with Down syndrome also had narrower foot width (diagonal foot width and horizontal foot width), narrower heel width and smaller girth measurements (ball girth and instep girth) compared to typically developing children. In children with Down syndrome, first toe height and instep height were lower than for typically developing children. There were no differences in ball of foot length and fifth toe height measurements between the groups.

Differences between groups in normalised foot dimensions

Seven out of 11 normalised foot measurements were significantly different between groups (Table 4). Children with Down syndrome had a shorter foot length, but a longer ball of foot length compared to typically developing children. Foot width (diagonal foot width and horizontal foot width) was greater in children with Down syndrome as was foot girth (ball girth and instep girth) compared to typically developing children. Lastly, fifth toe height was greater in children with Down syndrome. There were no significant differences between groups for outside ball of foot length, heel width, first toe height and instep height.

Discussion

Our findings show there are substantial differences in the foot shape of children with Down syndrome compared to typically developing children. Children with Down syndrome have smaller foot dimensions compared to their peers. However, when we adjusted our analyses to normalise for differences in height, we found children with Down syndrome had a relatively shorter foot length, and when normalised for foot length, a longer ball of foot length, wider forefoot width, larger girth measurements and a greater fifth toe height. The effect sizes for these differences were all moderate to large.

These findings are novel as this is the first study to comprehensively measure foot dimensions of children with Down syndrome using 3D foot scans. The absolute differences in foot dimensions found between groups (i.e. not adjusted for foot length) are in agreement with a previous study using a podoscope that reported boys with Down syndrome had shorter and narrower feet as compared to their peers [8]. The normalised results are also in agreement with other descriptive studies that report the feet of children with Down syndrome are wide [1, 7] and short [25]. Our findings add to the existing literature by expanding the number of foot dimensions of children with Down syndrome investigated.

Normalised differences in foot dimensions may explain why poorly-fitting footwear is relatively common among individuals with Down syndrome [1, 14, 25, 26]. Children with Down syndrome are more likely to wear footwear that is too long or too narrow in an attempt to accommodate their unique foot dimensions [1, 2, 25, 26]. Commercially available footwear is manufactured from shoe lasts made to fit the foot of individuals without Down syndrome. The dimensions of commercially available footwear are unable to accommodate the usual foot dimensions of an individual with Down syndrome which is shorter and wider, with increased girth and of greater height of the lesser toes (as measured by the fifth toe).

The implications from this study are that children with Down syndrome require footwear that is wider and deeper than commercially available footwear, to accommodate feet that are wider and more voluminous. Current footwear design has led to a challenging situation for parents of children with Down syndrome, which is to identify commercially-available footwear that satisfies this need, as custom-made footwear is unlikely to be a feasible option. A better solution would be that future footwear should account for differences in foot dimensions of children with Down syndrome. In addition to designing shoes that fit correctly, other issues need to be considered, including understanding the experiences and barriers to appropriate footwear selection of care givers, education of caregivers and footwear suppliers, and the development of a greater range of shapes of commercially available footwear that are aesthetically pleasing and affordable.

Our findings must be considered in the context of its limitations. First, the study did not stratify the analyses across specific age ranges or sex as it was not powered to do so. Future studies could explore if there are differences across specific age ranges or between sexes. Second, the assessor who measured all foot dimensions was not blinded to the groups when measuring foot dimensions, which may have introduced assessment bias, although we attempted to minimise this with a strict measurement protocol and assessor training. Lastly, we experienced a technical issue with the scanners used, which required half the sample to be measured using a different scanner. However, we do not believe this would have affected the results because both scanners create the same file output (STL files), all scans were measured using the same protocol and technique, and both scanners are accurate to 0.5 mm.

Conclusion

There are substantial variations in the 3D foot shape of children with Down syndrome. These findings should be considered when fitting and manufacturing footwear for children with Down syndrome.

Acknowledgements

We acknowledge all participants and their parents for being involved in the studies in which this data was derived from. NMH is a recipient of an Australian Government Research Training Scholarship 2015. This work was supported by the La Trobe University Living with Disability Research Centre, as well as the La Trobe University Sport, Exercise and Rehabilitation Research Focus Area.

Declarations

Conflict of interest

The authors declare that they have no competing interests.

Ethical approval and consent to participate

Ethical approval was obtained from the La Trobe University Human Ethics Committee (ethics approval number HEC13-035, HEC16-027 and HEC19-290).

Consent for publication

All authors have read and approved the final manuscript.

Availability of data and material

The datasets used and/or analysed during the current study are available from the corresponding author upon reasonable request.

Funding

NMH is a recipient of an Australian Government Research Training Scholarship 2015. This work was supported by the La Trobe University Living with Disability Research Centre.

References

1. Lim, Shields N, Nikolopoulos N, Barrett J, Evans A, Taylor N, et al. The association of foot structure and footwear fit with disability in children and adolescents with Down syndrome. J Foot Ankle Res. 2015;8:4.

2. Calvo-Lobo C, Ramos Garcia A, Losa Iglesias ME, Lopez-Lopez D, Rodriguez-Sanz D, Romero-Morales C, et al. The Relationship between Shoe Fitting and Foot Health of Persons with Down Syndrome: A Case Control Study. Int J Environ Res Public Health. 2018;15(5):983.

3. Roizen N, Patterson D. Down's syndrome. Lancet. 2003;361(9365):1281-9.

4. Buldt AK, Menz HB. Incorrectly fitted footwear, foot pain and foot disorders: a systematic search and narrative review of the literature. J Foot Ankle Res. 2018;11:43.

5. Prasher VP, Robinson L, Krishnan VH, Chung MC. Podiatric disorders among children with Down syndrome and learning disability. Dev Med Child Neurol. 1995;37(2):131-4.

6. Mansour E, Yaacoub JJ, Bakouny Z, Assi A, Ghanem I. A podoscopic and descriptive study of foot deformities in patients with Down syndrome. Orthop Traumatol Surg Res. 2017;103(1):123-7.

7. Concolino D, Pasquzzi A, Capalbo G, Sinopoli S, Strisciuglio P. Early detection of podiatric anomalies in children with Down syndrome. Acta Paediatr. 2006;95(1):17-20.

8. Puszczałowska-Lizis E, Nowak K, Omorczyk J, Ambroży T, Bujas P, Nosiadek L. Foot Structure in Boys with Down Syndrome. Biomed Res Int. 2017;7047468.

9. Lee Y-C, Lin G, Wang M-JJ. Comparing 3D foot scanning with conventional measurement methods. J Foot Ankle Res. 2014;7:44.

10. Lee YC, Kouchi M, Mochimaru M, Wang MJ. Comparing 3D foot shape models between Taiwanese and Japanese females. J Hum Ergol (Tokyo). 2015;44(1):11-20.

11. Lee YC, Wang MJ. Taiwanese adult foot shape classification using 3D scanning data. Ergonomics. 2015;58(3):513-23.

12. Saghazadeh M, Kitano N, Okura TJJoF, Research A. Gender differences of foot characteristics in older Japanese adults using a 3D foot scanner. J Foot Ankle Res. 2015;8:29.

13. von Elm E, Altman DG, Egger M, Pocock SJ, Gotzsche PC, Vandenbroucke JP. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. J Clin Epidemiol. 2008;61(4):344-9.

14. Hassan NM, Shields N, Landorf KB, Buldt AK, Taylor NF, Evans AM, et al. Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study. Disabil Rehabil. 2019:1-10. [Epub ahead of print].

15. Redmond AC, Crosbie J, Ouvrier RA. Development and validation of a novel rating system for scoring standing foot posture: The Foot Posture Index. Clin Biomech (Bristol, Avon). 2006;21(1):89-98.

16. Morrison SC, Ferrari J. Inter-rater reliability of the Foot Posture Index (FPI-6) in the assessment of the paediatric foot. J Foot Ankle Res. 2009;2:26.

17. Menz HB, Auhl M, Ristevski S, Frescos N, Munteanu SE. Evaluation of the accuracy of shoe fitting in older people using three-dimensional foot scanning. J Foot Ankle Res. 2014;7:3.

18. Goonetilleke R. The Science of Footwear. 1st ed ed. Boca Raton: CRC Press; 2013.

19. Lee Y-C, Wang M-J. Taiwanese adult foot shape classification using 3D scanning data. Ergonomics. 2015;58(3):513-23.

20. Hassan NM, Buldt AK, Shields N, Landorf KB, Menz HB, Munteanu SE. Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome. Manuscript submitted for publication. 2020:[Epub ahead of print].

21. Bacchetti P. Current sample size conventions: flaws, harms, and alternatives. BMC Med. 2010;8:17.

22. Menz HB. Two feet, or one person? Problems associated with statistical analysis of paired data in foot and ankle medicine. Foot (Edinb). 2004;14(1):2-5.

23. Mickle KJ, Munro BJ, Lord SR, Menz HB, Steele JR. Foot shape of older people: implications for shoe design. Footwear Sci. 2010;2(3):131-9.

24. Cohen J. Statistical Power Analysis for the Behavioral Sciences. 2nd ed. Hillsdale, NJ: Erlbaum1988.

25. Jenkins DW, Cooper K, O'Connor R, Watanabe L. Foot-to-shoe mismatch and rates of referral in Special Olympics athletes. J Am Podiatr Med Assoc. 2012;102(3):187-97.

26. Shields N, Lim P, Wollersheim D, Nikolopoulos N, Barrett J, Evans A, et al. Do foot posture, deformity, and footwear fit influence physical activity levels in children with Down syndrome? A prospective cohort study. J Intellect Disabil Res. 2017;42(4):332-8.

Tables

Table 1. Definitions of the foot measurements.

Length	
Foot length	Distance between foot end (pternion) and foot tip (anterior point of most protruding toe).
Ball of foot length	distance between foot end (pternion) and the first metatarsophalangeal protrusion.
Outside ball of foot length	Distance between foot end (pternion) and the fifth metatarsophalangeal protrusion.
Width	
Diagonal foot width	Connecting line between the first metatarsophalangeal joint and the fifth metatarsophalangeal joint.
Horizontal foot width	Orthogonal connection line starting at the first metatarsophalangeal joint to the outside curvature of the foot.
Heel width	Maximum orthogonal connection line starting at the medial side of the heel to the outside curvature of the heel.
Girth	
Ball girth	Maximum circumference at the level of the first and the fifth metatarsophalangeal joint protrusion.
Instep girth	Maximum circumference measured from the most plantar aspect of the foot to the most dorsal aspect of the foot, at the level of the navicular.
Height	
First toe height	Maximum height of the hallux measured from the most plantar aspect of the hallux to the most dorsal aspect of the hallux.
Fifth toe height	Maximum height of the fifth toe measured from the most plantar aspect of the fifth toe to the most dorsal aspect of the fifth toe.
Instep height	Measured from the most plantar aspect of the foot to the most dorsal aspect of soft tissue.
Forefoot shape	Three categories: (i) first toe longest, (ii) second toe longest, and (iii) first and second toe equal in length.

Table 2. Participant characteristics. Values are mean (SD) unless otherwise noted.

	Children with Down syndrome	**Typically developing children**
Age, years	10.5 (3.7)	10.8 (3.7)
Age, n (%)		
5 to 10 years	27 (53)	24 (47)
11 to 15 years	17 (33)	21 (41)
16 to 19 years	7 (14)	6 (12)
Females/males, n	28/23	28/23
Height (m)	1.3 (0.2)	1.4 (0.2)
Weight (kg)	40.4 (21.4)	43.2 (18.5)
BMI[1] (kg/m^2)	22.1 (6.7)	20.1 (4.5)
Type of Down syndrome, n (Trisomy 21/Translocation/Mosaic)	44/6/1	N/A
Foot Posture Index, right	8.8 (2.1)	5.4 (2.2)
FPI -12 to +1, n (%)	0 (0)	2 (4)
FPI +2 to +9, n (%)	31 (61)	48 (94)
FPI +10 to +12, n (%)	20 (39)	1 (2)

[1]Body mass index. The Foot Posture Index scores range from -12 to +12. For children, normal foot posture in children lies within the range of +2 to +9. Scores greater than +9 indicate a pes planus foot posture, while scores less than +2 indicate a pes cavus foot posture.

Table 3. Comparison of absolute differences in foot dimensions. Values are the mean (SD) and measured in mm unless indicated otherwise.

Foot measurement	Children with Down syndrome	Typically developing children	Mean difference (95% CI)	Cohen's *d* (95% CI, size of effect)
Foot length	193.0 (26.6)	223.2 (28.4)	-30.1 (-40.9 to -19.3)	1.1 (0.7 to 1.5, large effect)
Ball of foot length	146.5 (20.6)	166.1 (21.1)	-19.7 (-27.9 to 11.5)	1.0 (0.6 to 1.4, large effect)
Outside ball of foot length	125.8 (17.2)	147.0 (17.6)	-21.2 (-28.1 to -14.4)	1.2 (0.8 to 1.6, large effect)
Diagonal foot width	78.2 (11.7)	86.5 (11.2)	-8.3 (-12.8 to -3.8)	0.7 (0.3 to 1.1, moderate effect)
Horizontal foot width	76.3 (10.9)	83.1 (10.3)	-6.8 (-11.0 to -2.6)	0.7 (0.3 to 1.1, moderate effect)
Heel width	49.1 (7.6)	56.1 (6.9)	-6.9 (-9.8 to -4.1)	1.0 (0.6 to 1.4, large effect)
Ball girth	187.3 (26.1)	205.4 (24.9)	-18.0 (-28.0 to -8.0)	0.7 (0.3 to 1.1, moderate effect)
Instep girth	201.3 (25.2)	224.0 (26.6)	-23.3 (-33.5 to -13.1)	0.9 (0.5 to 1.3, large effect)
First toe height	19.5 (3.6)	21.9 (2.8)	-2.4 (-3.7 to -1.1)	0.8 (0.4 to 1.2, large effect)
Fifth toe height	16.2 (2.9)	15.5 (2.4)	0.7 (-0.3 to 1.7)	0.3 (-0.1 to 0.7, small effect)
Instep height	54.9 (7.5)	64.5 (7.4)	-9.6 (-12.5 to -6.7)	1.3 (0.9 to 1.7, large effect)
Forefoot shape			1.58[1] (p = 0.483)[2]	
1st toe longest, n (%)	42 (82)	46 (90)		
2nd toe longest, n (%)	3 (5)	1 (2)		
1st and 2nd toe equal length, n (%)	6 (12)	4 (8)		

[1]Pearson's chi-square statistical test value. [2]*P*-value.

Table 4. Comparison of normalised differences in foot dimensions. Values are the mean (SD) and measured in mm unless indicated otherwise.

Foot measurement	Children with Down syndrome	Typically developing children	Mean difference (95% CI)	Cohen's *d* (95% CI, size of effect)
Foot length[1]	202.3 (10.0)	213.9 (7.3)	-11.6 (-15.1 to -8.2)	1.2 (0.8 to 1.6, large effect)
Ball of foot length	157.8 (4.9)	155.0 (3.7)	2.9 (1.2 to 4.6)	0.7 (0.3 to 1.1, moderate effect)
Outside ball of foot length	135.8 (6.3)	137.4 (6.3)	-1.6 (-4.1 to 0.9)	0.3 (-0.1 to 0.7, small effect)
Diagonal foot width	84.4 (6.2)	80.8 (3.8)	3.7 (1.6 to 5.7)	0.7 (0.3 to 1.1, moderate effect)
Horizontal foot width	82.4 (5.8)	77.6 (3.7)	4.8 (2.9 to 6.7)	1.0 (0.6 to 1.4, large effect)
Heel width	53.1 (5.5)	52.4 (3.5)	0.7 (-1.1 to 2.5)	0.2 (-0.2 to 0.6, small effect)
Ball girth	202.5 (14.2)	191.9 (8.8)	10.6 (5.9 to 15.3)	0.9 (0.5 to 1.3, large effect)
Instep girth	217.9 (14.7)	209.9 (9.7)	7.9 (3.0 to 12.8)	0.7 (0.3 to 1.1, moderate effect)
First toe height	21.0 (2.6)	20.5 (2.2)	0.5 (-0.5 to 1.4)	0.2 (-0.2 to 0.6, small effect)
Fifth toe height	17.6 (3.0)	14.5 (1.4)	3.1 (2.2 to 4.1)	1.3 (0.9 to 1.7, large effect)
Instep height	59.5 (6.1)	60.4 (4.1)	-0.9 (-2.9 to 1.1)	0.2 (-0.1 to 0.6, small effect)
Forefoot shape			1.58[2] (0.483)[3]	
1st toe longest, n (%)	42 (82)	46 (90)		
2nd toe longest, n (%)	3 (5)	1 (2)		
1st and 2nd toe equal length, n (%)	6 (12)	4 (8)		

[1]Foot length was normalised to height while remaining dimensions were normalised to foot length. [2]Pearson's chi-square statistical test value. [3]*P*-value

Figures

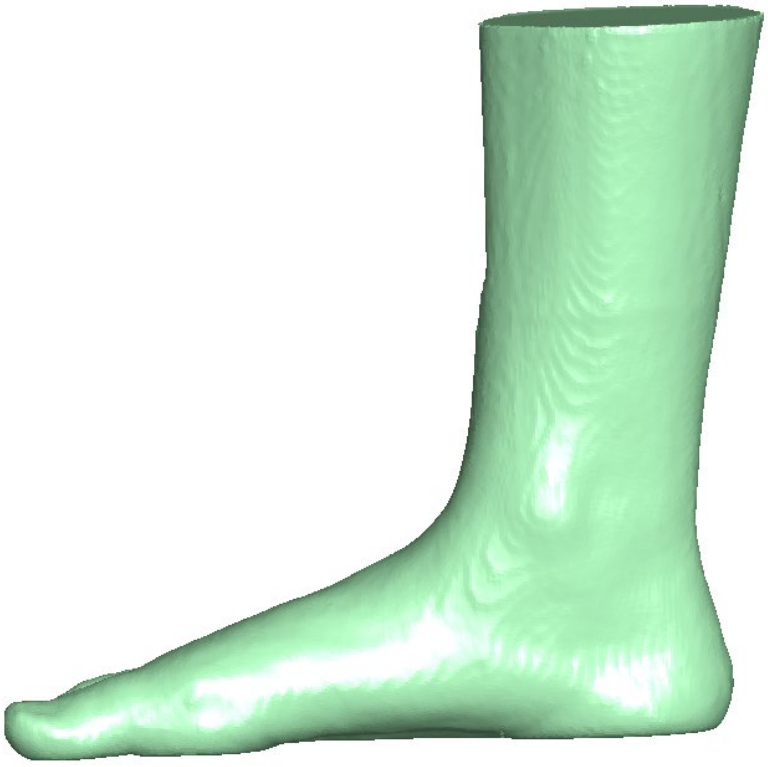


Figure 1. 3D model of the foot.

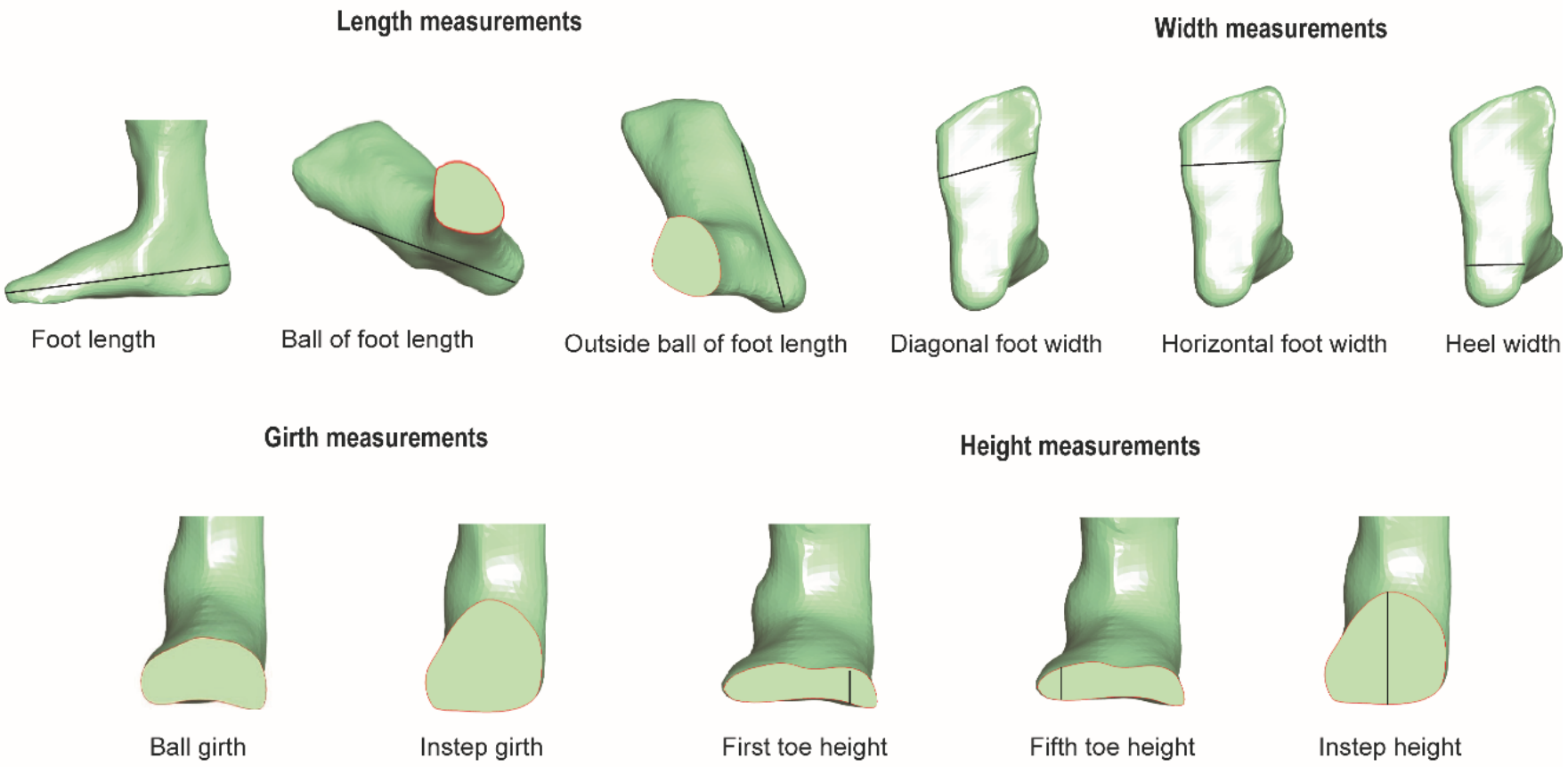


Figure 2. Foot dimension measurement technique.

List of figures

List of abbreviations

3D: Three-dimensional.

STL: Stereolithography.

STROBE: Strengthening the Reporting of Observational Studies in Epidemiology Statement.

Authors' contributions

Conception and design – NMH, KBL, NS and SEM

Collection and assembly of data – NMH and AKB

Analysis and interpretation of data – NMH, NS, KBL, HBM and SEM

Drafting of article – NMH, KBL, NS and SEM

Final approval of article – NMH, AKB, NS, KBL, HBM and SEM

Chapter 6 – Discussion

6.1 Summary of key findings

This thesis examined two interrelated problems – reduced participation in physical activity and poorly-fitting footwear in children with Down syndrome. The two primary aims of this thesis were to determine the: (i) effectiveness of interventions (including custom-fitted footwear) to increase physical activity in children with Down syndrome, and (ii) foot dimensions of children with Down syndrome, to better understand the issue of poorly-fitting footwear. Four related studies were completed that address these aims. The following section summarises the key findings identified in these studies.

Key finding 1 (Chapter 2) – No previous studies investigated the effectiveness of footwear to increase physical activity in children with Down syndrome

The aim of the study presented in Chapter 2 was to determine the effectiveness of interventions to increase physical activity in individuals with intellectual disabilities. In order to do this, a systematic review of randomised trials was conducted to synthesise findings from high level evidence. Key findings were: (i) a small body of evidence (9 studies) currently exists regarding the effectiveness of interventions in increasing physical activity in individuals with intellectual disabilities; (ii) no trials had evaluated the potential effects of footwear in increasing physical activity in individuals with intellectual disabilities, including those with Down syndrome; (iii) several trials had methodological limitations and none were rated as low risk of bias; (iv) a limited number of interventions were effective in increasing physical activity in individuals with intellectual disabilities, and the magnitude of effect was small to large; (v) three trials included children or adolescents with Down syndrome as participants; and (vi) only one trial, involving adolescents with Down syndrome, demonstrated positive effects of an intervention (progressive resistance training program) on physical activity.

No previous randomised trials had evaluated the effects of footwear on physical activity in children with Down syndrome. However, there is preliminary evidence to show poorly-fitting footwear is associated with reduced physical activity in children with Down syndrome [80], so further research was warranted to evaluate the effectiveness of appropriate footwear fit in increasing physical activity.

Key finding 2 (Chapter 3) – A definitive randomised trial to evaluate the efficacy of custom-fitted footwear to increase physical activity in children with Down syndrome is feasible, but commercially available footwear may not be suitable

The aim of the study presented in Chapter 3 was to investigate the feasibility of a definitive randomised trial to evaluate the efficacy of custom-fitted footwear to increase physical activity in children with Down syndrome. To achieve this, a randomised pilot study was performed that evaluated six feasibility domains; demand, implementation, acceptability, practicality, limited efficacy testing and adaptation. Key findings from this study were: (i) a definitive randomised trial is feasible, (ii) intervention adherence and acceptability were high, and (iii) positive trends for effects on physical activity in the intervention group were found in the short term (6 weeks). However, several issues need to be considered prior to embarking on a definitive trial, including: (i) the rate of recruitment was lower than anticipated due to long travel distances and competing family responsibilities of participants' caregivers, (ii) the use of co-interventions was high, and importantly, (iii) footwear fit of the intervention was no better than participants' existing footwear.

The primary issue with the intervention (i.e. commercially available footwear) used was insufficient width fittings to accommodate the width requirement of the forefoot in children with Down syndrome. Although steps were taken to custom-fit the footwear – such as using footwear manufactured by a reputable children's footwear manufacturer, using styles of footwear with multiple width fittings, adhering to a measurement and fitting protocol provided by the manufacturer, using footwear styles that were adjustable to suit width (i.e. Velcro and lace fixation options, and using footwear that had a removable foot bed to increase space) – this was not sufficient to accommodate foot width. Therefore, these findings indicate commercially available footwear does not provide an appropriate fit for children with Down syndrome, which may explain the high prevalence of poorly-fitting footwear in this population.

Key finding 3 (Chapter 4) – Measuring the foot dimensions of children with Down syndrome is reproducible using 3D foot scans

The aim of the study presented in Chapter 4 was to determine the intra- and inter-rater reproducibility of measuring foot dimensions of children with Down syndrome using 3D foot scans. In order to improve the understanding of variations in foot structure of children with Down syndrome, its impact on footwear fit, and subsequently, to design

appropriately fitting footwear, it is critical that 3D measurements of foot dimensions are reproducible.

A method for obtaining detailed measurements of foot dimensions (including lengths, widths, girths and height measurements) of children with Down syndrome using 3D foot scans was developed and the intra- and inter-rater reproducibility was evaluated. Key findings from this study were: (i) measuring the foot dimensions of children with Down syndrome is reliable, as there was moderate to excellent reliability for all measurements performed; (ii) agreement for the measurement of foot length was acceptable; (iii) the acceptability of agreement for the remaining measurements was unclear. Overall, the findings from the study in Chapter 4 indicate that the reproducibility of foot dimension measurements from 3D foot scans is acceptable, which means the method can be used in future studies in this population.

Key finding 4 (Chapter 5) – Children with Down syndrome have shorter, wider feet with increased girth and fifth toe height compared to typically developing children

The aim of the study presented in Chapter 5 was to compare the 3D foot dimensions of children with Down syndrome to typically developing children. A cross-sectional observational study was conducted with 51 typically developing children who were matched according to age (±2 years) and sex to 51 children with Down syndrome. Both absolute and normalised (to scale) differences in foot dimensions were assessed. Key findings were: (i) children with Down syndrome have smaller foot dimensions overall; and (ii) when foot dimensions were normalised to scale, children with Down syndrome have a shorter foot (heel to toe measurement) but a longer ball of foot length measurement (heel to ball measurement), a wider forefoot with increased girth (ball and instep girth), and a greater fifth toe height measurement. These findings confirm there are differences in the 3D foot shape of children with Down syndrome, and this should be considered when fitting and manufacturing footwear for this population.

These findings are consistent with the previously conducted studies that infer differences in foot structure between children with and without Down syndrome in terms of length and width [59, 61-63, 65, 66]. However, the study presented in Chapter 5 has added to the existing knowledge by describing other dimensions that have not been previously evaluated. Specifically, these findings show children with Down syndrome have a longer ball of foot length, increased girth measurements (which highlights the volume of the foot), and a greater fifth toe height measurement. These findings are important as they

provide information regarding critical dimensions that are important in assessing footwear fit, fitting footwear, as well as footwear manufacturing.

6.2 Discussion of the main findings

6.2.1 There are limited interventions that have been shown to increase physical activity in children with Down syndrome

There are limited interventions that increase physical activity in children with Down syndrome. Only three interventions have been shown to be effective in improving physical activity; but these interventions include individuals with intellectual disabilities of any origin and only one intervention involved adolescents with Down syndrome. These interventions are:

i. A 12-week gym-based, progressive resistance training program (compared to a social group) [124], which had a large effect on maintaining physical activity;
ii. A multicomponent diet and physical activity program (compared to a wait-list control group [99]), which had a small effect on physical activity;
iii. A physical activity framework and education program (compared to usual care [102], which had a large effect on physical activity.

These interventions, which all had a positive effect on physical activity, share common characteristics that may play a role in their effectiveness: access, familiarity, routine and support. First, each of the interventions were implemented in a convenient, accessible location for participants. The first intervention was implemented at a local community gym [124], the second at participants' homes [99], and the final at a day activity centre regularly attended by participants [102]. Utilising a convenient location improves access by reducing barriers to participation (such as long travel distances or transportation issues) and has the additional advantage of enabling physical activity without the need for specific equipment, as the physical activity programs can be adapted to an individual's environment. Second, implementing a physical activity intervention in a familiar environment (such as participants homes) and involving the individuals who provide care is also more likely to encourage individuals with an intellectual disability to engage in organised activities as familiarity facilitates physical activity [129, 130]. Third, the interventions encouraged an individual to develop a routine. A routine of physical activity provides individuals with regular opportunities to practice and improve their skills [119, 129, 131]. This may help improve their confidence in their abilities to engage in physical activity. Establishing a routine of physical activity is also beneficial because it creates a habit. Finally, a support person can facilitate the physical activity intervention by encouraging and supporting individuals with an intellectual disability to participate. A

support person can initiate physical activity as well as participate in physical activity alongside individuals with an intellectual disability to help their adherence. Involving a support person during physical activity is also useful in assisting individuals with an intellectual disability in learning new skills through imitation [119, 129].

Some characteristics of the interventions outlined above, however, may have had less of an influence on its effectiveness. For example, although the interventions were adapted to suit the needs of individuals with intellectual disabilities (e.g. by reducing complex components or modifying equipment), they were not population-specific, as they were adaptations of existing programs created for the general public. It is possible that some components of the interventions were too complex, or adaptations were inadequate, for participants with an intellectual disability to benefit. In addition, several interventions were based on theories of behavioural change such as Social Cognitive Theory, which is grounded on self-efficacy and motivation. Although behavioural change is more likely to occur when based on an appropriate theory [132], the use of behaviour change theories for those with intellectual disabilities has been questioned due to the level of abstract thinking required [133]. Further, despite the lengthy duration of some interventions (which allows individuals enough time to develop new routines or behaviours), no long-term intervention was effective.

The generalisability of the findings from the existing trials to children with Down syndrome also needs consideration. Only three trials [124-126] included children with Down syndrome, and only one intervention (progressive resistance training) was shown to be effective in maintaining physical activity in young adults (aged between 14 to 22 years) [124]. Further, a gym-based progressive resistance training intervention may not be appropriate for younger children. Therefore, there are no interventions supported by randomised controlled trials that have been shown to be effective at increasing physical activity in school aged children with Down syndrome.

Similar to the findings of Chapter 2 (systematic review), there are few interventions to increase physical activity in children with intellectual disabilities (i.e. not only children with Down Syndrome) and have limited effectiveness [133]. More broadly, while interventions aimed to increase physical activity in children with any kind of disability (including physical, intellectual, sensory and developmental disabilities) have short terms benefits, there are methodological limitations to available published studies, such as generalisability, transferability and scientific rigour [134].

6.2.2 Children with Down syndrome have a unique foot structure and this has implications for footwear fit

Children with Down syndrome have a unique foot structure, which is a proportionally shorter foot length (heel to toe measurement), a longer ball of foot length (heel to ball measurement), greater forefoot width, greater foot girth (ball and instep girth), and greater fifth toe height. These findings, outlined in Chapter 5, broadly align with previous descriptions of the feet of individuals with Down syndrome [59, 61-63, 65, 66], but they also add to existing knowledge by providing a thorough description of the 3D foot dimensions of children with Down syndrome. These findings have identified new information regarding the variations of the ball of foot length, girth and height measurements of the feet of children with Down syndrome. These novel findings suggest that achieving appropriate footwear fit extends beyond fitting based on length and width measurements, as traditionally performed. These findings also indicate that footwear fit can be inappropriate even with the correct length and width fitting.

The improved understanding of the unique foot shape of children with Down syndrome allows for a better evaluation of footwear fit in this population, and this may lead to the supply of more appropriately fitting footwear. At present, children with Down syndrome commonly wear footwear that is either too long when fitted according to width, or too narrow when fitted to length. This can be explained by the unique dimensions of the feet of children with Down syndrome that are wider at the forefoot (with greater girth measurements) and shorter in length [70]. Further, additional considerations for width may be required in the presence of a foot deformity, such as hallux valgus. When attempting to accommodate a wider foot, larger footwear sizes are often selected, which increases footwear length. This subsequent change in length (which is based on the foot width fitting as opposed to the true length of the foot) then affects the position of the metatarsophalangeal joints, particularly the first metatarsophalangeal joints, relative to the treadline of footwear. This distorts the overall fit, as footwear fitted will be too long [65, 79-81]. Therefore, width fitting is the most problematic issue when fitting footwear [135], and explains why children with Down syndrome frequently wear poorly-fitting footwear.

Evaluating the range of foot dimensions allows for the identification of key areas of footwear fit that can be improved. The findings from Chapter 5 suggest in order to appropriately accommodate the feet of children with Down syndrome, there are some key considerations. First, greater width and girth fittings are necessary to accommodate the increased volume of the foot at the forefoot and midfoot. Second, a deeper toe box is needed to accommodate for increased toe height. Lastly, ball of foot length as well as overall footwear length (heel to toe measurement) need to be considered in the fitting

process. Fitting according to the ball of foot length measurement will ensure correct alignment of the first metatarsophalangeal joint at the treadline of the shoe.

Due to the limited research on footwear fit in children with Down syndrome, it may be useful to consider the outcomes of other studies that have improved footwear fit in other populations. Footwear fitting issues are common in older individuals who have structural variations. These variations may be related to ethnic or sex-differences to foot structure (e.g. different lengths, widths, girth and height measurements) or structural deformities (such as forefoot deformities), which are prevalent in older people. In previous clinical trials, off-the shelf, extra-depth and width footwear (known as medical-grade footwear) has been shown to improve footwear fit in older people [136, 137]. Further, extra-depth and width footwear significantly improved foot pain and function, which is an indication of improved foot health [137]. These findings may be relevant to children with Down syndrome, since both groups share similar foot characteristics – older individuals typically have a broader forefoot [79], forefoot deformities, and are more likely to have a flatter medial arch [79].

While extra-depth and width footwear might be a potential option for improving footwear fit in children with Down syndrome, access to such footwear needs to be considered. Although extra-depth and width footwear for children is available online, it is not readily available in a retail setting (Figure 3). This limits the options available for children with Down syndrome where being fitted prior to purchase could be considered to be critical to ensuring the fit is correct. It is also unclear whether extra-depth and width footwear would be aesthetically pleasing for children with Down syndrome, which may affect wear adherence. Therefore, off-the-shelf, extra-depth and width footwear may be an option for children with Down syndrome; however further research is required to determine if: (i) such footwear can be purchased in a way to ensure that it fits correctly; and (ii) the styles of footwear available are aesthetically acceptable.

Figure 3. Examples of off-the-shelf, extra-depth and width footwear for children and adults. Top row: children's footwear obtained from an online retail store (left: Healthyfeetstore.com, Mt Emey 3301 model, retailing for $100 USD, with extra wide fitting [138]; right: Wellandable.com.au, Boston school shoe model retailing at $268 AUD, with wide fitting [139]). Bottom row: adults' footwear available online or through healthcare providers (left: Dr. Comfort, Maggie X model retailing at $289 AUD [140]; right: Dr Comfort, William X model retailing at $279 AUD [141], with both models having extra wide fitting).

6.2.3 Commercially available footwear is not suitable for children with Down syndrome

A key finding from the pilot study presented in Chapter 3 was that custom-fitted footwear did not accommodate the foot structure of children with Down syndrome. Indeed, the fit of the footwear was found to be no better fitting than participants' existing footwear, highlighting a major limitation in the suitability of commercially available footwear. This indicates that commercially available footwear is not suitable for all children with Down syndrome, and as such, parents are likely to experience difficulty acquiring footwear for their children that fits appropriately.

A limitation of commercially available footwear is that there are insufficient width fittings to accommodate the relatively wide forefoot shape of children with Down syndrome [135]. There are limited styles of footwear that are designed to accommodate variations in foot structure or structural deformities that increase width dimensions of the forefoot, as these differences are not usually seen in typically developing children. This limits

children with Down syndrome from being able to wear certain styles of footwear. It is possible that these issues may not be a problem across all footwear brands. However, the challenge when fitting children with Down syndrome is identifying commercially available footwear that has sufficient dimensions to accommodate their feet.

Besides limited width fittings, there are a number of other reasons to explain why commercially available footwear is not suitable for children with Down syndrome. The first is the existing practice of fitting footwear according to length and width measurements, as footwear sizes are based on these measurements [135]. This approach oversimplifies footwear fitting as it does not consider the way in which other foot dimensions, such as foot girth, influence overall fit. Footwear fit is further compounded by the inconsistencies in sizing that occur across footwear brands. This sizing issue occurs because of the different sizing and grading systems (including the French, Chinese, Japanese, American, British and Mondo Point systems) used across the world. Each system varies in its incremental differences between sizes, and this may affect the resulting footwear fit when attempting to purchase footwear that is the same size as an individual's existing footwear [135]. This misfit may be further accentuated in the presence of foot deformities or significant variations in the average foot shape, which is typically seen in the Down syndrome population.

Although commercially available footwear is not able to accommodate the foot structure of children with Down syndrome, a number of compensatory footwear fitting techniques may be used to address the limitations of commercially available footwear in the interim. Initially, applying appropriate footwear fitting techniques in conjunction with selecting footwear with favourable features may improve footwear fit. This includes use of footwear with: (i) a malleable upper material to accommodate structural variations, (ii) a deep round-shaped toe box to accommodate differences in toe height, (iii) multiple width fittings at a given size (e.g. New Balance® footwear, or Dr Comfort® footwear for older children who may fit into adult sizes), and (iv) lace-up or Velcro fixation to allow for adjustment.

Although these compensatory footwear fitting techniques may be useful, these adjustments may not be able to improve all aspects of footwear fit for all children, as happened in our trial (Chapter 3). To address this, two alternative options are available. First, extra-depth and width footwear (medical-grade footwear) can be used, which is likely to cater for the additional depth and width required. Second, if extra-depth and width footwear does not provide an adequate fit, custom-made footwear can be accessed. Both of these options are relatively expensive, particularly when it is considered that the footwear will be replaced regularly to allow for growth. Fortunately, external funding schemes are available in Australia (e.g. State-wide Equipment Program

[SWEP] and the National Disability Insurance Scheme [NDIS]) for children with Down syndrome and can help families access footwear by covering the costs of medical-grade footwear or custom-made footwear as required.

A long-term solution to improving footwear fit of children with Down syndrome is to create footwear that is based on the foot dimensions of this population. Manufacturing footwear for children with Down syndrome using normative data for this population would result in footwear that appropriately accommodates their unique foot dimensions. While the findings from Chapter 5 provide preliminary data on the foot dimensions of children with Down syndrome, the sample size included in the study was relatively small (51 children with Down syndrome) and is unlikely to be representative of all children with Down syndrome. A representative sample would need to consider: (i) the number of children with Down syndrome within a region (e.g. within Australia); (ii) the distribution of ages within the age bracket of 5 to 18 years and (iii) the number of boys and girls with Down syndrome, including those within each age group. In previous studies, the number of 3D foot scans analysed for foot dimension data have ranged substantially, from 42 to 9,220 [70, 71, 76, 142-150]. A more recent study evaluated 1.2 million foot scans across three geographical regions [151]. Therefore, a significant body of further work (with the input of a statistician) is required to obtain a large database of 3D foot scans that is representative of children with Down syndrome to allow for footwear manufacturing for this population.

6.2.4 Using innovative technology to design and manufacture customised footwear is a possible alternative to commercially available footwear, which can help solve footwear fitting issues in children with Down syndrome

With advancements in technology, innovative technologies may play a pivotal role in the future of footwear design and manufacture of footwear, particularly in developing affordable, customised footwear. This could have a substantial effect on resolving footwear fitting issues experienced by children with Down syndrome as it addresses the limitations of mass-produced, generic footwear.

Mass-customisation of footwear uses technology to develop products that are tailored to the needs of individuals while minimising the associated cost of production [152]. As discussed previously, children with Down syndrome require footwear that is appropriately fitting, affordable and readily available, which suggests that mass-customisation of footwear may be an ideal approach for this population. Aesthetics is also an important factor as the appearance of footwear can have social implications (e.g. identity and

personal style a child may identify with), and thus it may affect adherence [153]. A framework for mass-customisation has been described elsewhere [154]. Essentially, mass-customisation of footwear uses a computer-automated design system to design customised footwear in relation to style and fit. 3D scanning is used to obtain foot dimension data that are used to create the digitised foot model for that individual. Footwear style is then decided upon by the consumer through an existing database of footwear styles. Once the style has been finalised, the available footwear lasts are adapted according to the dimensions of the consumer (obtained through the 3D foot scan) and the footwear is manufactured [154]. This system bridges the gap between manufacturers and consumers and provides consumers a significant role in determining their preferred footwear that have a superior fit to commercially available footwear [154]. In future, mass-customisation of footwear may replace traditional methods of footwear manufacturing for both custom-made footwear (which is time consuming, labour intensive and costly) and generic footwear. Although exciting, this advanced process of footwear manufacture is still in development and likely to take time before it is fully implemented [155].

6.3 Future research

To build on the knowledge generated from this thesis, it would be important to solve the issue of poor footwear fit in children with Down syndrome before a definitive randomised trial is conducted to evaluate the effectiveness of custom-fitted footwear to increase physical activity. The use of extra-depth and width footwear is a possibility, and an initial step would be to collaborate with manufacturers of extra-depth and width footwear to compare footwear last dimensions with the average foot dimensions of children with Down syndrome obtained from 3D foot scans (e.g. data from Chapter 5). If it was found that their foot dimensions can be accommodated in such footwear, a definitive trial using extra-depth and width footwear could be conducted. Important outcome measures such as appropriateness of footwear fit and acceptability of footwear (i.e. acceptability of the footwear in relation to its aesthetics, and adherence to wear as measured in the randomised pilot study) would need to be evaluated. Other limitations identified in the pilot study presented in Chapter 3 need to be addressed prior to a definitive trial. For example, the slow recruitment rate as a result of long travel distances to assessment location can be improved by altering data collection methods. This may include collecting outcome measurements online where feasible (e.g. using REDCap software), as well as allowing some flexibility with the location of the trial to collect data that cannot be collected through an online platform (e.g. multi-site trial).

To further improve footwear fit, creating footwear that is specific to the foot dimensions of children with Down syndrome (i.e. wider, deeper footwear that can accommodate for the increased girth and toe height, at a given size) is necessary. To do this, further research is required to establish normative data for the foot dimensions of children with Down syndrome. Data on foot dimensions can be collected through the use of 3D foot scans for analysis, and the analysis can be further extended to evaluate sex- and age-based differences, as evaluated in studies of other populations [70, 143, 151]. Findings from this study can then be factored into the footwear development process. A prototype of the footwear at a range of sizes can be trialled in future studies to determine the appropriateness of fit and acceptability in children with Down syndrome prior to developing this range on a commercial scale.

6.4 Strengths and limitations of this thesis

The strengths of this thesis lie in its examination of the two interrelated concepts of physical activity and footwear in children with Down syndrome, which have not been investigated previously. The findings build upon previous literature by: (i) synthesising research that has evaluated the effectiveness of physical activity interventions; (ii) identifying key considerations for a definitive randomised trial; (iii) providing preliminary findings on a novel intervention (custom-fitted footwear) to improve physical activity; (iv) establishing 3D scanning is a reproducible method of measuring foot dimensions for children with Down syndrome; and (v) providing an in-depth understanding of the foot dimensions of children with Down syndrome.

A variety of study designs were implemented to thoroughly answer the research questions that were posed. Rigorous methods reduced the effects of confounding factors (such as risks of bias), and valid and reliable tools were consistently used to collect data, thus increasing confidence in the findings. The systematic review and the randomised pilot study were registered prospectively (with PROSPERO and ANZCTR). The use of relevant reporting guidelines (e.g. PRISMA, TIDier, CONSORT, GRAAS, STROBE) also ensured transparency and consistency when reporting the studies.

The findings from the studies conducted in this thesis have several applications across clinical, research and industry settings. There is potential for the findings to have an impact on clinical and research practice, that will ultimately lead to positive improvements to the health and quality of life of children with Down syndrome. For example, for clinicians, the findings provide a better understanding of footwear fitting issues commonly experienced in children with Down syndrome and highlight how footwear fit can be evaluated and improved. For researchers, future studies can be guided by the available data, including using: (i) the randomised pilot study (Chapter 3) to plan a definitive randomised trial, and (ii) the measurement technique (presented in Chapter 4 and 5) to obtain normative data on the foot dimensions of children with Down syndrome. For industry, the findings will assist in the design and manufacture of footwear specific to children with Down syndrome. Additionally, this thesis focused on a young age group, which is an important group for research to focus on as health improvements during childhood years have implications that persist into adulthood.

There are also some limitations. Randomised trials investigating the effectiveness of physical activity interventions (i.e. those included in Chapter 2) had significant heterogeneity in the types of interventions and time-points that were evaluated, which limited the ability to conduct a meta-analysis. A meta-analysis would have been useful to improve statistical power and the precision of the effect estimates. There were also methodological issues associated with the studies (such as missing data and risk of

bias), which reduces the certainty of the confidence of the findings. In addition, because physical activity interventions are physical, participant blinding is difficult. Although this is recognised as an inherent issue in randomised trials [156], the possibility of ascertainment bias affecting the estimate of effects of such interventions cannot be overlooked.

A further limitation relates to the inclusion criteria used in the randomised pilot study presented in Chapter 3. Participants did not have their existing footwear screened for appropriateness of fit prior to being enrolled in the study. Although this was not an issue for the majority of participants (as 88% of participants wore poorly-fitting footwear prior to being enrolled), this is an important consideration in any future trial as it may interfere with the difference in the effect between the experimental footwear and the existing footwear (as there would be little difference in fit between the two shoes, thus masking any effect if the experimental intervention was effective). Therefore, any future trial should revise the eligibility criteria and exclude children with Down syndrome who present with appropriately fitting footwear (if any).

In addition to this, future research involving the lower extremity in children with Down syndrome should also consider any conditions that may affect joints and their function (such as Juvenile Idiopathic Arthritis [51]) due to such a condition being under-diagnosed in this population. This is also necessary when using interventions that may impact joint function. Future studies should consider these conditions when designing studies, for example, when developing participant inclusion and exclusion criteria.

Finally, in the case-matched study presented in Chapter 5, a technical error with the original foot scanner (3D FotoScan foot scanner) occurred, so the scanner was not able to be used for participants in the control group. Therefore, all participants in the control group had their foot scanned using a more recent scanner (INFOOT foot scanner). Even though both scanning devices have the high accuracy (within 0.5 to 1.0 mm) and create the same output file, it was not ideal to measure participants using different scanners. It was also not feasible to ask the children with Down syndrome to attend a further testing session for the purpose of having their foot rescanned.

6.5 Conclusion

Footwear fit is a problem for children with Down syndrome. Footwear may have potential to improve participation in physical activity, but this is difficult to evaluate empirically until there is a solution to the issue of limited availability of appropriately-fitting footwear. Possible solutions to this problem that warrant further investigation are extra-depth and width footwear, and innovative approaches to footwear production using 3D scanning

and manufacture. Further studies could then test the hypothesis that foot health status and the amount of physical activity of children with Down syndrome could be improved through footwear modification. Given footwear plays a substantial role in performing day-to-day activities, it is imperative that footwear fit is improved for children with Down syndrome.

References

1. Lanphear NE, Castillo HA. Down Syndrome. In: Garfunkel LC, Kaczorowski JM, Christy C, editors. Pediatric Clinical Advisor (Second Edition). Philadelphia: Mosby; 2007. p. 175-6.

2. Down Syndrome Australia. Down syndrome population statistics. 2020. [Accessed 21 July 2020]. Available from: https://www.downsyndrome.org.au/about-down-syndrome/statistics/.

3. Collins VR, Muggli EE, Riley M, Palma S, Halliday JL. Is Down syndrome a disappearing birth defect? J Pediatr. 2008;152(1):20-4.

4. Halliday JL, Watson LF, Lumley J, Danks DM, Sheffield LJ. New estimates of Down syndrome risks at chorionic villus sampling, amniocentesis, and livebirth in women of advanced maternal age from a uniquely defined population. Prenat Diagn. 1995;15(5):455-65.

5. Versacci P, Di Carlo D, Digilio MC, Marino B. Cardiovascular disease in Down syndrome. Curr Opin Pediatr. 2018;30(5):616-22.

6. Glasson EJ, Sullivan SG, Hussain R, Petterson BA, Montgomery PD, Bittles AH. The changing survival profile of people with Down's syndrome: implications for genetic counselling. Clin Genet. 2002;62(5):390-3.

7. Bittles AH, Bower C, Hussain R, Glasson EJ. The four ages of Down syndrome. Eur J Public Health. 2006;17(2):221-5.

8. Halliday J, Collins V, Riley M, Youssef D, Muggli E. Has Prenatal Screening Influenced the Prevalence of Comorbidities Associated with Down Syndrome and Subsequent Survival Rates? Pediatrics. 2009;123(1):256-61.

9. Kahlem P. Gene-dosage effect on chromosome 21 transcriptome in trisomy 21: implication in Down syndrome cognitive disorders. Behav Genet. 2006;36(3):416-28.

10. Hassold T, Sherman S. Down syndrome: genetic recombination and the origin of the extra chromosome 21. Clin Genet. 2000;57(2):95-100.

11. Oliver TR, Feingold E, Yu K, Cheung V, Tinker S, Yadav-Shah M, et al. New insights into human nondisjunction of chromosome 21 in oocytes. PLoS Genet. 2008;4(3):e1000033.

12. Kazemi M, Salehi M, Kheirollahi M. Down Syndrome: Current Status, Challenges and Future Perspectives. Int J Mol Cell Med. 2016;5(3):125-33.

13. Papavassiliou P, Charalsawadi C, Rafferty K, Jackson-Cook C. Mosaicism for trisomy 21: a review. Am J Med Genet A. 2015;167a(1):26-39.

14. Kelminson KL, Elias ER, Goldson E. Down syndrome. In: Bajaj L, Hambidge SJ, Kerby G, Nyquist A-C, editors. Berman's Pediatric Decision Making (Fifth Edition). Saint Louis: Mosby; 2011. p. 680-3.

15. Coppedè F. Risk factors for Down syndrome. Arch Toxicol. 2016;90(12):2917-29.

16. Warren AC, Chakravarti A, Wong C, Slaugenhaupt SA, Halloran SL, Watkins PC, et al. Evidence for reduced recombination on the nondisjoined chromosomes 21 in Down syndrome. Science. 1987;237(4815):652-4.

17. Weijerman ME, de Winter JP. Clinical practice. The care of children with Down syndrome. Eur J Pediatr. 2010;169(12):1445-52.

18. American Academy of Pediatrics. Health supervision for children with Down syndrome. Pediatrics. 2001;107(2):442-9.

19. Mégarbané A, Noguier F, Stora S, Manchon L, Mircher C, Bruno R, et al. The intellectual disability of trisomy 21: differences in gene expression in a case series of patients with lower and higher IQ. Eur J Hum Genet. 2013;21(11):1253-9.

20. Cipriani G, Danti S, Carlesi C, Di Fiorino M. Aging With Down Syndrome: The Dual Diagnosis: Alzheimer’s Disease and Down Syndrome. Am J Alzheimers Dis Other Demen. 2018;33(4):253-62.

21. Castellani RJ, Rolston RK, Smith MA. Alzheimer disease. Dis Mon. 2010;56(9):484-546.

22. Castro P, Zaman S, Holland A. Alzheimer's disease in people with Down's syndrome: the prospects for and the challenges of developing preventative treatments. J Neurol. 2017;264(4):804-13.

23. Stephen R, Hongisto K, Solomon A, Lönnroos E. Physical Activity and Alzheimer's Disease: A Systematic Review. J Gerontol A Biol Sci Med Sci. 2017;72(6):733-9.

24. Roper RJ, Reeves RH. Understanding the basis for Down syndrome phenotypes. PLoS Genet. 2006;2(3):e50.

25. Dimopoulos K, Kempny A. Patients with Down syndrome and congenital heart disease: survival is improving, but challenges remain. Heart. 2016;102(19):1515-7.

26. Levenson D. Talking about Down syndrome. Am J Med Genet A. 2009;149a(4):vii-viii.

27. Fernhall B, Pitetti KH, Rimmer JH, McCubbin JA, Rintala P, Millar AL, et al. Cardiorespiratory capacity of individuals with mental retardation including Down syndrome. Med Sci Sports Excer. 1996;28(3):366-71.

28. Whooten R, Schmitt J, Schwartz A. Endocrine manifestations of Down syndrome. Curr Opin Endocrinol Diabetes Obes. 2018;25(1):61-6.

29. Amr NH. Thyroid Disorders in Subjects with Down Syndrome: An Update. Acta Biomed. 2018;89(1):132-9.

30. Pierce MJ, LaFranchi SH, Pinter JD. Characterization of Thyroid Abnormalities in a Large Cohort of Children with Down Syndrome. Horm Res Paediatr. 2017;87(3):170-8.

31. Kao CH, Chen CC, Wang SJ, Yeh SH. Bone mineral density in children with Down's syndrome detected by dual photon absorptiometry. Nucl Med Commun. 1992;13(10):773-5.

32. Carfi A, Liperoti R, Fusco D, Giovannini S, Brandi V, Vetrano DL, et al. Bone mineral density in adults with Down syndrome. Osteoporos Int. 2017;28(10):2929-34.

33. Gavris M, Courteix D, Serbanoiu S, Hantiu I, Tifrea C, Ferry B. Bone Tissue in Down Syndrome Patients Deteriorates Following Aging: A Study Based on Bone Ultrasound Analysis. J Med Ultrasound. 2014;22(1):29-36.

34. Bertapelli F, Pitetti K, Agiovlasitis S, Guerra-Junior G. Overweight and obesity in children and adolescents with Down syndrome-prevalence, determinants, consequences, and interventions: A literature review. Res Dev Disabil. 2016;57:181-92.

35. Hatch-Stein JA, Zemel BS, Prasad D, Kalkwarf HJ, Pipan M, Magge SN, et al. Body Composition and BMI Growth Charts in Children with Down Syndrome. Pediatrics. 2016;138(4):e20160541.

36. Melville CA, Cooper SA, McGrother CW, Thorp CF, Collacott R. Obesity in adults with Down syndrome: a case-control study. J Intellect Disabil Res. 2005;49(Pt 2):125-33.

37. Benhamu-Benhamu S, Garcia-de-la-Peña R, Gijon-Nogueron G, Jimenez-Cristino MD, Gordillo-Fernández LM, Dominguez-Maldonado G. Range of Ankle Dorsiflexion in a Group of Adults with Ligamentous Laxity. J Am Podiatr Med Assoc. 2018;108(3):245-52.

38. Agiovlasitis S, McCubbin JA, Yun J, Mpitsos G, Pavol MJ. Effects of Down syndrome on three-dimensional motion during walking at different speeds. Gait Posture. 2009;30(3):345-50.

39. Dey A, Bhowmik K, Chatterjee A, Chakrabarty PB, Sinha S, Mukhopadhyay K. Down Syndrome Related Muscle Hypotonia: Association with COL6A3 Functional SNP rs2270669. Front Genet. 2013;4:57.

40. Mendonca GV, Pereira FD, Fernhall B. Reduced exercise capacity in persons with Down syndrome: cause, effect, and management. Ther Clin Risk Manag. 2010;6:601-10.

41. Foley C, MacDermott E, Killeen O. Musculoskeletal anomalies in a national cohort of children and adolescents with trisomy 21. Pediatr Rheumatol Online J. 2014;12:P160.

42. Pikora TJ, Bourke J, Bathgate K, Foley K-R, Lennox N, Leonard H. Health Conditions and their impact among adolescents and young adults with Down Syndrome. PLoS One. 2014;9(5):e96868.

43. Mik G, Gholve PA, Scher DM, Widmann RF, Green DW. Down syndrome: orthopedic issues. Curr Opin Pediatr. 2008;20(1):30-6.

44. Ali FE, Al-Bustan MA, Al-Busairi WA, Al-Mulla FA, Esbaita EY. Cervical spine abnormalities associated with Down syndrome. Int Orthop. 2006;30(4):284-9.

45. Kocon H, Kabacyj M, Zgoda M. The results of the operative treatment of patellar instability in children with Down's syndrome. J Pediatr Orthop B. 2012;21(5):407-10.

46. Milbrandt TA, Johnston CEI. Down Syndrome and Scoliosis: A Review of a 50-Year Experience at One Institution. Spine. 2005;30(18):2051-5.

47. Talbot C, Alshryda S. Evidence-Based Treatment for Musculoskeletal Disorders in Children with Down's Syndrome. In: Alshryda S, Huntley JS, Banaszkiewicz PA, editors. Paediatric Orthopaedics: An Evidence-Based Approach to Clinical Questions. Cham, Switzerland: Springer; 2017. p. 519-26.

48. Caird MS, Wills BP, Dormans JP. Down syndrome in children: the role of the orthopaedic surgeon. J Am Acad Orthop Surg. 2006;14(11):610-9.

49. Galli M, Rigoldi C, Brunner R, Virji-Babul N, Giorgio A. Joint stiffness and gait pattern evaluation in children with Down syndrome. Gait Posture. 2008;28(3):502-6.

50. Foley CM, Deely DA, MacDermott EJ, Killeen OG. Arthropathy of Down syndrome: an under-diagnosed inflammatory joint disease that warrants a name change. RMD open. 2019;5:e000890.

51. Nicek A, Talib N, Lovell D, Smith C, Becker ML, Jones JT. Assessment and treatment of Down syndrome-associated arthritis: a survey of pediatric rheumatologists. Pediatr Rheumatol Online J. 2020;18:57.

52. Cremers MJG, Bol E, de Roos F, van Gijn J. Risk of sports activities in children with Down's syndrome and atlantoaxial instability. Lancet. 1993;342(8870):511-4.

53. Maranho D, Fuchs K, Kim Y-j, Novais EN. Hip Instability in Patients with Down Syndrome. J Am Acad Orthop Surg. 2018;26(13):455-62.

54. Bennet GC, Rang M, Roye DP, Aprin H. Dislocation of the hip in trisomy 21. J Bone Joint Surg Br. 1982;64(3):289-94.

55. Hresko MT, McCarthy JC, Goldberg MJ. Hip disease in adults with Down syndrome. J Bone Joint Surg Br. 1993;75(4):604-7.

56. Rebouças Moreira TA, Demange MK, Gobbi RG, Mustacchi Z, Pécora JR, Passarelli Tírico LE, et al. Trochlear dysplasia and patellar instability in patients with Down syndrome. Rev Bras Ortop. 2015;50(2):159-63.

57. Post WR, Fithian DC. Patellofemoral Instability: A Consensus Statement From the AOSSM/PFF Patellofemoral Instability Workshop. Orthop J Sports Med. 2018;6(1):2325967117750352.

58. Ercis M, Balci S, Atakan N. Dermatological manifestations of 71 Down syndrome children admitted to a clinical genetics unit. Clin Genet. 1996;50(5):317-20.

59. Prasher VP, Robinson L, Krishnan VH, Chung MC. Podiatric disorders among children with Down syndrome and learning disability. Dev Med Child Neurol. 1995;37(2):131-4.

60. Merrick J, Ezra E, Josef B, Hendel D, Steinberg DM, Wientroub S. Musculoskeletal problems in Down Syndrome European Paediatric Orthopaedic Society Survey: the Israeli sample. J Pediatr Orthop B. 2000;9(3):185-92.

61. Concolino D, Pasquzzi A, Capalbo G, Sinopoli S, Strisciuglio P. Early detection of podiatric anomalies in children with Down syndrome. Acta Paediatr. 2006;95(1):17-20.

62. Lim, Shields N, Nikolopoulos N, Barrett J, Evans A, Taylor N, et al. The association of foot structure and footwear fit with disability in children and adolescents with Down syndrome. J Foot Ankle Res. 2015;8:4.

63. Mansour E, Yaacoub JJ, Bakouny Z, Assi A, Ghanem I. A podoscopic and descriptive study of foot deformities in patients with Down syndrome. Orthop Traumatol Surg Res. 2017;103(1):123-7.

64. Perotti LR, Abousamra O, Del Pilar Duque Orozco M, Rogers KJ, Sees JP, Miller F. Foot and ankle deformities in children with Down syndrome. J Child Orthop. 2018;12(3):218-26.

65. Calvo-Lobo C, Ramos Garcia A, Losa Iglesias ME, Lopez-Lopez D, Rodriguez-Sanz D, Romero-Morales C, et al. The Relationship between Shoe Fitting and Foot Health of Persons with Down Syndrome: A Case Control Study. Int J Environ Res Public Health. 2018;15(5):983.

66. Puszczałowska-Lizis E, Nowak K, Omorczyk J, Ambroży T, Bujas P, Nosiadek L. Foot Structure in Boys with Down Syndrome. Biomed Res Int. 2017;7047468.

67. Lee YC, Wang MJ. Taiwanese adult foot shape classification using 3D scanning data. Ergonomics. 2015;58(3):513-23.

68. Goonetilleke R, Luximon A. Designing for comfort: a footwear application. Proceedings of Computer-Aided Ergonomics and Safety Conference Maui, Hawaii. 2001.

69. Goonetilleke R. The Science of Footwear. 1st ed ed. Boca Raton: CRC Press; 2013.

70. Krauß I, Grau S, Mauch M, Maiwald C, Horstmann T. Sex-related differences in foot shape. Ergonomics. 2008;51(11):1693-709.

71. Lee Y-C, Wang M-J. Taiwanese adult foot shape classification using 3D scanning data. Ergonomics. 2015;58(3):513-23.

72. Goonetilleke R. Foot anthropometry in Hong Kong. Proceedings of the ASEAN 97 Conference; Kuala Lumpur, Malaysia. 1997.

73. Lee Y-C, Lin G, Wang M-JJ. Comparing 3D foot scanning with conventional measurement methods. J Foot Ankle Res. 2014;7:44.

74. Saghazadeh M, Kitano N, Okura T. Gender differences of foot characteristics in older Japanese adults using a 3D foot scanner. J Foot Ankle Res. 2015;8:29.

75. Wunderlich RE, Cavanagh PR. Gender differences in adult foot shape: implications for shoe design. Med Sci Sports Exerc. 2001;33(4):605-11.

76. Lee YC, Kouchi M, Mochimaru M, Wang MJ. Comparing 3D foot shape models between Taiwanese and Japanese females. J Hum Ergol (Tokyo). 2015;44(1):11-20.

77. Telfer S, Woodburn J. The use of 3D surface scanning for the measurement and assessment of the human foot. J Foot Ankle Res. 2010;3:19.

78. Goonetilleke RS, Luximon A, Tsui KL. The Quality of Footwear Fit: What we know, don't know and should know. Proc Hum Factors Ergon Soc Annu Meet. 2000;44(12):515-8.

79. Buldt AK, Menz HB. Incorrectly fitted footwear, foot pain and foot disorders: a systematic search and narrative review of the literature. J Foot Ankle Res. 2018;11:43.

80. Shields N, Lim P, Wollersheim D, Nikolopoulos N, Barrett J, Evans A, et al. Do foot posture, deformity, and footwear fit influence physical activity levels in children with Down syndrome? A prospective cohort study. J Intellect Disabil Res. 2017;42(4):332-8.

81. Jenkins DW, Cooper K, O'Connor R, Watanabe L. Foot-to-shoe mismatch and rates of referral in Special Olympics athletes. J Am Podiatr Med Assoc. 2012;102(3):187-97.

82. Roizen N, Patterson D. Down's syndrome. Lancet. 2003;361(9365):1281-9.

83. Caspersen CJ, Powell KE, Christenson GM. Physical activity, exercise, and physical fitness: definitions and distinctions for health-related research. Public Health Rep. 1985;100(2):126-31.

84. Galson SK. The importance of being active your way. Public Health Rep. 2009;124(6):772-3.

85. Sharma A, Madaan V, Petty FD. Exercise for mental health. Prim Care Companion J Clin Psychiatry. 2006;8(2):106.

86. Warburton DER, Nicol CW, Bredin SSD. Health benefits of physical activity: the evidence. CMAJ. 2006;174(6):801-9.

87. World Health Organization. Global recommendations on physical activity for health 2010 [Accessed 23 July 2019]. Available from: http://apps.who.int/iris/bitstream/10665/44399/1/9789241599979_eng.pdf].

88. Janssen I. Physical activity guidelines for children and youth. Can J Public Health. 2007;98 Suppl 2:S109-21.

89. Janssen I, Leblanc AG. Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. Int J Behav Nutr Phys Act. 2010;7:40.

90. Dishman RK, Washburn RA, Schoeller DA. Measurement of Physical Activity. Quest. 2001;53(3):295-309.

91. Alhusaini AA, Ali Al-Walah M, Melam GR, Buragadda S. Pedometer-determined physical activity levels of healthy children and children with Down's syndrome. Somatosens Mot Res. 2017;34(4):219-25.

92. Serra MC, Balraj E, DiSanzo BL, Ivey FM, Hafer-Macko CE, Treuth MS, et al. Validating accelerometry as a measure of physical activity and energy expenditure in chronic stroke. Top Stroke Rehabil. 2017;24(1):18-23.

93. Voss C, Harris KC. Physical activity evaluation in children with congenital heart disease. Heart. 2017;103(18):1408-12.

94. Cain KL, Sallis JF, Conway TL, Van Dyck D, Calhoon L. Using accelerometers in youth physical activity studies: a review of methods. J Phys Act Health. 2013;10(3):437-50.

95. Smith M, Taylor S, Iusitini L, Stewart T, Savila Fa, Tautolo E-S, et al. Accelerometer data treatment for adolescents: Fitting a piece of the puzzle. Prev Med Rep. 2016;5:228-31.

96. Sirard JR, Slater ME. Compliance with wearing physical activity accelerometers in high school students. J Phys Act Health. 2009;6(Suppl 1):S148-S55.

97. Whitt-Glover MC, O'Neill KL, Stettler N. Physical activity patterns in children with and without Down syndrome. Pediatr Rehabil. 2006;9(2):158-64.

98. Shields N, Dodd KJ, Abblitt C. Do children with Down syndrome perform sufficient physical activity to maintain good health? A pilot study. Adapt Phys Activ Q. 2009;26(4):307-20.

99. Bergström H, Hagströmer M, Hagberg J, Elinder LS. A multi-component universal intervention to improve diet and physical activity among adults with intellectual disabilities in community residences: a cluster randomised controlled trial. Res Dev Disabil. 2013;34(11):3847-57.

100. McDermott S, Whitner W, Thomas-Koger M, Mann JR, Clarkson J, Barnes TL, et al. An efficacy trial of 'Steps to Your Health', a health promotion programme for adults with intellectual disability. Health Educ J. 2012;71(3):278-90.

101. Melville CA, Mitchell F, Stalker K, Matthews L, McConnachie A, Murray HM, et al. Effectiveness of a walking programme to support adults with intellectual disabilities to increase physical activity: walk well cluster-randomised controlled trial. Int J Behav Nutr Phys Act. 2015;12:125.

102. van Schijndel-Speet M, Evenhuis HM, van Wijck R, van Montfort KC, Echteld MA. A structured physical activity and fitness programme for older adults with intellectual disabilities: results of a cluster-randomised clinical trial. J Intellect Disabil Res. 2017;61(1):16-29.

103. Tudor-Locke C, Johnson WD, Katzmarzyk PT. U.S. population profile of time-stamped accelerometer outputs: impact of wear time. J Phys Act Health. 2011;8(5):693-8.

104. Tudor-Locke C, Barreira TV, Schuna JM, Jr., Mire EF, Chaput JP, Fogelholm M, et al. Improving wear time compliance with a 24-hour waist-worn accelerometer protocol in

the International Study of Childhood Obesity, Lifestyle and the Environment (ISCOLE). Int J Behav Nutr Phys Act. 2015;12:11.

105. Gabriel KP, McClain JJ, Schmid KK, Storti KL, High RR, Underwood DA, et al. Issues in accelerometer methodology: the role of epoch length on estimates of physical activity and relationships with health outcomes in overweight, post-menopausal women. Int J Behav Nutr Phys Act. 2010;7:53.

106. Kumahara H, Schutz Y, Ayabe M, Yoshioka M, Yoshitake Y, Shindo M, et al. The use of uniaxial accelerometry for the assessment of physical-activity-related energy expenditure: a validation study against whole-body indirect calorimetry. Br J Nutr. 2004;91(2):235-43.

107. Edwardson CL, Gorely T. Epoch length and its effect on physical activity intensity. Med Sci Sports Exerc. 2010;42(5):928-34.

108. Trost SG, McIver KL, Pate RR. Conducting accelerometer-based activity assessments in field-based research. Med Sci Sports Exerc. 2005;37(11 Suppl):S531-43.

109. Trost SG, Loprinzi PD, Moore R, Pfeiffer KA. Comparison of accelerometer cut points for predicting activity intensity in youth. Med Sci Sports Exerc. 2011;43(7):1360-8.

110. Fox B, Moffett GE, Kinnison C, Brooks G, Case LE. Physical Activity Levels of Children with Down Syndrome. Pediatr Phys Ther 2019;31(1):33-41.

111. Phillips AC, Holland AJ. Assessment of objectively measured physical activity levels in individuals with intellectual disabilities with and without Down's syndrome. PLoS One. 2011;6(12):e28618-e.

112. Izquierdo-Gomez R, Martínez-Gómez D, Acha A, Veiga OL, Villagra A, Diaz-Cueto M. Objective assessment of sedentary time and physical activity throughout the week in adolescents with Down syndrome. The UP&DOWN study. Res Dev Disabil. 2014;35(2):482-9.

113. Izquierdo-Gomez R, Martinez-Gomez D, Esteban-Cornejo I, Hallal PC, Garcia-Cervantes L, Villagra A, et al. Changes in objectively measured physical activity in adolescents with Down syndrome: the UP&DOWN longitudinal study. J Intellect Disabil Res. 2017;61(4):363-72.

114. Esposito PE, MacDonald M, Hornyak JE, Ulrich DA. Physical Activity Patterns of Youth with Down Syndrome. Intellect Dev Disabil. 2012;50(2):109-19.

115. Troiano RP, Berrigan D, Dodd KW, Masse LC, Tilert T, McDowell M. Physical activity in the United States measured by accelerometer. Med Sci Sports Exerc. 2008;40(1):181-8.

116. Booth FW, Roberts CK, Laye MJ. Lack of exercise is a major cause of chronic diseases. Compr Physiol. 2012;2(2):1143-211.

117. King LK, March L, Anandacoomarasamy A. Obesity & osteoarthritis. Indian J Med Res. 2013;138(2):185-93.

118. Pi-Sunyer X. The medical risks of obesity. Postgrad Med. 2009;121(6):21-33.

119. Barr M, Shields N. Identifying the barriers and facilitators to participation in physical activity for children with Down syndrome. J Intellect Disabil Res. 2011;55(11):1020-33.

120. Pitetti KH, Fernhall B. Comparing run performance of adolescents with mental retardation, with and without Down syndrome. Adapt Phys Activ Q. 2004;21(3):219-28.

121. Cioni M, Cocilovo A, Rossi F, Paci D, Valle MS. Analysis of ankle kinetics during walking in individuals with Down syndrome. Am J Ment Retard. 2001;106(5):470-8.

122. Alwhaibi RM, Aldugahishem HM. Factors affecting participation in physical activities in Saudi children with Down syndrome: mothers' perspectives. Disabil Rehabil. 2019;41(13):1524-35.

123. Alesi M, Pepi A. Physical Activity Engagement in Young People with Down Syndrome: Investigating Parental Beliefs. J Appl Res Intellect Disabil. 2017;30(1):71-83.

124. Shields N, Taylor NF, Wee E, Wollersheim D, O'Shea SD, Fernhall B. A community-based strength training programme increases muscle strength and physical activity in young people with Down syndrome: a randomised controlled trial. Res Dev Disabil. 2013;34(12):4385-94.

125. Angulo-Barroso R, Burghardt AR, Lloyd M, Ulrich DA. Physical activity in infants with Down syndrome receiving a treadmill intervention. Infant Behav Dev. 2008;31(2):255-69.

126. Ulrich DA, Burghardt AR, Lloyd M, Tiernan C, Hornyak JE. Physical Activity Benefits of Learning to Ride a Two-Wheel Bicycle for Children with Down Syndrome: A Randomized Trial. Phys Ther. 2011;91(10):1463-77.

127. Bartlo P, Klein PJ. Physical activity benefits and needs in adults with intellectual disabilities: systematic review of the literature. Am J Intellect Dev Disabil. 2011;116(3):220-32.

128. Heller T, McCubbin JA, Drum C, Peterson J. Physical activity and nutrition health promotion interventions: what is working for people with intellectual disabilities? Intellect Dev Disabil. 2011;49(1):26-36.

129. Mahy J, Shields N, Taylor NF, Dodd KJ. Identifying facilitators and barriers to physical activity for adults with Down syndrome. J Intellect Disabil Res. 2010;54(9):795-805.

130. van Schijndel-Speet M, Evenhuis HM, van Wijck R, van Empelen P, Echteld MA. Facilitators and Barriers to Physical Activity as Perceived by Older Adults with Intellectual Disability. Intellect Dev Disabil. 2014;52(3):175-86.

131. Downs SJ, Boddy LM, Knowles ZR, Fairclough SJ, Stratton G. Exploring opportunities available and perceived barriers to physical activity engagement in children and young people with Down syndrome. Eur J Spec Needs Educ. 2013;28(3):270-87.

132. Davis R, Campbell R, Hildon Z, Hobbs L, Michie S. Theories of behaviour and behaviour change across the social and behavioural sciences: a scoping review. Health Psychol Rev. 2015;9(3):323-44.

133. McGarty AM, Downs SJ, Melville CA, Harris L. A systematic review and meta-analysis of interventions to increase physical activity in children and adolescents with intellectual disabilities. J Intellect Disabil Res. 2018;62(4):312-29.

134. Lai B, Lee E, Wagatsuma M, Frey G, Stanish H, Jung T, et al. Research Trends and Recommendations for Physical Activity Interventions Among Children and Youth with Disabilities: A Review of Reviews. Adapt Phys Activ Q. 2020;37(2):211-34.

135. Kim N, Do W. Developing elderly men’s footwear sizing system based on their foot shapes. Fash Text. 2019;6(1):28.

136. Hurst B, Branthwaite H, Greenhalgh A, Chockalingam N. Medical-grade footwear: the impact of fit and comfort. J Foot Ankle Res. 2017;10:2.

137. Menz HB, Auhl M, Ristevski S, Frescos N, Munteanu SE. Effectiveness of off-the-shelf, extra-depth footwear in reducing foot pain in older people: a randomized controlled trial. J Gerontol A Biol Sci Med Sci. 2015;70(4):511-7.

138. Healthy Feet Store. Mt Emey 3305 - Kid's Orthopedic Boots 2020 [Accessed 14 June 2020]. Available from: https://www.healthyfeetstore.com/mt-emey-3305-kids-orthopedic-boot.html].

139. Well & Able. Boston School 2020 [Accessed 14 June 2020]. Available from: https://www.wellandable.com.au/boston-school].

140. Dr Comfort. Dr. Comfort Maggy-X Women's Double Depth Shoes 2020 [Accessed 14 June 2020]. Available from: https://www.drcomfort.com.au/shoes/women-s-shoes/double-depth/dr-comfort-maggy-x-women-s-double-depth-shoes.html].

141. Dr Comfort. Dr. Comfort William-X Men's Double Depth Shoes 2020 [Accessed 14 June 2020]. Available from: https://www.drcomfort.com.au/shoes/men-s-shoes/double-depth/dr-comfort-william-x-men-s-double-depth-shoes.html].

142. Luo G, Houston VL, Mussman M, Garbarini M, Beattie AC, Thongpop C. Comparison of male and female foot shape. J Am Podiatr Med Assoc. 2009;99(5):383-90.

143. Krauss I, Valiant G, Horstmann T, Grau S. Comparison of Female Foot Morphology and Last Design in Athletic Footwear—Are Men's Lasts Appropriate for Women? Res Sports Med. 2010;18(2):140-56.

144. Xiong S, Goonetilleke RS, Witana CP, Lee Au EY. Modelling foot height and foot shape-related dimensions. Ergonomics. 2008;51(8):1272-89.

145. Mickle KJ, Munro BJ, Lord SR, Menz HB, Steele JR. Foot shape of older people: implications for shoe design. Footwear Sci. 2010;2(3):131-9.

146. Jurca A, Kolsek T, Vidic T. Dorothy mass foot measurement campaign. Footwear Sci. 2011;3(sup1):S83-S5.

147. Krauss I, Langbein C, Horstmann T, Grau S. Sex-related differences in foot shape of adult Caucasians--a follow-up study focusing on long and short feet. Ergonomics. 2011;54(3):294-300.

148. Rodrigo AS, Goonetilleke RS, Witana CP. Model based foot shape classification using 2D foot outlines. Comput Aided Design. 2012;44(1):48-55.

149. Domjanic J, Fieder M, Seidler H, Mitteroecker P. Geometric morphometric footprint analysis of young women. J Foot Ankle Res. 2013;6:27.

150. Stanković K, Booth BG, Danckaers F, Burg F, Vermaelen P, Duerinck S, et al. Three-dimensional quantitative analysis of healthy foot shape: a proof of concept study. J Foot Ankle Res. 2018;11:8.

151. Jurca A, Žabkar J, Džeroski S. Analysis of 1.2 million foot scans from North America, Europe and Asia. Sci Rep. 2019;9(1):19155.

152. Novak B, Fatur P, Dolinsek S. Mass customization in footwear industry: A case study. 2007.

153. Farndon L, Robinson V, Nicholls E, Vernon W. If the shoe fits: development of an on-line tool to aid practitioner/patient discussions about ‘healthy footwear’. J Foot Ankle Res. 2016;9:17.

154. Zhang Y-F, Luximon A, Pattanayak A, Zhang M. Shoe-last design exploration and customization. J Text Inst. 2011;103:1-8.

155. Rout N, Zhang Y-F, Khandual A, Luximon A. 3D foot scan to custom shoe last. Special Issue IJCCT. 2010;1(2):14-8.

156. Boutron I, Tubach F, Giraudeau B, Ravaud P. Blinding was judged more difficult to achieve and maintain in nonpharmacologic than pharmacologic trials. J Clin Epidemiol. 2004;57(6):543-50.

Appendices

Appendix 1. Online supplementary files associated with the published systematic review

Supplementary file 1 – Search records

#	Search terms	MEDLINE	EMBASE	CINAHL	SPORTDiscus	Cochrane Library
1	Exp Intellectual Disability/or intellectual disability*.mp or exp Cognition Disorders/ or cognitive deficit*.mp or *mentally disabled persons/	184,365	455,893	14,143	1,140	3,299
2	Exercise.mp or exp Exercise/ or exp physical activity/ or physical activity*.mp	390,959	672,246	124,573	235,537	60,482
3	1 AND 2	2,841	11,352	360	248	234
4	Search filters*	908	1,389	129	Nil	Nil

Note: mp: title, original title, abstract, name of substance word, subject heading word. Exp: explode.*Details of the search filters are located in Additional file 2.

Supplementary file 2 – Search filters used in each database

MEDLINE	EMBASE	CINAHL	Cochrane Library[1] and SPORTDiscus
Exp Intellectual disability/or intellectual disabilit* .mp.	Exp Intellectual Disability/ or intellectual disabilit*.mp.	(MH "exp Intellectual Disability+") OR "intellectual disability*"	Intellectual disabilit*.ti,ab,kw
Exp Cognition Disorders/or cognitive deficit*.mp.	Cognitive deficit*.mp.	"cognitive deficit*" OR (MH exp "Cognition Disorders+")	Cognitive deficit*.ti,ab,kw
*mentally disabled persons/	*Mentally disabled persons/	(MH exp "Mentally Disabled Persons") OR "mentally disabled persons"	*mentally disabled persons:ti,ab,kw
1 or 2 or 3	1 or 2 or 3	1 OR 2 OR 3	1 or 2 or 3
Exp Exercise/ or exercise.mp.	Exp physical activity/ or physical activit*.mp.	(MH exp "Exercise+") OR "exercise"	Physical activit*:ti,ab,kw
Physical activity*.mp.	Exp exercise/ or exercise.mp.	(MH exp "Physical Activity") OR "physical activity*"	Exercise:ti,ab,kw
5 or 6	5 or 6	5 OR 6	5 or 6
4 and 7	4 and 7	4 AND 7	4 and 7
Randomized controlled trial.pt	Random:.tw.	(MH "Clinical Trials+")	
Controlled clinical trial.pt.	Placebo:.mp.	PT Clinical Trial	
Randomized.ab.	Double-blind:.tw.	TX clinic* n1 trial*	
Placebo.ab.	9 or 10 or 11	TX ((singl* n1 blind*) or (singl* n1 mask*)) or TX ((doubl* n1 blind*) or (doubl* n1 mask*)) or TX ((tripl* n1 blind*) or (tripl* n1 mask*)) or TX (trebl* n1 blind*) or (trebl* n1 mask*))	
Drug therapy.fs.	8 and 12	TX randomis* control* trial*	
Randomly.ab.		(MH "Random Assignment ")	
Trial.ab.		TX random* allocat*	
Groups.ab.		TX placebo	
9 or 10 or 11 or 12 or 13 or 14 or 15 or 16		(MH "Placebos")	
Exp animals/ not humans.sh.		(MH "Quantitative Studies")	
17 not 18		TX allocate* random*	

8 and 19	9 OR 10 0R 11 OR 12 OR 13 OR 14 OR 15 OR 16 OR 17 OR 18 OR 19
	8 AND 20

Abbreviations: exp, explode; MH, major heading; sh, medical subject heading; ti, title; ab, abstract; kw, keyword; pt, publication type; tw, title/abstract; mp denotes free text search; fs denotes floating subheading;. [1]ti, ab, kw used for Cochrane search strategy.

Supplementary file 3 – Risk of bias judgement

Angulo-Barroso et al. (2008)

Methods	Randomised controlled trial.	
Participants	30 infants with Down syndrome.	
Interventions	Individualised treadmill training protocols (high-intensity and low-intensity).	
Outcome	Changes to physical activity levels between groups during the intervention phase and at four different occasions during a 1-year follow up.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Unclear	Insufficient information provided to permit a judgement of risk.
Allocation concealment	Unclear	Insufficient information provided to permit a judgement of risk.
Blinding of participants	High	Participants not blinded due to the nature of the intervention.
Blinding Outcome assessor	Low	Not blinded, but main outcome was an objective outcome measure (activity log), so not likely to be influenced by lack of blinding.
Incomplete outcome data	High	Six out of 30 participants dropped out and were subsequently left out of the study, and the authors have not mentioned which group they dropped out of.
Selective reporting	Unclear	Insufficient information provided to permit a judgement of risk.
Other biases	Unclear	Insufficient information provided to permit a judgement of risk.

Bergstrom et al. (2011)

Methods	Cluster randomised controlled trial.	
Participants	129 adults with mild to moderate intellectual disabilities aged between 20-66 years.	
Interventions	Multi-component intervention for caregivers and participants based on the social cognitive theory and health promotion.	
Outcome	Moderate to vigorous physical activity measured by steps per day.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Low	Simple randomised design was used and a researcher with no knowledge of the participants performed randomisation. Participant identification numbers were mixed in a basket and chosen to allocate participants into groups.
Allocation concealment	Low	Sealed envelopes with participant identification numbers were used.
Blinding of participants	High	Not blinded due to nature of intervention.
Blinding outcome assessor	Low	Not blinded however primary outcome was an objective outcome measure which is not likely to be influenced by lack of blinding.
Incomplete outcome data	Low	Three out of 33 clusters dropped out after recruitment but before the intervention was implemented. One out of 130 participants dropped out and was subsequently left out of the trial. Missing data unlikely to be related to the outcome.
Selective reporting	Low	Protocol published and outcomes pre-specified.
Other biases	Low	The trial appears to be free of other sources of bias.

Curtin et al. (2013)

Methods	Pilot randomised controlled trial.	
Participants	21 participants aged between 13-26 with Down syndrome and a BMI of equal or greater than 85th percentile were enrolled in the trial.	
Interventions	Nutrition and activity education versus nutrition and activity education with a behavioural intervention.	
Outcome	Change in moderate to vigorous physical activity.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Unclear	Insufficient information provided to permit a judgement of risk.
Allocation concealment	High	Group assignment was made in the order of enrolment by the research coordinator working from a printed list of assignments.
Blinding of participants	High	Participants not blinded due to the nature of the intervention.
Blinding Outcome assessor	High	Outcome assessors were not blinded to group assignment.
Incomplete outcome data	High	4 out of 21 participants dropped out (19%) with unclear reasons for drop-outs.
Selective reporting	Unclear	Insufficient information to determine risk of bias. Unable to locate trial protocol.
Other biases	Unclear	Insufficient information to determine risk of bias.

McDermott et al. (2012)

Methods	Randomised active controlled intervention trial.	
Participants	443 community dwelling adults aged between 19-70 with mild to moderate ID.	
Interventions	'Steps to Your Health' participatory classes.	
Outcome	Change in mean of minutes of moderate to vigorous physical activity.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Unclear	Participants were assigned on random assignment however it is unclear how the process of randomisation took place.
Allocation concealment	Unclear	The authors did not address this domain.
Blinding of participants	High	Participants were not blinded due to the nature of intervention.
Blinding of outcome assessor	Low	Primary outcome was measured objectively and is unlikely to be affected by lack of blinding.
Incomplete outcome data	High	Significant amounts of outcome data were missing. 236 out of 445 participants (53%) dropped out and were subsequently left out of the trial.
Selective reporting	Unclear	Insufficient information provided to determine risk of bias. No protocol was located.
Other biases	Unclear	Unclear if other forms of bias are present.

Melville et al. (2015)

Methods	Cluster randomised controlled trial.	
Participants	102 participants over 18 with any level of ID.	
Interventions	Walk Well program aimed to encourage walking and engagement in physical activity.	
Outcome	Percentage in time in moderate to vigorous physical activity.	
Risk of bias		
Item	Authors' judgement	Description.
Adequate sequence generation	Low	Sequence generation was random by using a computer to generate permuted blocks.
Allocation concealment	low	Used an interactive voice response system that was hosted externally.
Blinding of participants	High	Not blinded due to nature of intervention.
Blinding of outcome assessor	Low	Not blinded, but primary outcome was an objective outcome measure (activity log via an accelerometer), so not likely to be influenced by lack of blinding.
Incomplete outcome data	High	82 out of 102 participants dropped out (20%) and were subsequently left out of the trial.
Selective reporting	Low	Reported pre-specified outcomes. Qualitative outcomes published elsewhere.
Other biases	Low	Appears to be free of other forms of bias.

Shields et al. (2013)

Methods	Randomised controlled trial.	
Participants	68 young people with Down syndrome who were aged between 14-22 and had mild to moderate ID.	
Interventions	Student-led progressive resistance training program versus attention controlled social program.	
Outcome	Physical activity measured as the average vector magnitude activity per minute.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Low	Sequence generation process performed through block randomisation method where participants were considered in blocks of 4, 6 & 8. Order of blocks were generated from a web-based program.
Allocation concealment	Low	Allocation concealment (1:1 allocation) was performed through sequentially numbered, opaque and sealed envelopes.
Blinding of participants	High	Participants were unable to be blinded due to nature of intervention.
Blinding of outcome assessor	Low	Assessments were completed by an assessor blinded to group allocation and had no involvement in recruitment, randomisation or training participants.
Incomplete outcome data	Low	A small amount of missing data occurred for reasons other than that related to the intervention.
Selective reporting	Low	Outcomes were reported as specified in the trial protocol. Two minor variations however were made to data analysis, including the calculation of SMD and using Pearson's r correlation coefficient to improve interpretability of results.
Other biases	Low	This trial appears to be free of other forms of bias.

Shields and Taylor (2015)

Methods	A phase II randomised controlled trial.	
Participants	16 participants aged from 18-35 with Down syndrome.	
Interventions	Mentored physical activity program.	
Outcome	Physical activity measured as vector magnitude per minute.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Low	Sequence generation process performed through block randomisation method where participants were considered in blocks of 4, 6 & 8. Order of blocks were generated from a web-based program.
Allocation concealment	Low	Allocation concealment (1:1 allocation) was performed through sequentially numbered, opaque and sealed envelopes.
Blinding of participants	High	No blinding due to nature of intervention.
Blinding of outcome assessor	Low	Assessments were completed by an assessor blinded to group allocation and had no involvement in recruitment, randomisation or training participants.
Incomplete outcome data	Low	Data missing for 4 participants (3 controls, 1 intervention). Reasons for missing sessions were not related to the intervention, carry forward technique used for missing data.
Selective reporting	Low	Trial protocol not located however the authors reported on results for key outcomes expected to be reported.
Other biases	Low	Free of other forms of bias.

Ulrich et al. (2011)

Methods	Randomised controlled trial.	
Participants	72 participants aged between 8-15 with Down syndrome.	
Interventions	Modified bicycle versus a wait-list control.	
Outcome	Physical activity measured in number of minutes per day spent in moderate to vigorous physical activity.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Unclear	Insufficient information on randomisation and sequence generation process.
Allocation concealment	Unclear	Method of allocation not described and possibly not performed.
Blinding of participants	High	Not blinded due to the nature of the intervention.
Blinding of outcome assessor	High	Although some outcomes were objective (e.g. physical activity via accelerometers), on balance, there is too much room for bias.
Incomplete outcome data	High	11 out of 72 participants (15%) dropped out and were subsequently left out of the trial, plus 15 were not analysed because they did not learn to ride the bike (36% in total not analysed).
Selective reporting	High	Trial registry shows different time points to what was reported.
Other biases	High	Potential conflict of interest regarding funding source and use of fleet bikes however there is insufficient information provided to rule this out.

van Schijndel-Speet et al. (2017)

Methods	Cluster randomised clinical trial.	
Participants	151 participants who were 40 years or older.	
Interventions	Physical activity and education programme versus usual care.	
Outcome	Physical activity measured in steps per day.	
Risk of bias		
Item	Authors' judgement	Description
Adequate sequence generation	Unclear	Person responsible for randomising groups may be aware of treatment allocation. Clients and family were concealed to the allocation, not day centre managers. Investigators/participants not blinded for allocation.
Allocation concealment	High	Person responsible for randomising groups may be aware of treatment allocation. Only clients and family were concealed to the allocation, not day centre managers.
Blinding of participants	High	Blinding of participants not possible due to nature of intervention.
Blinding of outcome assessor	High	Executors of baseline and effect measurements were not blinded to allocation.
Incomplete outcome data	High	Substantial amount of missing data on the primary outcome measure of PA.
Selective reporting	Low	The trial protocol is available, and all pre-specified outcome measured (primary/secondary) have been reported as specified in the protocol however subsets of data used.
Other biases	Unclear	Insufficient information to rule out other forms of bias.

Supplementary file 4 – Definition of adherence to physical activity monitors used across studies.

Trial	Definition of adherence
Angulo-Barroso et al. (2008)	Not reported.
Bergstrom et al. (2013)	Around the waist in line with the knee for seven consecutive days.
Curtin et al. (2013)	Equal or greater than 600 minutes per day (10 hours) on at least three weekdays and one weekend.
McDermott et al. (2012)	To be worn around the waist during all waking hours for five days minimum including two weekend days.
Melville et al. (2015)	All walking hours for seven days, minimum being six hours of data on at least three out of the seven days of use.
Shields et al. (2013)	Four days including one weekend day with at least 10 hours of data a day.
Shields and Taylor (2015)	Four days including one weekend day with at least 10 hours of data a day.
Ulrich et al. (2011)	No minimum time specified, to be used for all activities except water activities.
van Schijndel-Speet et al. (2017)	At least four days of use.

Supplementary file 5 – Description of interventions

Trial	Intervention
Angulo-Barroso et al. (2008)	A 9-month high-intensity interval training program was compared to a low-intensity interval training program on a custom-built treadmill. The high-intensity protocol involved progressive increases to treadmill belt speed, time and ankle weights. The low-intensity protocol did not change during the longitudinal interval of the intervention.
Bergstrom et al. (2013)	A three-component intervention (with a duration of 12 to 16 months) was developed in line with the Social Cognitive Theory and aimed to improve health literacy and behaviour through improving social and physical environments. The three components were: (i) appointment for a health ambassador in each community residence attending network meetings, (ii) a study circle for caregivers and (iii) a health course for residents.
Curtin et al. (2013)	A 9-month intervention was used in two active intervention groups. One group received a 6-month nutrition and activity education intervention, and the other group received a 6-month nutrition and activity education plus a behavioural intervention. The education program allocated to one group was a nutrition and activity education program that taught participants simple nutritional concepts and exercises via verbal instruction, demonstration taste tests and activities. The other group received same nutrition and activity education program, plus the addition of a behavioural intervention that involved sessions with a behavioural specialist who provided instructions on behavioural strategies (i.e. monitoring diet and activity).
McDermott et al. (2012)	The 12-week intervention used, known as the 'Steps to Your Health' classes consisted of eight participatory classes that covered a variety of topics that encouraged moderate to vigorous physical activity and body mass reduction. Topics included nutrition, exercise, thinking patterns, and behaviour management. Sessions were led by a health educator. The control group covered topics relating to hygiene and safety.
Melville et al. (2015)	The 12-week intervention involved physical activity consultations to implement a behaviour change model (encompassing goal-setting, self-efficacy, self-monitoring and mobilising social support for change) and involved carers. The Walk Well intervention was previously used on parents without intellectual disabilities. The aim of the walking program was for participants to gradually increase their daily walking time by thirty minutes.
Shields et al. (2013)	The 10-week intervention used included a progressive resistance training program (using readily available resistance machines) at a local gymnasium. It involved pairing a physiotherapy student as a mentor with a participant to assist them complete the training program and provide support.

Shields and Taylor (2015)	Using an 8-week walking program, participants in the intervention group worked with their student mentor to complete walking sessions together and to plan for an additional session of walking without the student mentor while participants in the control group engaged in social activities with their student mentors.
Ulrich et al. (2011)	Participants were taught how to learn to ride a bicycle (intervention) for one week. The bicycle used was a specially designed, adapted bicycle that provided stability while learning. The bicycle was designed to allow for incremental progress to a two-wheel bicycle and could be altered to suit the needs of the rider. Special rollers could be fitted in place of the rear wheel to facilitate movement that was similar to a two-wheel bicycle. The rollers included a series of eight different sizes that increased in difficulty level. The rollers tapered as the rider progressed in skill, which eventually led to riding a standard two-wheel bicycle.
Van Schijndel-Speet et al. (2017)	There were two components to the 8-month intervention. The first component was an education program (to improve participants' knowledge on physical activity and its health benefits). The education component was inspired by another health promotion program (known as 'Health Matters'), which is developed with experts in educating people with intellectual disabilities and developing appropriate educational content. The second component was a physical activity program (based on guidelines set by the American College of Sports Medicine and the American Heart Association). The physical activity program was set to address fitness components, including strength, endurance, balance and flexibility.

Appendix 2. Online supplementary files associated with published randomised pilot study

Supplementary file 1 – Effect of Clarks® (intervention) casual shoes on gait parameters.

Biomechanical variable	Mean (SD) at baseline Custom-fitted footwear group (n = 17)	Mean (SD) at baseline Control group (n = 16)	Mean (SD) at 12 weeks Custom-fitted footwear group (n = 17)	Mean (SD) at 12 weeks Control group (n = 16)	Adjusted mean difference (95% CI)	*P*-value
Velocity (cm/sec)	111.24 (23.38)	108.28 (17.26)	122.27 (21.90)	112.54 (23.80)	8.10 (-3.71 to 19.90)	0.18
Cadence	123.67 (16.01)	124.83 (16.97)	125.38 (13.76)	125.41 (17.88)	0.71 (-6.81 to 8.23)	0.85
Step time (sec) L[1]	0.49 (0.07)	0.49 (0.06)	0.48 (0.05)	0.49 (0.07)	0.00 (-0.04 to 0.023)	0.79
Step length (cm) L	53.35 (9.12)	52.24 (7.84)	57.58 (10.17)	53.99 (10.04)	2.59 (-1.88 to 7.06)	0.26
Step time (sec) R[2]	0.47 (0.08)	0.49 (0.07)	0.486 (0.05)	0.48 (0.07)	-4.02 (-0.03 to 0.00)	1.00
Step length (cm) R	54.88 (10.73)	52.62 (7.59)	60.08 (9.84)	53.87 (10.6)	4.25 (-0.33 to 8.84)	0.07
Stride length (cm) L	108.45 (19.47)	105.42 (5.11)	117.25 (21.05)	108.33 (23.08)	3.74 (-5.03 to 12.52)	0.39
Stride length (cm) R	108.53 (19.38)	105.14 (15.32)	118.52 (20.27)	108.43 (21.00)	5.17 (-2.83 to 13.17)	0.20
HH[3] base support (cm) L	11.38 (3.14)	9.76 (1.60)	10.17 (2.87)	9.28 (2.85)	-0.12 (-1.79 to 1.56)	0.89
HH base support (cm) R	11.45 (3.07)	9.96 (1.68)	10.11 (2.96)	9.43 (2.76)	-0.14 (-1.82 to 1.54)	0.87
Stance time (sec) L	0.61 (0.11)	0.60 (0.09)	0.59 (0.07)	0.59 (0.10)	0.00 (-0.05 to 0.04)	0.87
Stance time (sec) R	0.61 (0.11)	0.60 (0.89)	0.59 (0.09)	0.60 (0.10)	-0.01 (-0.07 to 0.04)	0.66
Single support time (sec) L	0.38 (0.04)	0.37 (0.05)	0.38 (0.03)	0.38 (0.04)	0.00 (-0.03 to 0.02)	0.70
Single support time (sec) R	0.38 (0.04)	0.38 (0.03)	0.38 (0.03)	0.38 (0.04)	0.00 (-0.02 to 0.02)	0.96
Double support time (sec) L	0.23 (0.08)	0.22 (0.05)	0.21 (0.04)	0.21 (0.06)	-0.01 (-0.04 to 0.02)	0.48
Double support time (sec) R	0.23 (0.08)	0.22 (0.05)	0.21 (0.04)	0.22 (0.06)	-0.01 (-0.04 to 0.02)	0.38
Toe in/out angle L	1.02 (10.09)	2.73 (5.97)	3.89 (8.69)	3.76 (6.52)	1.49 (-1.44 to 4.41)	0.30
Toe in/out angle R	2.08 (7.71)	2.95 (5.92)	4.52 (8.69)	2.75 (6.40)	2.32 (-0.91 to 5.55)	0.16
Step width (cm)	51.22 (7.89)	49.57 (6.20)	54.52 (7.99)	50.15 (8.68)	2.75 (-0.34 to 5.84)	0.08
Stride width (cm)	10.29 (3.04)	9.11 (3.04)	9.11 (2.23)	8.89 (2.30)	-0.24 (-1.75 to 1.28)	0.76

Supplementary file 1 (cont.) – Effect of Clarks® (intervention) school shoe on gait parameters.

Biomechanical variable	Mean (SD) at baseline Custom-fitted footwear group (n = 17)	Mean (SD) at baseline Control (n = 16)	Mean (SD) at 12 weeks Custom-fitted footwear group (n = 17)	Mean (SD) at 12 weeks Control group (n = 16)	Adjusted mean difference (95% CI)	*P*-value
Velocity (cm/sec)	110.57 (21.38)	108.98 (21.96)	115.77 (20.27)	113.48 (25.28)	1.27 (-10.99 to 13.52)	0.84
Cadence	123.42 (15.10)	124.45 (14.60)	124.87 (14.5)	125.91 (15.64)	-0.30 (-7.54 to 6.94)	0.94
Step time (sec) L	0.49 (0.06)	0.50 (0.05)	0.49 (0.05)	0.49 (0.06)	0.00 (-0.04 to 0.04)	0.89
Step length (cm) L	53.64 (9.19)	52.81 (10.21)	58.54 (10.75)	55.00 (10.96)	2.89 (-1.63 to 7.41)	0.21
Step time (sec) R	0.50 (0.06)	0.49 (0.06)	0.49 (0.05)	0.48 (0.06)	0.01 (-0.02 to 0.04)	0.56
Step length (cm) R	54.26 (10.42)	52.73 (10.11)	58.35 (10.13)	54.21 (11.44)	2.94 (-2.59 to 8.47)	0.29
Stride length (cm) L	108.33 (19.40)	105.95 (19.89)	112.01 (20.76)	108.42 (21.76)	1.58 (-8.07 to 11.23)	0.75
Stride length (cm) R	107.87 (19.14)	105.94 (20.21)	113.08 (19.85)	110.19 (22.76)	1.32 (-9.24 to 11.88)	0.80
HH base support (cm) L	10.94 (2.75)	10.34 (1.80)	11.11 (2.69)	9.79 (1.83)	0.99 (-0.30 to 2.28)	0.13
HH base support (cm) R	10.96 (2.83)	10.49 (1.71)	11.19 (2.60)	9.67 (2.10)	1.24 (-0.04 to 2.51)	0.06
Stance time (sec) L	0.61 (0.11)	0.60 (0.08)	0.60 (0.07)	0.59 (0.08)	0.01 (-0.03 to 0.06)	0.53
Stance time (sec) R	0.6109 (0.10)	0.60 (0.08)	0.60 (0.08)	0.60 (0.09)	0.00 (-0.04 to 0.05)	0.95
Single support time (sec) L	0.38 (0.04)	0.37 (0.04)	0.37 (0.03)	0.37 (0.04)	0.00 (-0.02 to 0.02)	0.89
Single support time (sec) R	0.38 (0.03)	0.37 (0.04)	0.37 (1.54)	0.38 (0.04)	0.00 (-0.03 to 0.02)	0.67
Double support time (sec) L	0.23 (0.08)	0.23 (0.04)	0.224 (0.05)	0.22 (0.05)	0.00 (-0.03 to 0.04)	0.84
Double support time (sec) R	0.23 (0.08)	0.23 (0.04)	0.227 (0.05)	0.22 (0.05)	0.00 (-0.03 to 0.04)	0.82
Toe in/out angle L	2.13 (9.21)	2.25 (5.07)	3.48 (8.97)	2.55 (5.73)	1.01 (-2.84 to 4.87)	0.60
Toe in/out angle R	3.56 (7.10)	3.53 (6.04)	4.29 (6.46)	2.92 (6.84)	1.34 (-1.33 to 4.01)	0.32
Step width (cm)	51.01 (7.80)	49.89 (8.08)	53.02 (7.90)	51.02 (8.88)	1.05 (-2.23 to 4.32)	0.53
Stride width (cm)	9.92 (2.54)	9.46 (1.57)	9.77 (2.27)	8.69 (1.74)	0.82 (-0.25 to 1.89)	0.13

*A system-generated error resulted in loss of data for five participants at the 12-week assessment, however multiple imputation was used prior to analysis. A negative adjusted mean difference indicates the result favoured the control group. [1]L = left and [2]R = right. [3]Heel to heel base of support.

Appendix 3. Online supplementary files associated with the published reproducibility study

Supplementary file 1 – 3D foot scan measurement protocol

Software required

The 3D-Tool© Version 13 (3D-Tool GmbH, Weinheim, Germany): https://www.3d-tool.com/

Canvas© 11 software (ACD Systems International, Seattle, WA, USA).

Notes

- All length and width measurements can be measured using 3D-Tool© viewer
- All girth measurements or cross-sections taken will require Canvas©
- Unit of measurement for all dimensions are in mm (both software)

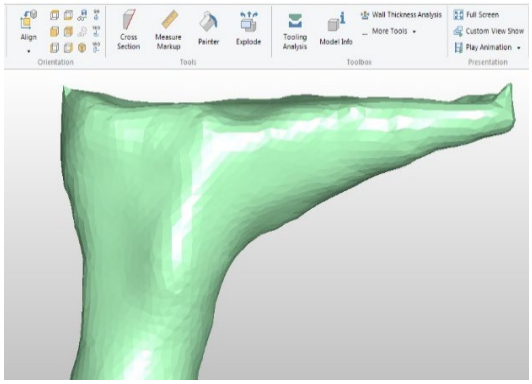
Length

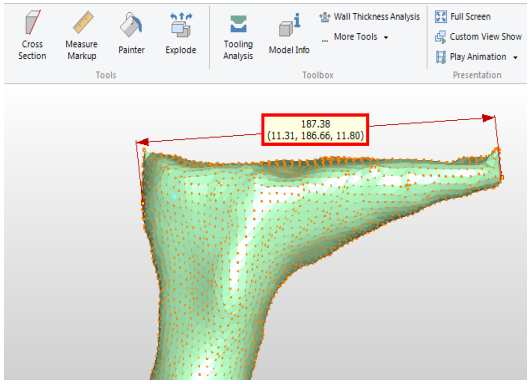
Foot length

Definition

Distance between foot end (heel) and foot tip (anterior point of most protruding toe).

Instructions





To measure length, click on right view under the Align tab. Select measure mark-up and select your landmarks from the pternion to the most protruding toe.

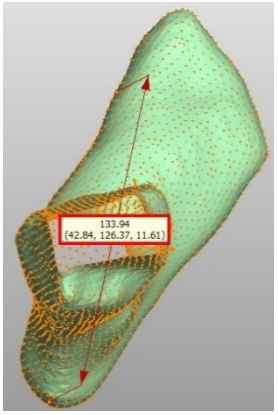
Ball of foot length

Definition

Distance between foot end (heel/pternion) and the 1st metatarsophalangeal protrusion.

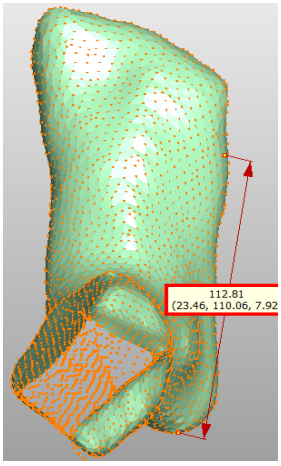
Instructions

To measure ball of foot length, rotate the 3D model to view the medial side of the foot. Select ‘measure mark-up’ and click on the heel from the pternion. Rotate the image to view the planter surface of the model. Select the landmark at the protrusion of the 1st metatarsal. Rotate the model to check the dorsal side of the foot to confirm positioning (see image).



Outside foot length

Definition

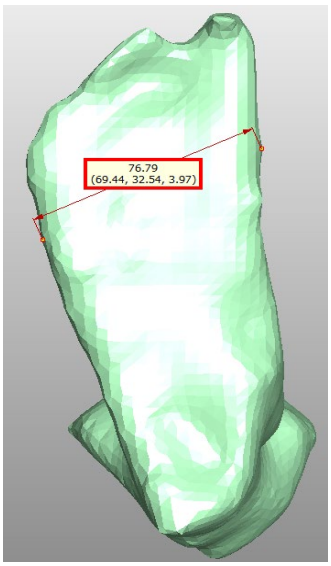


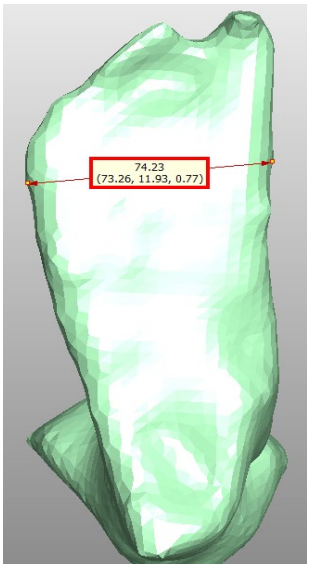
Distance between foot end (heel/pternion) and the fifth metatarsophalangeal protrusion.

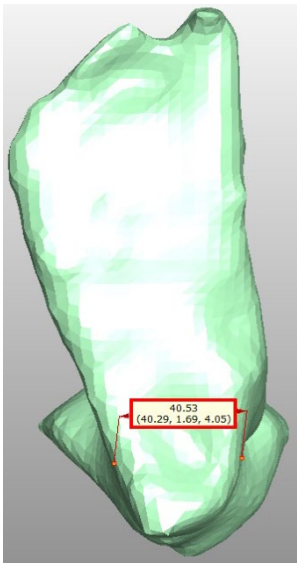
Instructions

Select the same point of the pternion used for foot length and ball of foot length, rotate the model to view the plantar side. Select the point of the protrusion of the 5th metatarsal. Rotate the model to check the dorsal side of the foot to confirm positioning (see image).

Foot width







(A) (B) (C)

(A) Diagonal foot width

Definition

Connecting line between the 1st metatarsophalangeal joint and the 5th metatarsophalangeal joint.

Instructions

Position to view the plantar surface of the foot. Select ‘measure mark-up’ and measure the widest points of the forefoot, at the 1st metatarsal to the 5th metatarsal, following the height of the metatarsal heads.

(B) Horizontal foot width

Definition

Orthogonal connection line starting at the 1st metatarsophalangeal joint to the outside curvature of the foot.

Instructions

Similarly, select the 1st metatarsophalangeal joint and measure to the outside curvature of the foot.

(C) Heel width

Definition

Orthogonal connection line starting on the medial side of the heel to the outside curvature of the heel.

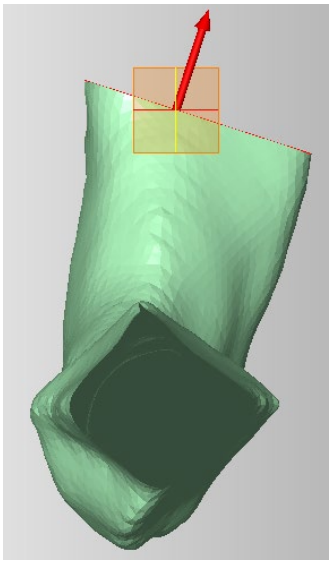
Instructions

Measure the widest part of the heel from the medial to lateral aspect of the heel.

Girth

Ball girth

Definition



Maximum circumference over the first to the fifth metatarsophalangeal joint protrusion.

Instructions

Rotate the model to view the dorsal side of the foot. Select ‘cross section’. Select the ‘XZ-plane’ and drag the arrow to the ball of the foot. Adjust the ‘angle Z’ to follow the metatarsal head positioning. Select the ‘setting button’, select ‘export cross-section as DXF’ and save. You will need to open this file in Canvas© to measure ball girth.

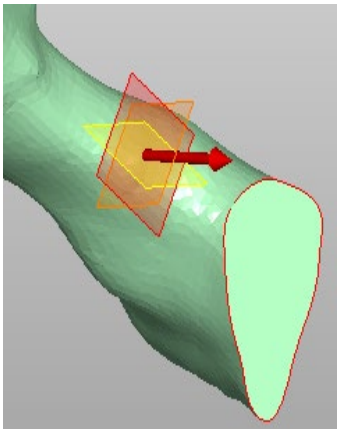
Instep girth

Definition

Measured from the most plantar surface of the foot to the most dorsal aspect of the foot, in alignment with the navicular.

Instructions

Similar to ball girth, drag the arrow to the instep region and adjust the angle to align with the navicular. Rotate the image for the best view of the final area, then export as a DXF to open and measure in Canvas©. For all girth measurements that are exported as a DXF and opened in Canvas©, a dialogue box will allow you to select from a range of options. Ensure settings are in mm, open the file and select the image. The top panel will show the perimeter value.



Height

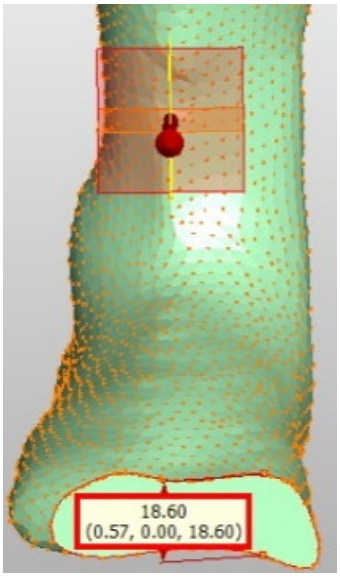
First and fifth toe height

Definition (first toe height)

Maximum height of the hallux measured from the most plantar aspect of the hallux to the most dorsal aspect of the hallux.

Instructions

Position the model to view the medial side. Take a cross-section of the highest point of the digit, reposition the foot model (to view from the front) and measure height.

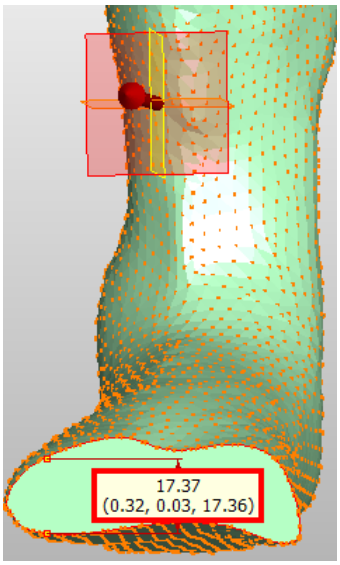


Definition (fifth toe height)

Maximum height of the 5th toe measured from the most plantar aspect of the toe to the most dorsal aspect of the toe.

Instructions

Position the model to view the lateral side. Take a cross-section of the highest point of the digit, reposition the foot model (to view from the front) and measure height.



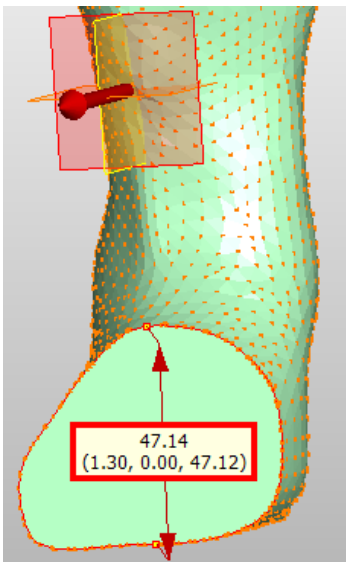
Instep height

Definition

Measured from the most plantar aspect of the foot to the highest dorsal aspect of soft tissue (plantar foot end to the junction of shank and foot dorsum).

Instructions

Rotate the model to view the medial side of the foot. Select ‘cross section’. Select the ‘XZ-plane’ and drag the arrow to the region according to the definition. With this cross-section of the instep height, measure from the most plantar aspect to the most dorsal aspect of soft tissue.

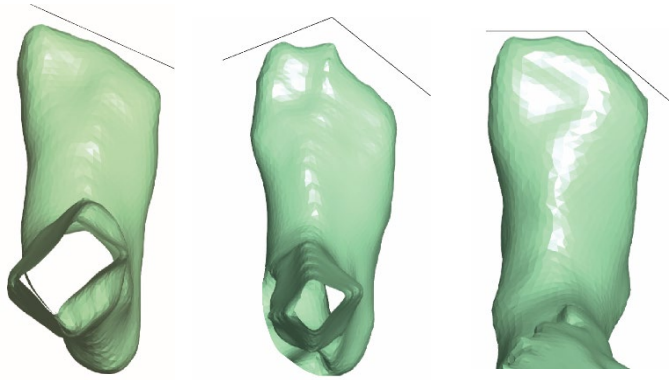


Forefoot shape

The forefoot region of each scan has been categorised into 3 shapes, which reflect the length of toes relative to each other. This section involves evaluating the forefoot and determining which category best suits the shape of the toes for each scan.

Shapes are:

(1) 1st digit is the longest digit (1>2>3>4>5)
(2) 2nd digit is the longest digit (2>1>3>4>5)
(3) 1st and 2nd digits are equal length, and longer than the remaining (1=2>3>4>5)



(1) (2) (3)

Appendix 4. Study registrations (systematic review and pilot study)

Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials

Nirmeen Hassan, Shannon Munteanu, Karl Landorf, Nora Shields

Citation

Nirmeen Hassan, Shannon Munteanu, Karl Landorf, Nora Shields. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. PROSPERO 2016 CRD42016046948 Available from: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42016046948

Review question

What interventions increase physical activity in individuals with intellectual disabilities?

Searches

We will search the following electronic databases: MEDLINE, CINAHL, Cochrane library, SPORTDiscus and EMBASE.
The search strategy will include only terms relating to physical activity and intellectual disabilities. The terms will be combined with the Cochrane MEDLINE filter for randomised controlled trials of interventions. The search terms will be adapted across other databases in combination with database-specific filters for randomised controlled trials where possible. No restrictions will be placed on language or publication period.

Types of study to be included

Only RCTs will be included.

Condition or domain being studied

Physical activity in people (children and adults) with an intellectual disability as a primary diagnosis.

Participants/population

Inclusion: Any age with an intellectual disability. No restrictions on level of intellectual disability or geographical location of individual.

Intervention(s), exposure(s)

Any intervention that aims to improve overall participation in physical activity. Interventions may include but are not limited to structured walking programs, structured exercise programs, health promotion programs, multi component interventions (nutritional and exercise programs) and use of exercise equipment.

Comparator(s)/control

Usual care or groups not expected to have an effect on physical activity e.g. social activities not involving physical activities.

Main outcome(s)

Studies need to include at least one count of physical activity measured objectively, and at least one outcome measuring physical activity objectively (e.g. accelerometers, pedometers, doubly labelled water).

* Measures of effect

Standardised mean differences and associated 95% confidence intervals.

Additional outcome(s)

None.

* Measures of effect

N/A

Data extraction (selection and coding)

Titles and/or abstracts of studies retrieved using the search strategy will be screened independently by two review authors to identify studies that potentially meet the inclusion criteria. The full text of potentially eligible studies will be retrieved and independently assessed for eligibility by two review authors. Any disagreement between the two authors will be resolved through discussion. A modified data extraction form will be used to extract data from the included studies.
Extracted information will include: study setting; study population and participant characteristics at baseline; details of the intervention and control conditions; study methods, including outcome measures and times of measurement.
Two review authors will extract data independently, discrepancies will be identified and resolved through discussion (with a third author where necessary). Missing data will be requested from authors of included studies.

Risk of bias (quality) assessment
Two review authors will independently assess the risk of bias of included studies using the Cochrane Risk of Bias tool. Any disagreement will be discussed between review authors until consensus is reached. A third reviewer will be involved if an agreement cannot be reached.

Strategy for data synthesis
A quantitative synthesis is planned and the means and standard deviations of the outcome measures (assuming continuous scaled data) will be extracted. Standardised mean differences with 95% confidence intervals will be determined. A meta-analysis will be performed where there are multiple studies comparing the effectiveness of a similar intervention at a similar time point.

Analysis of subgroups or subsets
If feasible, sub-group analysis for people with differing levels of intellectual disability or differing medical conditions associated with the disability will be performed.

Contact details for further information
Ms. Hassan
N.Hassan@latrobe.edu.au

Organisational affiliation of the review
Discipline of Podiatry, School of Allied Health, Human Services and Sport, College of Science, Health and Engineering, La Trobe University, Melbourne, Victoria 3086, Australia
http://www.latrobe.edu.au/

Review team members and their organisational affiliations
Ms Nirmeen Hassan. Discipline of Podiatry, School of Allied Health, La Trobe University
Assistant/Associate Professor Shannon Munteanu. Discipline of Podiatry, School of Allied Health, La Trobe University
Professor Karl Landorf. Discipline of Podiatry, School of Allied Health, La Trobe University
Professor Nora Shields. Discipline of Physiotherapy, School of Allied Health, La Trobe University

Type and method of review
Systematic review

Anticipated or actual start date
04 August 2016

Anticipated completion date
01 May 2017

Funding sources/sponsors
This review will not be funded.

Conflicts of interest
None known

Language

English

Country
Australia

Stage of review
Review Completed published

Details of final report/publication(s) or preprints if available
Hassan NM, Landorf KB, Shields N, Munteanu SE. Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials. Journal of Intellectual Disability Research; 2018:63; 168 – 191. doi: https://doi.org/10.1111/jir.12562
https://onlinelibrary.wiley.com/doi/full/10.1111/jir.12562

Subject index terms status
Subject indexing assigned by CRD

Subject index terms
Humans; Intellectual Disability; Motor Activity; Quality of Life

Date of registration in PROSPERO
13 October 2016

Date of first submission
18 June 2020

Stage of review at time of this submission

Stage	Started	Completed
Preliminary searches	Yes	Yes
Piloting of the study selection process	Yes	Yes
Formal screening of search results against eligibility criteria	Yes	Yes
Data extraction	Yes	Yes
Risk of bias (quality) assessment	Yes	Yes
Data analysis	Yes	Yes

Revision note
This record is being updated as the publication is now available online.

The record owner confirms that the information they have supplied for this submission is accurate and complete and they understand that deliberate provision of inaccurate information or omission of data may be construed as scientific misconduct.

The record owner confirms that they will update the status of the review when it is completed and will add publication details in due course.

Versions
13 October 2016
17 October 2016

10 September 2020

Trial Review

COVID-19 sudies are our top priority. For all other trials, there is a 4-week delay in processing a trial submitted to the ANZCTR and additional delays for updates of regisered trials. We appreciate your patience.

Trial regisered on ANZCTR

Regisration number	ACTRN12616001118493
Ethics application satus	Approved

Date submitted	28/07/2016
Date regisered	17/08/2016
Date las updated	1/08/2018
Type of regisration	Prospectively regisered

Titles & IDs

Public title	Do cusomised ft shoes increase physical activity in children with Down syndrome?
Scientifc title	A pilot randomised controlled trial to invesigate if cusomised ft shoes increase the amount of physical activity performed by children and adolescents with Down syndrome
Secondary ID [1]	None
Universal Trial Number (UTN)	
Trial acronym	ShoeFIT
Linked sudy record	

Health condition

Health condition(s) or problem(s) sudied:	
Physical activity in Down syndrome	
Foot health in Down syndrome	
Condition category	**Condition code**
Physical Medicine / Rehabilitation	Other physical medicine / rehabilitation
Human Genetics and Inherited Disorders	Down's syndrome

Study type	Interventional
Description of intervention(s) / exposure	The intervention group will receive two pairs of sandard commerically available shoes (from Clarks Ausralia); one pair for school and one pair for ordinary wear. The shoes will be cusom fitted during a single one-on-one appointment (approximate duration of 1 hour) with a podiatris trained in fitting shoes. The school shoes will be Clarks Laura (females) or Lochie (males) model shoes. Participants that have feet that are too large for the Laura/Lochie syles or prefer lace-up shoes will be provided with Daytona model shoes. Ordinary wear shoes will be Clarks Ventura model (males and females) shoes. Participants that prefer lace-up shoes will be provided with Vancouver model shoes. The duration of the intervention will be 12 weeks. Participants will be expected to use the school shoes when they go to school and use the ordinary wear shoes at other times (i.e. when they are not at school but are wearing shoes). Adherence will be monitored by specially designed quesionnaire that the parents will complete at 6 and 12 weeks.
Intervention code [1]	Treatment: Other
Intervention code [2]	Lifesyle
Intervention code [3]	Treatment: Devices
Comparator / control treatment	The control group (wait lis control) will continue wearing their exising shoes and will then receive two pairs of cusomised fit shoes at the end of the sudy (12 weeks).
Control group	Active

Outcomes

Primary outcome [1]	Physical activity (using an Actigraph activity monitor collected over 7 days)
Timepoint [1]	Baseline and at 6 and 12 weeks. Twelve weeks is the primary end-point
Secondary outcome [1]	Foot-specifc disability (physical domain) using the physical domain of the Oxford Ankle Foot Quesionnaire for Children) - parent reported version
Timepoint [1]	Baseline, 6 and 12 weeks
Secondary outcome [2]	Foot-specifc disability (school and play domain) using the Oxford Ankle Foot Quesionnaire for Children - parent reported version
Timepoint [2]	Baseline, 6 and 12 weeks
Secondary outcome [3]	Foot-specifc disability (emotional domain) using the Oxford Ankle Foot Quesionnaire for Children - parent reported version
Timepoint [3]	Baseline, 6 and 12 weeks
Secondary outcome [4]	Acceptability of the cusom fit shoes (experimental group) via semi-sructured interview of the participants and carers/parents
Timepoint [4]	12 weeks
Secondary outcome [5]	Walking velocity (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom fitted shoes (both ordinary wear and school shoes)
Timepoint [5]	Baseline and 12 weeks
Secondary outcome [6]	Cadence (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom fitted shoes (both ordinary wear and school shoes)
Timepoint [6]	Baseline and 12 weeks
Secondary outcome [7]	Stride length (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom fitted shoes (both ordinary wear and school shoes)
Timepoint [7]	Baseline and 12 weeks
Secondary outcome [8]	Step length (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom fitted shoes (both ordinary wear and school shoes)
Timepoint [8]	Baseline and 12 weeks
Secondary outcome [9]	Base of support (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom fitted shoes (both ordinary wear and school shoes)
Timepoint [9]	Baseline and 12 weeks
Secondary outcome [10]	Toe in/out angle (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom fitted shoes (both ordinary wear and school shoes)

Timepoint [10]	Baseline and 12 weeks
Secondary outcome [11]	Step width (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom ftted shoes (both ordinary wear and school shoes)
Timepoint [11]	Baseline and 12 weeks
Secondary outcome [12]	Adverse events (such as new pains in the body, rolled ankles, blisers, swelling) will be assessed via survey completed by the parents/next of kin. The survey has been specifcally designed for this sudy.
Timepoint [12]	6 and 12 weeks
Secondary outcome [13]	Adherence to the intervention (experimental group only) will be determined by survey of parents/next of kin. Parents of the participants will provide information regarding the number of hours per day and number of days they have worn their shoes during the previous 6 weeks. The survey has been specifcally designed for this sudy.
Timepoint [13]	6 and 12 weeks
Secondary outcome [14]	Use of co-interventions (such as new shoes that have not been prescribed in this sudy , visits to health care providers for treatment of any foot conditions) will be assessed via a survey. The survey has been specifcally designed for this sudy.
Timepoint [14]	6 and 12 weeks
Secondary outcome [15]	Stride width (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom ftted shoes (both ordinary wear and school shoes)
Timepoint [15]	Baseline and 12 weeks
Secondary outcome [16]	Single leg support time (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom ftted shoes (both ordinary wear and school shoes)
Timepoint [16]	Baseline and 12 weeks
Secondary outcome [17]	Double leg support time (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom ftted shoes (both ordinary wear and school shoes)
Timepoint [17]	Baseline and 12 weeks
Secondary outcome [18]	Stance time (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom ftted shoes (both ordinary wear and school shoes)
Timepoint [18]	Baseline and 12 weeks
Secondary outcome [19]	Step time (using the GAITRite sysem) wearing the pre-sudy entry shoes and cusom ftted shoes (both ordinary wear and school shoes)
Timepoint [19]	Baseline and 12 weeks

Eligibility

Key inclusion criteria	Children and adolescents with Down syndrome will be eligible for inclusion in the trial if they are aged between 5 and 17 years and have the ability to follow simple verbal insructions in English.
Minimum age	5Years
Maximum age	17Years
Gender	Both males and females
Can healthy volunteers participate?	Yes
Key exclusion criteria	Participants will be excluded from the trial if there is an acute or subacute medical reason why their physical activity levels might be afected on an on-going basis (e.g. previous lower limb surgery , unable to walk without a supportive device such as a walker or brace), or a concomitant medical condition or injury that could afect their physical function (e.g. neurological or infammatory disorder). Participants will also be excluded from the trial after the initial assessment if they cannot adhere to wearing a monitor to measure the amount of physical activity they do for at leas 10 hours on at leas 4 days including at leas one weekend day .

Study design

Purpose of the sudy	Treatment
Allocation to intervention	Randomised controlled trial
Procedure for enrolling a subject and allocating the treatment (allocation concealment procedures)	Participants will be randomly allocated to one of two groups after baseline assessment using sealed opaque envelopes. The sealed envelopes will be prepared beforehand by a researcher with no other involvement in participant recruitment, allocation or assessment.

Methods used to generate the sequence in which subjects will be randomised (sequence generation)	Permuted block randomisation (with random block sizes)
Masking / blinding	Blinded (masking used)
Who is / are masked / blinded?	The people adminisering the treatment/s The people assessing the outcomes The people analysing the results/data
Intervention assignment	Parallel
Other design features	
Phase	Not Applicable
Type of endpoint(s)	Efcacy
Statisical methods / analysis	Statisical analysis will be completed using SPSS (IBM Corp, NY, USA). Demographic characterisics and baseline data will be summarized by descriptive satisics. Data on outcome measures will be analysed on an intention-to-treat basis. Multiple imputation will be used to replace any missing data using fve iterations, with age, baseline scores, and group allocation as predictors. Groups will be compared on primary and secondary outcomes at week 6 and 12. Continuously-scored outcome measures will be analysed using analysis of covariance with the intervention group and baseline scores entered as independent variables. Efect sizes and associated 95% confdence intervals will be calculated for each outcome. A sample size calculation for a fully powered randomised controlled trial will also be completed. Acceptability data from semi-sructured interviews will be analysed using thematic analysis by two independent assessors and fnal themes will be agreed by consensus. Statisical signifcance for hypothesis tess will be set at the conventional level of a = 0.05.

Recruitment satus	Completed

Date of frs participant enrolment			
Anticipated	23/08/2016	**Actual**	21/09/2016

Date of las participant enrolment			
Anticipated	29/09/2017	**Actual**	8/09/2017

Date of las data collection			
Anticipated	22/12/2017	**Actual**	20/12/2017

Sample size					
Target	30	**Accrual to date**		**Final**	33

Recruitment in Ausralia	
Recruitment sate(s)	VIC

Funding & Sponsors

Funding source category [1]	University
Name [1]	La Trobe University. Sport, Exercise and Rehabilitation Research Focus Area Grant Scheme 1 – Pilot Project
Address [1]	La Trobe University Melbourne VIC 3086
Country [1]	Ausralia
Primary sponsor type	Individual
Name	Shannon Munteanu
Address	School of Allied Health,

	La Trobe University, Melbourne VIC 3086
Country	Ausralia
Secondary sponsor category [1]	Individual
Name [1]	Nora Shields
Address [1]	School of Allied Health, La Trobe University, Melbourne VIC 3086
Country [1]	Ausralia
Other collaborator category [1]	Individual
Name [1]	Hylton Menz
Address [1]	School of Allied Health, La Trobe University, Melbourne VIC 3086
Country [1]	Ausralia
Other collaborator category [2]	Individual
Name [2]	Nicholas Taylor
Address [2]	School of Allied Health, La Trobe University, Melbourne VIC 3086
Country [2]	Ausralia
Other collaborator category [3]	Individual
Name [3]	Angela Evans
Address [3]	School of Allied Health, La Trobe University, Melbourne VIC 3086
Country [3]	Ausralia
Other collaborator category [4]	Individual
Name [4]	Cylie Williams
Address [4]	Faculty of Medicine, Nursing & Health Sciences, Monash University, Wellington Rd & Blackburn Rd, Clayton VIC 3800
Country [4]	Ausralia

Ethics approval

Ethics application satus	Approved
Ethics committee name [1]	La Trobe University Human Ethics Committee
Ethics committee address [1]	La Trobe University Melbourne VIC 3086
Ethics committee country [1]	Ausralia
Date submitted for ethics approval [1]	30/03/2016
Approval date [1]	19/05/2016
Ethics approval number [1]	HEC16-027

Summary

Brief summary	The aim of this sudy is: To conduct a pilot randomised controlled trial to evaluate if wearing cusomised ft shoes increases physical activity among children and adolescents with Down syndrome.

	The research quesions are: In children with Down syndrome: * Does improving shoe ft increase physical activity? * Does improving shoe ft improve foot specifc disability, and walking patterns? * Are cusomised ft shoes acceptable? The sudy methods are: We will conduct a pilot randomised trial to compare cusomised ft shoes (intervention) to a waitlis (control) group for children with Down syndrome. Thirty participants with Down syndrome aged 5 to 17 years will be recruited, and randomly allocated to one of two groups. The intervention group (15 children) will receive two pairs of cusomised ft shoes (from Clarks Ausralia): one pair for school and one pair for ordinary wear. The control group (15 children) will continue wearing their exising shoes for 12 weeks (and will then receive two pairs of cusomised ft shoes). Participants will be assessed at the sart of the sudy (baseline/week 0), then at 6, and 12 weeks. Four outcomes will be measured: (1) physical activity (using an activity monitor collected over 7 days) (main outcome), (2) computerised gait analysis, (3) foot-specifc disability using a quesionnaire called the Oxford Ankle Foot Quesionnaire for Children, and (4) acceptability of the intervention (using an interview approach).
Trial website	
Trial related presentations / publications	
Public notes	

Contacts

Principal invesigator	
Name	Dr Shannon Munteanu
Address	School of Allied Health, La Trobe University Melbourne VIC 3086
Country	Ausralia

Phone	+61 3 94795866
Fax	
Email	s.munteanu@latrobe.edu.au

Contact person for public queries	
Name	Dr Shannon Munteanu
Address	School of Allied Health, La Trobe University Melbourne VIC 3086
Country	Ausralia
Phone	+61 3 94795866
Fax	
Email	s.munteanu@latrobe.edu.au

Contact person for scientifc queries	
Name	Dr Shannon Munteanu
Address	School of Allied Health, La Trobe University Melbourne VIC 3086
Country	Ausralia
Phone	+61 3 94795866
Fax	
Email	s.munteanu@latrobe.edu.au

Summary results

Have sudy results been published in a peer-reviewed journal?	

Other publications

Have sudy results been made publicly available in another format?	

Results – basic reporting Results – plain English summary	

Appendix 5. Author contributions

This thesis includes 3 studies that have been published with several co-authors, and one manuscript included as a chapter in this thesis. For each study, I was the first author and was the major contributor of the article. Below is an outline of my contributions for each piece of work.

Systematic review (Chapter 2)

I was involved in the concept and design of the systematic review.

I registered this review with PROSPERO.

I created the relevant search terms and applied these terms to conduct the search. I screened and selected all the relevant articles to be included in the review.

I completed the risk of bias assessment using a risk of bias tool and extracted the relevant data. I completed the data analysis using Revman software.

I drafted the final manuscript, prepared all tables and figures and worked in conjunction with my co-authors to finalise the article for publication.

Randomised pilot study (Chapter 3)

I recruited all participants included in the randomised pilot study and completed all initial and baseline assessments for all participants.

I completed all the progress reports required for ethics throughout the duration of the trial.

I collected and extracted all the data and performed the data analysis using SPSS software.

I wrote the manuscript and prepared all tables and figures. I worked in conjunction with my co-authors to finalise the article for publication.

Reproducibility study (Chapter 4)

I was involved in the concept and the design of the study.

I collected and extracted all the data and performed the data analysis using SPSS software.

I wrote the manuscript and prepared all tables and figures. I worked in conjunction with my co-authors to finalise the article for publication.

Cross-sectional observational study (Chapter 5)

I completed the ethics application for this study. I created all the relevant documentation required for this application (recruitment flyers, Participant/Parent Information Statements, consent forms and assessment forms).

I recruited all participants included in the cross-sectional observational study and completed all assessments for participants.

I completed all the progress reports required for ethics as required.

I collected and extracted all the data and performed the data analysis using SPSS software.

I wrote the manuscript and prepared all tables and figures. I worked in conjunction with my co-authors to finalise the article for publication.

Appendix 6. Ethical approval statements

Nora Shields

From: ResearchMasterEthics@latrobe.edu.au
Sent: Thursday, 19 May 2016 9:48 AM
To: ResearchMasterEthics; Nora Shields
Cc: Angela Evans; Hylton Menz; Nicholas Taylor; Shannon Munteanu; cylie.williams@Monash.edu
Subject: Application HEC16-027 (Finalised - Approved) - Application finalised as Approved

Dear Nora Shields,

The following project has been assessed as complying with the National Statement on Ethical Conduct in Human Research. I am pleased to advise that your project has been granted ethics approval and you may commence the study.

Application ID: HEC16-027
Application Status/Committee: Finalised - Approved

Project Title: Do custom fitted shoes change physical activity in children with Down syndrome?
Chief Investigator: Nora Shields
Other Investigators: Cylie Williams, Nicholas Taylor, Angela Evans, Shannon Munteanu, Hylton Menz

Date of Approval: 19/05/2016
Date of Ethics Approval Expiry: 22/12/2017

The following standard conditions apply to your project:

- Limit of Approval. Approval is limited strictly to the research proposal as submitted in your application.

- Variation to Project. Any subsequent variations or modifications you wish to make to your project must be formally notified for approval in advance of these modifications being introduced into the project.

- Adverse Events. If any unforeseen or adverse events occur the Chief Investigator must immediately notify the UHEC immediately. Any complaints about the project received by the researchers must also be referred immediately to the UHEC.

- Withdrawal of Project. If you decide to discontinue your research before its planned completion, you must inform the relevant committee and complete a Final Report form.

- Monitoring. All projects are subject to monitoring at any time by the University Human Ethics Committee.

- Annual Progress Reports. If your project continues for more than 12 months, you are required to submit a Progress Report annually, on or just prior to 12 February. The form is available on the Research Office website. Failure to submit a Progress Report will mean approval for this project will lapse.

- Auditing. An audit of the project may be conducted by members of the UHEC.

- Final Report. A Final Report (see above address) is required within six months of the completion of the project.

You may log in to ResearchMaster (https://rmenet.latrobe.edu.au) to view your application.

If you have any further questions, please contact the:
UHEC at humanethics@latrobe.edu.au
SHE College Human Ethics Sub-Committee at chesc.she@latrobe.edu.au ASSC College Human Ethics Sub-Committee at chesc.assc@latrobe.edu.au

HUMAN RESEARCH ETHICS APPLICATION FORM: DECLARATION FOR EXTERNAL INVESTIGATOR

This document must be submitted as an attachment to the online Human Research Ethics Application Form in ResearchMaster. All investigators who are not current staff or students of La Trobe University are required to complete this declaration form.

RESEARCH PROJECT	
Project Title	Do custom fitted shoes change physical activity in children with Down syndrome?
Chief Investigator	Dr Shannon Munteanu

DECLARATION

In preparing this application I/we, the undersigned, declare that I/we:

- have read and agree to abide by the La Trobe University Human Research Ethics Guidelines;
- have read and agree to abide by the conditions and constraints of the National Statement on Ethical Conduct in Human Research (2007) and any other relevant University and/or statutory requirements;
- accept responsibility for the accuracy of the information provided in this application and for the conduct of this research, in accordance with the principles contained in the NHMRC Guidelines and any other conditions specified by the University Human Ethics Committee;
- will ensure that the qualifications and / or experience of all personnel involved with the project are appropriate to the procedures performed;
- will ensure that appropriate permits from relevant external organisations, or State or Federal agencies will be obtained, that copies will be lodged with the UHEC and that any imposed conditions will be observed;
- understand that the information contained in this application is given on the basis that it remains confidential in accordance with relevant University and statutory requirements;
- abide by the terms and conditions set by the University Human Ethics Committee;
- certify that the information contained in this application is true and accurate;
- will seek approval for modifications to the research prior to their implementation.

SIGNATURE			
Name	Cylie Williams	**Date**	14/03/16
Signature			

Down Syndrome Victoria
Supporting Victorian families since 1978

12th April 2016

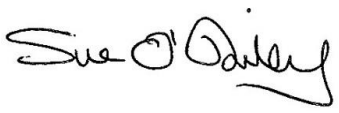
Prof Nora Shields,
Department of Physiotherapy,
La Trobe University,
Bundoora,
Victoria 3086.

Dear Nora,

Re: Benefits of custom fitted shoes in children with Down syndrome

The Down syndrome Association of Victoria (DSAV) have reviewed your research application and, subject to approval from La Trobe University Ethics Committee, we agree in principle to allow you to send project flyers to relevant members.

Yours sincerely,



Sue O'Riley
Executive Officer
Down Syndrome Association of Victoria

Down Syndrome Association of Victoria Inc.
18/71 Victoria Crescent Abbotsford VIC 3067 t: (03) 9486 9600 f: 03 9486 9601
e: info@dsav.asn.au w: downsyndromevictoria.org.au
Reg No A0008787R ABN 59 901 963 154

HEC19290 - New Application - Approved

HE **Human Ethics <humanethics@latrobe.edu.au>**
12/08/2019 12:15 PM

To: Shannon Munteanu Cc: NIRMEEN HASSAN; Andrew Buldt; Karl Landorf; Nora Shields

** This is an automatically generated email, please do not reply. Contact details are listed below.**

Dear Shannon Munteanu,

The following project has been assessed as complying with the National Statement on Ethical Conduct in Human Research. I am pleased to advise that your project has been granted ethics approval and you may commence the study.

Application ID: HEC19290
Application Status/Committee: University Human Ethics Committee

Project Title: Differences in foot anthropometry and foot health in children and adolescents with and without Down syndrome

Chief Investigator: Shannon Munteanu

Other Investigators: Karl Landorf, Andrew Buldt, Nora Shields, Ms Nirmeen Hassan

Date of Approval: 12/08/2019
Date of Ethics Approval Expiry: 12/08/2024

The following standard conditions apply to your project:

- Limit of Approval. Approval is limited strictly to the research proposal as submitted in your application.

- Variation to Project. Any subsequent variations or modifications you wish to make to your project must be formally notified for approval in advance of these modifications being introduced into the project.

- Adverse Events. If any unforeseen or adverse events occur the Chief Investigator must notify the UHEC immediately. Any complaints about the project received by the researchers must also be referred immediately to the UHEC.

- Withdrawal of Project. If you decide to discontinue your research before its planned completion, you must inform the relevant committee and complete a Final Report form.

- Monitoring. All projects are subject to monitoring at any time by the University Human Ethics Committee.

- Annual Progress Reports. If your project continues for more than 12 months, you are required to submit a Progress Report annually, on or just prior to 12 February. The form is available on the

Research Office website. Failure to submit a Progress Report will mean approval for this project will lapse.

- Auditing. An audit of the project may be conducted by members of the UHEC.

- Final Report. A Final Report (see above address) is required within six months of the completion of the project.

You may log in to ResearchMaster (https://rmenet.latrobe.edu.au) to view your application.

Should you require any further information, please contact the Human Research Ethics Team on:
T: +61 3 9479 1443| E: humanethics@latrobe.edu.au.

Warm regards,

Human Research Ethics Team
Ethics, Integrity & Biosafety, Research Office

LA TROBE UNIVERSITY **Research Data Management Planning Tool**

DMP Portal Home You are logged in as Rachel Saltry | Logout

La Trobe Home > DMP Portal Dashboard

System notifications

5 low priority message(s) show me »

Actions
- Object History
- Reharvest
- Reindex
- View Solr Index
- Edit Workflow: Complete
- View Audit Trail

Attachments

Differences in foot anthropometry and foot health in children and adolescents with and without Down syndrome

Project overview

Project title	Differences in foot anthropometry and foot health in children and adolescents with and without Down syndrome
Ethics Number	
FOR Codes	119999 - Medical and Health Sciences not elsewhere classified
College	SHE

Edit this plan
Download a PDF of this plan
Previous versions

Show sections individually

Data Identification, Collection & Planning

Data Types	Consent to collect	Collection Method	Source Data Identifiability Classification	Consent for future use of data (as outlined in PICF)
Physical	Prospective Do you require a waiver of consent? No	We will collect data that will be in hard-copy format. We will have printed questionnaires, assessment forms, take shoe tracings and foot prints using carbon paper.	Re-identifiable	Extended
Digital online tools	Prospective Do you require a waiver of consent? No	3D foot scans will be taken for each participant. The scanner is connected to a computer with a remote drive.	Re-identifiable	Extended

Data Storage & Destruction

Data Types	In what format will data be stored?	How is data stored securely?	Who has access to the data & are there any conditions/restriction to access?	Campus/Location	Building/Server Name	Storage period
Physical	Re-identifiable	Researchers listed on the ethics application will have access to the data. Restrictions to access include hard copies of collected data being stored in a lockable cabinet at all times, which will initially be located in the Research room (1.14) at the Health Sciences Clinic. This room has restricted access to researchers only. Hard copies will then be transferred to another lockable file cabinet in a researcher's office (room 523a, Health Sciences 3 building). Data will be compiled electronically and stored on the research drive (server managed by ICT) on password-protected computers, which is only accessible by researchers involved in the project.	Other	La Trobe University, Melbourne campus.	Health Sciences Clinic (Research room, room number 1.14), Health Sciences 3 building (room number 523a). Sever name: research drive (RDCS\PROJECTS\SHE\DownSyndromeFootHealthProject)	Research with Health information: 7 years post publication
Digital	Re-identifiable	Researchers listed on the ethics application will have access to the data. 3D scans will be stored on a computer with a remote drive. Data will then be compiled electronically and stored on the research drive (server managed by ICT) on password-protected computers, which is only accessible by researchers involved in the project.	password	La Trobe University, Melbourne campus.	Health Sciences Clinic (Research room, room number 1.14) on a remote drive. Sever name: research drive (RDCS\PROJECTS\SHE\DownSyndromeFootHealthProject).	Research with Health information: 7 years post-publication

Publication & dissemination

Describe any procedures or plans for verifying or cleaning data.
We will visually check for errors and run basic statistical tests to identify outliers. The data will be reviewed by more than one researcher to verify the data.

Describe any documentation you will use to inform project personnel and/or secondary users about project methods, data collection and data preparation.
Data is compiled in an excel sheet, where data is clearly labelled. Once data is moved into SPSS (statistical analysis software), each variable that is coded is labelled in detail in order for others to understand.

Post-project

Describe the protocols for organisation of data and records (for example digital folder, directory structures, physical filing systems). Post project.
Post project, all data will be compiled electronically and located in relevant sub-folders within the research drive. All hard copies will be filed away in a locked cabinet. Electronic data will be stored in RDCS\PROJECTS\SHE\DownSyndromeFootHealthProject.

What descriptive record will be created for the data to aid discovery and retrieval?
Mediated access via Figshare (research data La Trobe), La Trobe's collaborative digital repository

College
Figshare (research data.latrobe), La Trobe's collaborative digital repository

Will the research data be shared with other researchers once the project is complete? Explain how.

Mediated access (controlled access to physical or research data is allowed)

Intellectual property, copyright and ownership

Latrobe University. Date copyright was implemented

Other. Date copyright was implemented

Is the data owned by a third party?

No

Data ownership type

Purchase

What are the legal requirements for pre-existing data?

Are there additional requirements for the data?

No

Data ownership type

What are the legal requirements for pre-existing data?

Where are the pre-existing data located?

Are there additional requirements for the data?

Appendix 7. Forms associated with the randomised pilot study

LA TROBE UNIVERSITY

Participants Wanted!

Custom shoe fitting in young people with Down syndrome

We want to find out if custom fitted shoes are useful for young people with Down syndrome.
WE WOULD LIKE YOU TO HELP US!

What is involved?
Your child or adolescent will be allocated at random to one of two groups. One group will receive two pairs of custom fitted shoes immediately, and the second group will be asked to continue wearing their regular shoes for 12 weeks after which they will receive two pairs of customised fitted shoes.

We will ask all participants to complete a range of outcomes at the start of the study, after 6 weeks, and again 12 weeks later including assessments of their footwear, foot disability, physical activity, foot health and walking.

How much will it cost?
We will cover the cost of the two pairs of new Clarks shoes and will contribute a small amount towards the cost of your travel associated with the project.

Who will be taking part?
Young people with Down syndrome aged 5-17 years.

Who is organising this study?
Dr Shannon Munteanu, Dr Angela Evans, and Prof Hylton Menz are research podiatrists at La Trobe University. Dr Cylie Williams is a paediatric podiatrist and adjunct research fellow at Monash University. Prof Nora Shields and Prof Nick Taylor are research physiotherapists at La Trobe University.

Interested?
If you are interested in taking part or you have any questions regarding this study please contact Dr Shannon Munteanu (9479 5866 or s.munteanu@latrobe.edu.au) or Prof Nora Shields (03 9479 5852 or N.Shields@latrobe.edu.au).

College of Science Health and Engineering
School of Allied Health

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

PARTICIPANT INFORMATION STATEMENT (PARENT OR GUARDIAN)

PROJECT TITLE: Do custom fitted shoes change physical activity in children with Down syndrome?

INVESTIGATORS:

Dr Shannon Munteanu is senior lecturer in podiatry in the School of Allied Health at La Trobe University

Professor Nora Shields is the Professor of Clinical and Community Practice in the School of Allied Health at La Trobe University and Northern Health.

Dr Angela Evans is a paediatric podiatrist and a senior lecturer in the School of Allied Health at La Trobe University.

Dr Cylie Williams is a paediatric podiatrist and an adjunct Research Fellow at Monash University

Professor Hylton Menz is a NHMRC Senior Research Fellow in the School of Allied Health at La Trobe University

Professor Nicholas Taylor is the Professor of Allied Health in the School of Allied Health at La Trobe University and Eastern Health.

Ms Nirmeen Hassan is a podiatrist and Master of Research student in the School of Allied Health at La Trobe University.

Dr Andrew Buldt is a podiatrist and post-doctoral researcher in the School of Allied Health at La Trobe University.

ABOUT THE PROJECT:

Your child or adolescent is invited to take part in a project to see if custom fitted shoes changes the amount of physical activity young people with Down syndrome do. This project will test if young people with Down syndrome who wear custom fitted shoes and then wear the shoes regularly for 12 weeks do more physical activity than young people with Down syndrome who wear their own shoes.

WHO CAN TAKE PART?:

Your child is being asked to take part because they are aged 5-17 years and have Down syndrome.

Children with Down syndrome can take part in this project you need to be able to follow short instructions in English.

Children with Down syndrome will not be allowed to take part in this project if there is some reason why you cannot be active such as having an operation on their leg or needing to wear to brace to walk.

WHAT YOUR CHILD WILL BE ASKED TO DO:

If you agree for your child or adolescent to take part, they will be randomly put into either a group that will receive two pairs of custom fitted shoes (one pair of school shoes and one pair of casual shoes) or a group that will continue to wear their own shoes for 12 weeks (the control group).

All children taking part will be asked to do some tests at four time points:

- before the start of the project (initial assessment),
- two-weeks after their initial assessment (baseline assessment, week 0),
- 6-weeks from baseline (mid-way assessment, week 6) and
- at the end of the project (follow-up assessment, week 12).

If your child or adolescent has been randomly put into the control group (wear their usual shoes for 12 weeks), they will have an opportunity to receive two pairs of custom fitted shoes after they have finished all assessments.

TESTS:

Your child or adolescent will be asked to do some tests.

The tests will be done at La Trobe University, at the Melbourne campus in Bundoora, and will take about 90 minutes.

You will need to get your child or adolescent or young adult to the place where the tests are done.

We will pay $30 towards the cost of getting to the testing place and will give your adolescent or young adult that money on the day of the test. The exception is the midway test (week 6) which is done at home.

1. Wearing a little monitor: your child or adolescent will be asked to wear a little monitor for 8 days. It will measure the amount of movement your child does.

You child or adolescent will wear this monitor on their waist using a belt. You need to wear the monitor from when they get up in the morning until they go to bed at night.

If your child or adolescent is not able to wear the monitor for 10 hours each day for four days including either a Saturday or a Sunday, they cannot take part in the project.

Dr Shannon Munteanu will arrange for you and your child to be seen at the Health Sciences Clinic, La Trobe University to advise you on how to deal with the problem and determine the need for further referral, if any.

This project has been approved by the University Human Ethics Committee at La Trobe University. Clarks Australia is supporting this project by providing the shoes that will be prescribed by the podiatrists for free. This project has been given money from the Sport, Exercise and Rehabilitation Research Focus Area at La Trobe University ($24,137).

STORING INFORMATION:

All information we collect as part of this project will be confidential.
If your child or adolescent takes part in the study they will be given a code number, which we will use when entering their information on the computer. Their information will be stored during the project in a lockable filing cabinet in the Podiatry Research room at the La Trobe University Health Sciences Clinic. Data will be put on an electronic file on a computer that needs a password to use it.

Although the researchers will know who your child or adolescent is during the project, their name will not be put included in anything we write about the project. Their identity will remain confidential.

After the project is finished, the computer records and written forms will be transported to a lockable filing cabinet in the locked office of the principal researcher (Dr Shannon Munteanu) at La Trobe University (Room 540, Health Sciences Building 3). No one apart from the researchers will have access to data. At the end of the study a summary of the results will be kept as a computer file on the La Trobe University server. Data will be kept for fifteen years after publication of the results of the study and then it will be destroyed.

RESULTS OF THE STUDY:

When the study is finished, the researchers will send a report about the results to everyone who took part in the study. If your child or adolescent wants a copy of their results they will be given these.

The results of this project may appear in journal publications and in presentations at conferences but participants in the study will not be named.

WITHDRAWING FROM THE STUDY:

Your child or adolescent can stop taking part in the project at any time. If they change their mind, you can request for them to stop being part of the project, provided you tell us within four weeks of having finished

taking part in the project. Once we receive your request, we will destroy all data we have about your children or adolescent. To make this request, you will need to complete and sign a Revocation of Consent Form and send it to Dr Shannon Munteanu in the School of Allied Health at La Trobe University, Bundoora, VIC, 3086. You may only make this request if you change your mind within four weeks of finishing the project.

Taking part is purely voluntary and there are no disadvantages, penalties or adverse consequences for not taking part in or from stopping taking part in the research early.

QUESTIONS OR COMPLAINTS?

If you have any questions about this project you can telephone Dr Shannon Munteanu at La Trobe University, on (03) 9479 5866. If you have any complaints or questions that the researchers have not been able to answer to your satisfaction, you may contact the Senior Human Ethics Officer, Ethics and Integrity Research Office, La Trobe University, Victoria 3086 by telephone 03 9479 1443 or by e-mail humanethics@latrobe.edu.au.

Please quote the application reference number HEC16-027.

College of Science Health and Engineering
School of Allied Health

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

CONSENT FORM (PARENT OR GUARDIAN)

Do custom fitted shoes change physical activity in children with Down syndrome?

I _____have read (or, where appropriate, have had read to me) and understood the information above and any questions I have asked have been answered to my satisfaction.

I agree for my child or adolescent to take part in the project, realising that I may withdraw my child or adolescent from the study at any time. I know that I may ask that all traces of my child or adolescent's participation be removed from the project records up to four weeks after taking part in the project

I agree that the research data provided by my child or adolescent or with my permission during the project may be presented at conferences and published in journals on the condition that neither my child or adolescent's name nor any other identifying information is used.

Child or adolescent's name (block letters):_____

Name of parent (block letters):_____

Signature:_____ Date: _____

Name of researcher (block letters):_____

Signature:_____ Date: _____

ASSENT FORM (ADOLESCENT)

Do custom fitted shoes change physical activity in children with Down syndrome?

I _____have read (or someone else has read it to me) and understood the information above, and any questions I have asked have been explained to me so that I understand.

I agree to take part in the project, but I know that I may stop taking part in the study at any time and may ask that my name and all information about me or collected from me to be removed from the project records up to four weeks after I have finished the program.

I agree that the information provided by me during the project may be included in written reports and articles that might be presented at conferences or written in journals but that my name nor any other information that might identify me will not be used.

Name of participant (block letters):_____

Signature:_____ Date: _____

Name of researcher (block letters):_____

Signature:_____ Date: _____

LA TROBE UNIVERSITY
College of Science Health and Engineering
School of Allied Health

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

REVOCATION OF CONSENT FORM (PARENT OR GUARDIAN)

Do custom fitted shoes change physical activity in children with Down syndrome?

I hereby wish to WITHDRAW my consent for my child or adolescent to participate in the research proposal described above and understand that such withdrawal WILL NOT jeopardise my relationship with La Trobe University.

I understand that all traces of my child or adolescent's data will be removed from the project, provided I tell the principal researcher (Shannon Munteanu) this within four weeks of having finished taking part in the project.

Child or adolescent's name (block letters):_____

Name of parent (block letters):_____

Signature:_____ Date: _____

Name of researcher (block letters):_____

Signature:_____ Date: _____

College of Science Health and Engineering
School of Allied Health

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

REVOCATION OF CONSENT FORM (ADOLESCENT)

Do custom fitted shoes change physical activity in children with Down syndrome?

I do not want to be a part of the project anymore. I would like you to destroy any information I have given you and remove my name and details from the project. I understand that I need to tell the principal researcher (Shannon Munteanu) that I would like all traces of my data to be removed from the project within four weeks of having finished taking part in the project.

I understand that nothing bad will happen to me or my family because I have changed my mind.

Participant's name (block letters):_____

Signature:_____ Date:_____

Name of researcher (block letters):_____

Signature:_____ Date: _____

School of Allied Health, College of Science, Health and Engineering,
La Trobe University, Victoria 3086 Australia
T (03) 9479 5866, F (03) 9479 5768

INITIAL APPOINTMENT

Date: …….

Study Identification: SHOEFIT_

Do customised fit shoes increase physical activity in children with Down syndrome?

La Trobe University Human Ethics Committee application reference number HEC16-027

Investigators:

Professor Nora Shields	**Dr Shannon Munteanu**	**Dr Angela Evans**
Dr Cylie Williams	**Professor Hylton Menz**	**Professor Nicholas Taylor**
Ms Nirmeen Hassan	**Dr Andrew Buldt**	

LA TROBE UNIVERSITY

PARTICIPANT AND/OR GUARDIAN CONSENT

Informed consent form signed by:

Participant ☐ Guardian ☐

LA TROBE UNIVERSITY

PARTICIPANT CHARACTERISTICS AND MEDICAL HISTORY

Sex: Male / Female

Initials: First name / Surname:_____ (e.g SM)

Type of Down syndrome Trisomy 21 ☐ Translocation ☐ Mosaic ☐

DOB: _____ / _____/ _____ Age (years): (5-17 yrs)

Unable to be enrolled if <5 years or 18 years or older (or unable to follow basic verbal instructions)

Does your child currently use foot orthoses/arch supports: Yes / No (please circle)

Does your child use any other aids: Yes / No (please circle)

Please specify:_____

Unable to be enrolled if unable to walk without the aid of a walker or cane?

MEDCON Does your child have/ever had any of the following **medical conditions**?

Hearing impairment HEAR	Yes	[ ]	No	[ ]
Vision impairment VISION	Yes	[ ]	No	[ ]
Diabetes DIAB	Yes	[ ]	No	[ ]
Stroke STROKE	Yes	[ ]	No	[ ]
Cancer CANCER	Yes	[ ]	No	[ ]
Heart disease/ HEARTDISEASE	Yes	[ ]	No	[ ]
Incontinence – urinary INCONT	Yes	[ ]	No	[ ]
Broken bones FRCT	Yes	[ ]	No	[ ]
Hypothyroidism HYPOTHY	Yes	[ ]	No	[ ]
Leukemia LEUK	Yes	[ ]	No	[ ]
Atlantoaxial dysfunction ATLANTO	Yes	[ ]	No	[ ]
Epilepsy EPILEP	Yes	[ ]	No	[ ]
Constipation CONSTIP	Yes	[ ]	No	[ ]
Celiac disease CELIAC	Yes	[ ]	No	[ ]
Anxiety ANXIETY	Yes	[ ]	No	[ ]
Depression DEPRES	Yes	[ ]	No	[ ]
Autism AUTISM	Yes	[ ]	No	[ ]
Other medical condition (please specify) MEDOTHER	Yes	[ ]	No	[ ]

Unable to be enrolled if the presence of a concomitant medical condition or injury that could affect their physical function (e.g neurological or inflammatory disorder)

MEDS What medications is your child currently taking? Include medications bought directly from the chemist or shop without prescription. (Tablets, capsules, mixtures, powders, injections, eye drops, vitamins, herbs etc).

0 None [ ] → *Go to next question*

1.	4.
2.	5.
3.	6.

LA TROBE UNIVERSITY

PARTICIPANT ANTHROPOMETRICS

Test	Equipment	Instructions	Test result
Height	Stadiometer	• Take 2 height measurements using stadiometer • Remove shoes	**Test 1 (cm 1 dec point):** _____ **Test 2 (cm 1 dec point):** _____
Weight	Standard weighing scales	• Take 2 weight measurements using scales • Remove shoes	**Test 1 (kg 1 dec point):** _____ **Test 2 (kg 1 dec point):** _____
Waist circumference	Tape measure	• Take 2 measurements using tape measure	**Test 1 (cm 1 dec point):** _____ **Test 2 (cm 1 dec point):** _____
Hip circumference	Tape measure	• Take 2 measurements using tape measure	**Test 1 (cm 1 dec point):** _____ **Test 2 (cm 1 dec point):** _____

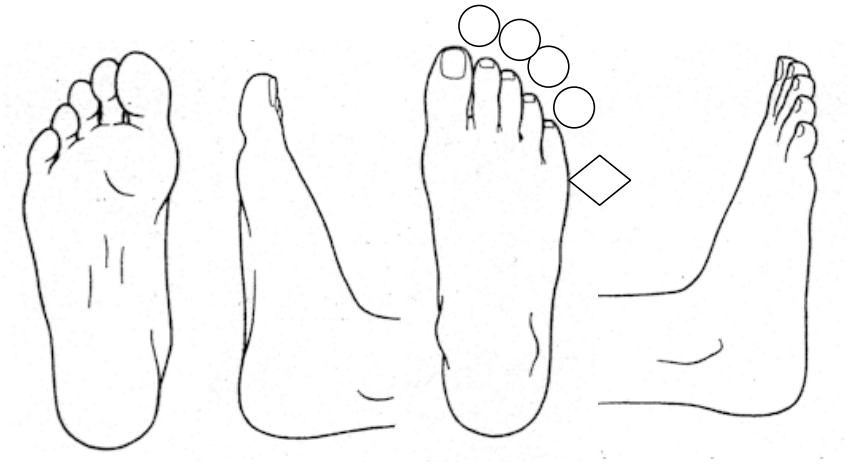
LA TROBE UNIVERSITY

FOOT ASSESSMENT

○	Lesser toe deformity (tick if present)	Blistering (sketch and label)
◇	Bunionette (tick if present)	Onychomycosis (sketch and label)
	Corns/callus (sketch areas and label)	Onychocryptosis (sketch and label)
	Warts (sketch and label)	Other…
	TInea (sketch and label)	

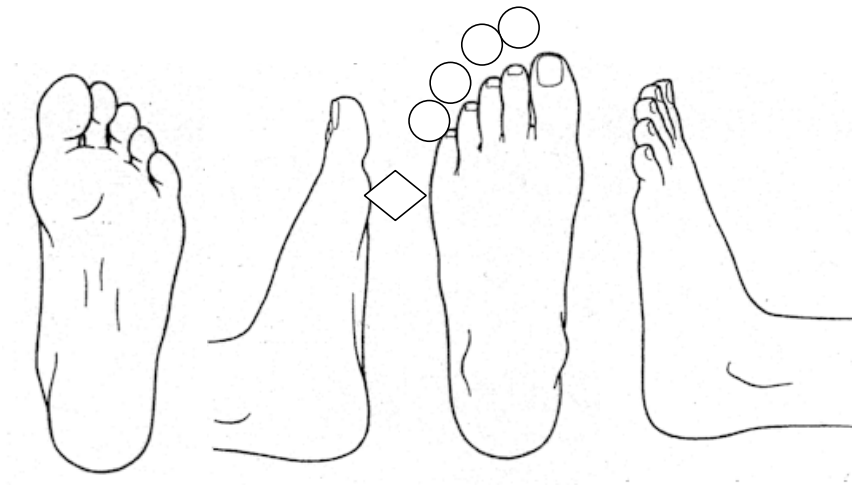
Right foot:

Xerosis scale: 0 1 2 3 4 5 6



Left foot

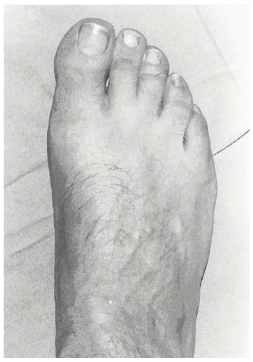
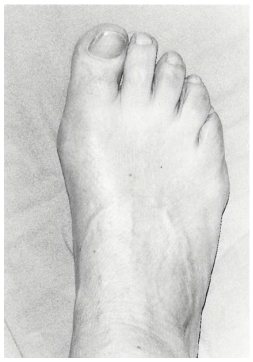
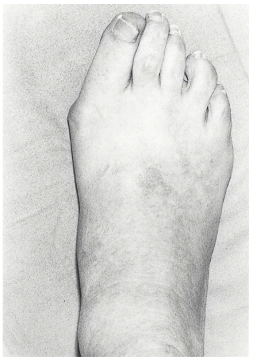
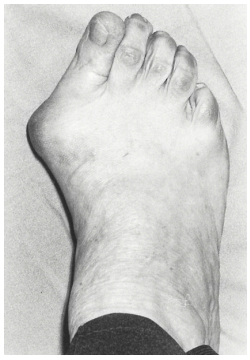
Xerosis scale: 0 1 2 3 4 5 6



5

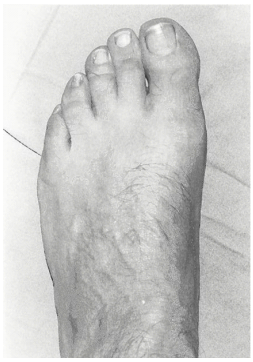
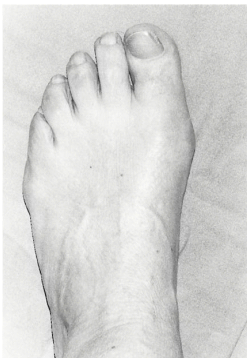
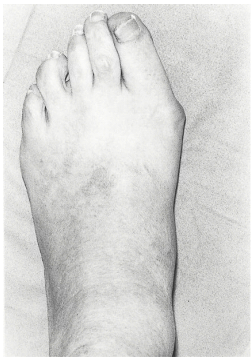
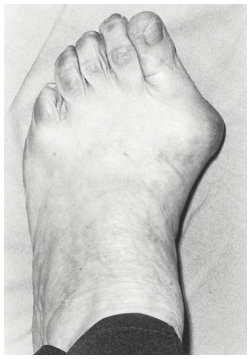
HVR **Hallux valgus** – Manchester scale – right foot

_____ grade

0  1  2  3 

HVL **Hallux valgus** – Manchester scale – left foot

_____ grade

0  1  2  3 

LA TROBE UNIVERSITY

Foot Posture Assessment

FPIL / FPIR Foot Posture Index (FPI)

	Left foot	Right foot
1 Talar head palpation	X	
2 Supra / infra malleolar curvature	X	
3 Inversion/eversion of calcaneus	X	
4 Prominence of TNJ	X	
5 Congruence of medial arch	X	
6 Abduction/adduction of FF on RF	X	
TOTAL	X	

AI R **Arch Index**

Left foot:	Right foot:
X	

7

ASSESSMENT OF DIMENSIONS OF FOOTWEAR

FIT_USUAL_nonschool

Usual non-school shoes

Left shoe:	Right shoe:
Length (mm):	Length (mm):
Maximum width (mm)	Maximum width (mm)
Total area (mm^2)	Total area (mm^2)

FIT_USUAL_school

Usual school shoes

Left shoe:	Right shoe:
Length (mm):	Length (mm):
Maximum width (mm)	Maximum width (mm)
Total area (mm^2)	Total area (mm^2)

FIT_USUAL_nonschool

Clarks non-school shoes

Left shoe:	Right shoe:
Length (mm):	Length (mm):
Maximum width (mm)	Maximum width (mm)
Total area (mm^2)	Total area (mm^2)

FIT_USUAL_nonschool

Clarks school shoes

Left shoe:	Right shoe:
Length (mm):	Length (mm):
Maximum width (mm)	Maximum width (mm)
Total area (mm^2)	Total area (mm^2)

LA TROBE UNIVERSITY

Trace outline here OF USUAL RIGHT NON-SCHOOL shoe

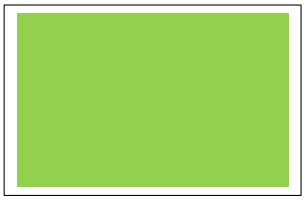
LA TROBE UNIVERSITY

Trace outline here OF USUAL RIGHT SCHOOL shoe

ASSESSMENT OF DIMENSIONS OF FEET

Ask subject to stand on carbon imprint paper and trace around RIGHT FOOT

Tick box when completed:



FIT_FEET **Foot**

Left foot:	**Right foot:**
Length (mm):	Length (mm):
Maximum width (mm)	Maximum width (mm)
Total area (mm^2)	Total area (mm^2)

ASSESSMENT OF SHOE SIZING USING FOOT GAUGE / SHOE PREFERENCE

SHOESIZE**Sizing**

Left foot:	**Right foot:**
Length:	Length
Width:	Width:

Recommended shoe length and width to order (e.g. 1 ½ H) :

_____

Shoe preference

	Male	Female	Unisex (if feet too large or prefer lace-ups)
School	Lochie	Laura	Daytona
Casual	Ventura (black, white/navy, white fuchsia, white/silver)		Vancouver (black, white/navy, white/silver)

Rules:
Fit with socks on
Sitting position

Length:
If more than a half number is covered, then **go up**;
If a ½ size but the shoe is only in full size length, then go up size
Width: If b/w two width fittings, then **go up** size.

LA TROBE UNIVERSITY

3D FOOT SCANS

Label as study SHOEFITRCT(study number)_right/left
e.g.SHOEFITRCT01_R
Use "VERY DARK" setting and turn off lights in the room. Participant should be standing with feet approx. shoulder width apart.

☐ Right foot ☐ Left foot

LA TROBE UNIVERSITY

FOOTWEAR ASSESSMENT TOOL

FAT_USUALNONSCHOOL **Usual NON-SCHOOL/CASUAL shoes**

1. Shoe details, including size _____

2. Age of shoes: _____ (months)

3. Footwear style:
1[ ] walking shoe 2[ ] athletic shoe 3[ ] oxford shoe 4[ ] moccasin
5[ ] boot 6[ ] ugg boot 7[ ] high heel 8[ ] thongs
9[ ] slipper 10[ ] backless slipper 11[ ] court-shoe 12[ ] mule
13[ ] sandal 14[ ] surgical/bespoke 15[ ] other:_____

4. Heel height:_____ (mm)

5. Forefoot height:_____ (mm)

6. Flexion point: 1[ ] at MTPJs 2[ ] proximal 3[ ] distal

7. Midfoot sole sagittal stability: 1[ ] minimal (>45º) 2[ ] moderate (≤ 45º) 3[ ] rigid (0-10º)

8. Fixation: 1[ ] none 2[ ] laces 3[ ] straps/buckles 4[ ] Velcro 5[ ] zips

FOOTWEAR ASSESSMENT TOOL

FAT_USUALSCHOOL **Usual SCHOOL shoes**

1. Shoe details, including size _____

2. Age of shoes: _____ (months)

3. Footwear style:
1[ ] walking shoe 2[ ] athletic shoe 3[ ] oxford shoe 4[ ] moccasin
5[ ] boot 6[ ] ugg boot 7[ ] high heel 8[ ] thongs
9[ ] slipper 10[ ] backless slipper 11[ ] court-shoe 12[ ] mule
13[ ] sandal 14[ ] surgical/bespoke 15[ ] other:_____

4. Heel height:_____ (mm)

5. Forefoot height:_____ (mm)

6. Flexion point: 1[ ] at MTPJs 2[ ] proximal 3[ ] distal

7. Midfoot sole sagittal stability: 1[ ] minimal (>45º) 2[ ] moderate (≤ 45º) 3[ ] rigid (0-10º)

8. Fixation: 1[ ] none 2[ ] laces 3[ ] straps/buckles 4[ ] Velcro 5[ ] zips

LA TROBE UNIVERSITY

ACTIGRAPH PHYSICAL ACTIVITY MONITOR TO BE PROVIDED TO PARENT

LA TROBE UNIVERSITY

CHECKLIST

Place tick next to each dot when complete

	Pre-Baseline (t-2 weeks)	Baseline (t0)	Week 6	Week 12
	Clinic	GAIT LAB	Postal	Clinic
Informed consent obtained and eligibility confirmed	●			
Initial / baseline assessments				
Measure for shoes	●			
Participant characteristics / medical history	●			
Foot assessment (deformity, dermatology, Foot Posture Index/Arch Index, toe deformity)	●			
Simplified Footwear Assessment Tool – non-school and school shoes	●	●		
Shoe Fit casual and school shoes (Menz Morris)	●	●		
3D foot scan (shoe fit) of right foot	●			
Biomechanical assessment		●		●
Primary outcome measure				
Physical activity (Actigraph monitor 7 days)	●		●	●
Secondary outcome measures				
Oxford Ankle Foot Questionnaire for Children for children - parent proxy		●	●	●
Acceptability of the custom fit shoes (experimental group interview)				●
Adherence (experimental group)			●	●
Adverse events (experimental group)			●	●
Use of cointerventions (including shoe changes)			●	●

OXFORD ANKLE AND FOOT QUESTIONNAIRE FOR CHILDREN – PARENT VERSION

The following questions are based on how children and adolescents have previously expressed how they have been affected by a problem to their feet or ankles.

Please fill out this questionnaire to the best of your ability and place a tick under each answer that describes your child or adolescent best for each question.

Considering the previous week,

1. Has your child found walking difficult because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

2. Has your child found it difficult to run because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

3. Has it been difficult for your child to stand up for long periods?

never	rarely	sometimes	very often	always
○	○	○	○	○

4. Has your child had pain in their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

5. Have your child's legs been sore or ached after walking or running?

never	rarely	sometimes	very often	always
○	○	○	○	○

6. Has your child felt tired because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

7. Has your child's foot or ankle stopped them joining in with others in the playground?

never	rarely	sometimes	very often	always
○	○	○	○	○

8. Has your child's foot or ankle stopped them playing in the park or outside?

never rarely sometimes very often always

○ ○ ○ ○ ○

9. Has your child's foot or ankle stopped them taking part in physical education lessons?

never rarely sometimes very often always

○ ○ ○ ○ ○

10. Has your child's foot or ankle stopped them taking part in any other lessons at school?

never rarely sometimes very often always

○ ○ ○ ○ ○

11. Has your child been bothered by how their foot or ankle looks?

never rarely sometimes very often always

○ ○ ○ ○ ○

12. Has the way your child walks bothered them?

never rarely sometimes very often always

○ ○ ○ ○ ○

13. Has your child been embarrassed because of their foot or ankle?

never rarely sometimes very often always

○ ○ ○ ○ ○

14. Has anyone been unkind to your child because of their foot or ankle?

never rarely sometimes very often always

○ ○ ○ ○ ○

15. Has your child's foot or ankle stopped them wearing any shoes they wanted to wear?

never rarely sometimes very often always

○ ○ ○ ○ ○

POST INTERVENTION INTERVIEW SCHEDULE (Participants with Down syndrome and their parents)

INTERVIEW PURPOSE & FRAMEWORK:

- To find out how the participants felt about their custom fitted shoes, and what effects they had, if any
- Use the following questions to guide your discussion
- Feel free to follow a participant's lead, and explore their thoughts as they arise

INITIAL STATEMENTS TO PARTICIPANTS:

- Let the participant know that you will be asking them questions about their custom fitted shoes, which they have been wearing for 12 weeks
- Remind the participant that you will be recording their answers, so that the researchers can look at them later
- Reassure the participant that they can give whatever answers they like, and that they won't get into trouble for saying what they think

Topic area:	Question examples:
Context questions & general thoughts	Tell me about the shoes you got What did you like about the shoes? What were the things you didn't like about the shoes? • Prompts: style, colour, comfort, fit Have you enjoyed wearing your shoes? Why/ why not? How often have you worn the shoes? • Prompts: daily, just for school, just at home, when asked
Shoes in the future	What would you change about the shoes? Would you like/ your child/ your adolescent like to continue wearing custom fitted these shoes? Why? Why not?

Appendix 8. Forms associated with the cross-sectional observational study

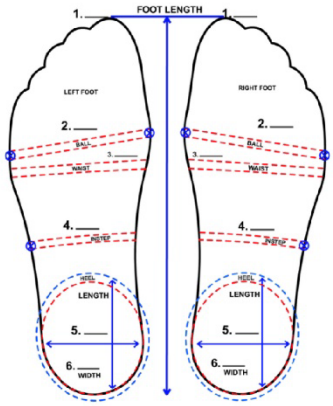
College of Science Health and Engineering
School of Allied Health, Human Services and Sport

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

FREE FOOT HEALTH ASSESSMENT FOR YOUR CHILD!

DIFFERENCES IN FOOT ANTHROPOMETRY AND FOOT HEALTH IN CHILDREN AND ADOLESCENTS WITH AND WITHOUT DOWN SYNDROME



Background

We are conducting a research project that aims to determine the foot dimensions and foot health of children and adolescents without Down syndrome and compare these findings to those with Down syndrome.

What is involved?

Your child or adolescent will attend a 30 minute appointment at the Health Sciences Clinic at La Trobe University to have their foot health and footwear assessed. Additionally, a 3D image will be taken of their foot. You will need to bring in the 2 pairs of footwear – one pair of most commonly worn casual shoes and one pair of school shoes.

Who is eligible?

Young people **without** Down syndrome aged between 5 – 19 years.

Investigators of the project

Nirmeen Hassan, Associate Professor Shannon Munteanu, Associate Professor Karl Landorf, Dr Andrew Buldt are research podiatrists at La Trobe University. Professor Nora Shields is a physiotherapist researcher at La Trobe University.

Interested?

If you're interested in having your child participate in this project or have any questions regarding this project, please contact Nirmeen Hassan (03 9479 6760 or n.hassan@latrobe.edu.au).

The research is being carried out in partial fulfilment of a PhD under the supervision of A/Prof Shannon Munteanu, A/Prof Karl Landorf and Professor Nora Shields. The following researchers will be conducting the study:		
Role	**Name**	**Organisation**
Chief Investigator	A/Prof Shannon Munteanu	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Associate investigator	Nirmeen Hassan	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Associate investigator	A/Prof Karl Landorf	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Associate investigator	Dr. Andrew Buldt	La Trobe Sport and Exercise Medicine Research Centre
Associate investigator	Prof. Nora Shields	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Research funder	This research receives in-kind support from La Trobe University.	

1. **What is the study about?**
   This is an invitation for your child to take part in a study. Children with Down syndrome have a unique foot shape that can complicate footwear fitting. As many children with Down syndrome have wider feet than other children, children with Down syndrome often wear poorly-fitted footwear, namely footwear that is too long to accommodate for the lack of width. This is problematic as poorly-fitted footwear can cause pain and interfere with normal walking patterns. This research project will recruit typically developing children to assess their foot dimensions and foot health. Children recruited to this project will then be matched to children with Down syndrome based on age and sex. Therefore, the aim of this project is to compare the differences in foot dimensions and foot health in children with and without Down syndrome.

2. **Does my child have to participate?**
   Being part of this study is voluntary. We ask that you discuss the study with your child when you are deciding if you want your child to take part. If you decide together for your child to be part of the study, we ask that you read this information carefully and ask us any questions.

   If you decide together you do not want your child to take part this won't affect your relationship with La Trobe University or any other listed organisation. You can read the information below and decide at the end if you do not want your child to take part.

3. **Who is being asked to participate?**
   Your child has been asked to be part of the study because:
   - They are aged between 5 to 17 years
   - They do not have Down syndrome

   Your child cannot participate in the study if they have:
   - Foot pain
   - A medical condition that may affect foot function
   - Lower limb amputation
   - Surgery to the lower limb in the last 12 months
   - Require the use of an ambulatory assistive device (such as a cane or walker)
   - Do not have the appropriate characteristics (age and sex) required to match the case group (children with Down syndrome who have been recruited previously).

4. What will my child be asked to do?

If your child wants to take part in this study, we will gather data about your child, including their date of birth, medical history and existing medication list (if applicable). We will measure your child's height and weight and calculate their body mass index. Your child will have their foot assessed through a number of ways that are described below.

Foot health assessment:

- Foot type: clinical scoring system and arch profile to determine foot posture
- Skin condition: visual assessment of the skin and nail to observe for corns, calluses, infections, ingrown toe nails
- Presence of bony deformities: visual assessment for the presence of bunions and toe deformities
- Foot shape: a 3D foot scan will be taken of your child's foot. Your child's face will not be visible in these scans. These scans create a 3D model of the foot which provides data on dimensions (i.e. length and width of the foot)
- Footwear characteristics: your child's shoes will be assessed for a number of characteristics such as style of footwear, age, shoe fit. **You will be required to bring in two pairs of footwear for the assessment– your child's school shoe and your child's casual shoes worn frequently**.

Foot and ankle disability assessment:

- You will be given a questionnaire to complete that collects information about how your child's feet or ankles may affect them.

It will take 30 minutes of your child's time to be part of this study. We will require you to be present at the same time.

When your child is taking part in this study, the following people will be present:

Name/Organisation	Position
Nirmeen Hassan, La Trobe University	PhD student and investigator of the project

5. What are the benefits?

The benefit of your child taking part in this study is that you will receive a free foot health assessment. In the event that the foot health assessment identifies an issue that may require treatment, a complimentary appointment at the La Trobe University Podiatry Clinic will be provided. The La Trobe University Podiatry Clinic is a student-run clinic that services the local community for all their podiatric needs. Students are exposed to a variety of patients and practice under the supervision of registered Podiatrists. Please note, we will waive the consultation fee of the complimentary appointment, however the cost of certain treatment (for example, purchasing consumables or requiring insoles) will need to be charged and will be explained by the student Podiatrist before any costs are incurred.

The expected benefits to society in general is that it will improve our understanding of variances in foot dimensions between children with and without Down syndrome which is useful information for footwear manufacturers. It is possible that this data can be used to guide footwear manufacturing for children with Down syndrome to create better fitting footwear.

6. What are the risks?

With any study there are (1) risks we know about, (2) risks we don't know about, and (3) risks we don't expect. If you or your child experience something that you aren't sure about, please contact us immediately so we can discuss the best way to manage your concerns.

Name/Organisation	Position	Telephone	Email
A/Prof Shannon Munteanu	Chief investigator	9479 5866	s.munteanu@latrobe.edu.au
Nirmeen Hassan	Associate investigator	9479 6760	nmhassan@students.latrobe.edu.au
A/Prof Karl Landorf	Associate investigator	9479 5300	k.landorf@latrobe.edu.au
Dr. Andrew Buldt	Associate investigator	9479 6760	a.buldt@latrobe.edu.au
Prof. Nora Shields	Associate investigator	94795852	n.shields@latrobe.edu.au

We do not foresee any risks associated with this study.

7. **What will happen to information about my child?**

We will **collect** information about your child in ways that do not reveal who they are.

We will **store** information about your child in ways that do not reveal who they are.

We will **publish** information about your child in ways that cannot be identified in any type of publication from this study.

We will **keep** your child's information for 7 after the project is completed. After this time, we will destroy all of your child's data.

The storage, transfer and destruction of your child's data will be undertaken in accordance with the Research Data Management Policy https://policies.latrobe.edu.au/document/view.php?id=106/.

The personal information provided will be handled in accordance with applicable privacy laws, any health information collected will be handled in accordance with the Health Records Act 2001 (Vic). Subject to any exceptions in relevant laws, you have the right to access and correct your child's personal information by contacting the research team.

8. **Will we hear about the results of the study?**

We will let you know about the results of the foot health assessment within two weeks of the data collection session. You will also receive the results of the study via email once complete. As the tests provide information about your child's foot health, you will be advised if there are any issues that require the attention of a Podiatrist. In this case, a complimentary consultation with student Podiatrists at the La Trobe University Podiatry Clinic will be organised.

9. **What if we change our minds?**

You or your child can choose to no longer be part of the study at any time, including during the appointment, until [four weeks] following the collection of your data. You can let us know by:

1. Completing the 'Withdrawal of Consent Form' (provided at the end of this document);
2. Phoning us; or
3. Emailing us

Your or your child's decision to withdraw at any point will **not** affect your relationship with La Trobe University or any other organisation listed.

When you withdraw your child from the study we will stop asking for information. Any identifiable information about your child will be withdrawn from the research study. However, once the results have been analysed we can only withdraw information, such as your child's name and contact details. If results haven't been analysed you can choose if we use those results or not.

10. **Who can we contact for questions or want more information?**

If you or your child would like to speak to us, please use the contact details below:

Name/Organisation	Position	Telephone	Email
A/Prof Shannon Munteanu	Chief investigator	9479 5866	s.munteanu@latrobe.edu.au
Nirmeen Hassan	Associate investigator	9479 6760	nmhassan@students.latrobe.edu.au

A/Prof Karl Landorf	Associate investigator	9479 5300	k.landorf@latrobe.edu.au
Dr. Andrew Buldt	Associate investigator	9479 6760	a.buldt@latrobe.edu.au
Prof. Nora Shields	Associate investigator	94795852	n.shields@latrobe.edu.au

11. What if we have a complaint?

If you or your child would like to make a complaint about any part of this study, please contact:

Ethics Reference Number	Position	Telephone	Email
HEC19-290	Senior Research Ethics Officer	+61 3 9479 1443	humanethics@latrobe.edu.au

Consent Form – Declaration by Parent/Guardian

I (the parent/guardian) have read (or, where appropriate, have had read to me) and understood the parent/guardian information statement, and any questions have been answered to my satisfaction. I understand I am being asked to provide consent for my child to be part of this study. I agree for my child to participate in the study, I know either myself or my child can withdraw at any time until [four weeks] following the collection of data. I agree information provided by my child or with my permission during the project may be included in a thesis, presentation and published in journals on the condition that my child cannot be identified.

I would like my child's information collected for this research study to be:

☐ Only used for this specific study (up until my child turns 18, and then they will be asked for their own consent);
☐ Used for future related studies (up until my child turns 18, and then they will be asked for their own consent);
☐ Used for any future studies (up until my child turns 18, and then they will be asked for their own consent)
☐ I would like to receive a copy of the results via email or post. I have provided my details below and ask that they only be used for this purpose and not stored with my information or for future contact.

Name	Email (optional)	Postal address (optional)

Parent/Guardian Signature

☐ I have received a signed copy of the Parent/Guardian Information Statement to keep
☐ If appropriate - I have discussed the study with my child and through these discussions, they have shown to me they want to be part of the study.

Parent/Guardian printed name	
Parent/Guardian signature	
Date	

Declaration by Researcher

☐ I have given a verbal explanation of the study, what it involves, and the risks and I believe the participant has understood;
☐ I am a person qualified to explain the study, the risks and answer questions

Researcher's printed name	
Researcher's signature	
Date	

* All parties must sign and date their own signature

Withdrawal of Consent

I wish to withdraw my consent for my child to participate in this study. I understand withdrawal will not affect my or my child's relationship with La Trobe University of any other organisation or professionals listed in the Participant Information Statem ent. I understand my child's information will be withdrawn as outlined below:

- ✓ My child will not be asked to provide any more information
- ✓ Any identifiable information will be withdrawn from the study
- ✓ The researchers cannot withdraw my child's information once it has been analysed

I would like my child's already collected and unanalysed data
☐ Destroyed and not used for any analysis
☐ Used for analysis

Parent/Guardian Signature

Parent/Guardian's printed name	
Parent/Guardian's signature	
Date	

Please forward this form to:

CI Name	Shannon Munteanu
Email	s.munteanu@latrobe.edu.au
Phone	9479 5866
Postal Address	Shannon Munteanu, Room 539, Level 5, Health Sciences 3, La Trobe University, 3086

College of Science Health and Engineering
School of Allied Health, Human Services and Sport

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

ASSENT FORM (CHILD/ADOLESCENT)

DIFFERENCES IN FOOT ANTHROPOMETRY AND FOOT HEALTH IN CHILDREN AND ADOLESCENTS WITH AND WITHOUT DOWN SYNDROME

I _____ have read and understood the information about the research project, and any questions I have asked has been answered to help me understand the information about the project.

I agree to join the project, and I know that this information may be used for future, related projects. I know that I am able to stop taking part in this project at any time I like, including now during the appointment and after the appointment. I also know that I am allowed to ask for my name and all the information I have given to be removed from the project records up to four weeks after I joined the project.

I know and agree to have my information shared in papers and articles that may be presented at conferences or in journals, however my personal information that may identify me will not be used.

Child or adolescent's name (block letters): _____

Name of parent or guardian (block letters): _____

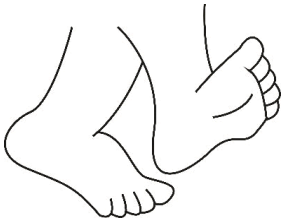
Signature: _____ Date: _____

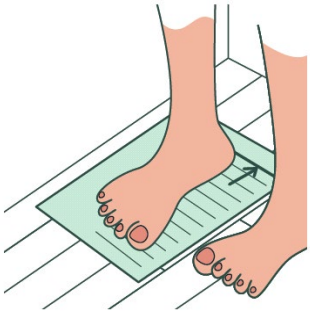
Name of researcher (block letters): _____

Signature: _____ Date: _____

CONSENT FORM FOR CHILDREN

We are doing a project about your feet. For this project, we will:





Measure and scan your feet

Look at how healthy your feet are

And measure your shoes to see how they fit

Lastly, your parent will answer a few questions……and that's it! ☺

Would you like to join? (Please circle)

Participant signature: ……

Parent signature: ……

Date:……

The research is being carried out in partial fulfilment of a PhD under the supervision of A/Prof Shannon Munteanu, A/Prof Karl Landorf and Professor Nora Shields. The following researchers will be conducting the study:		
Role	**Name**	**Organisation**
Chief Investigator	A/Prof Shannon Munteanu	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Associate investigator	Nirmeen Hassan	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Associate investigator	A/Prof Karl Landorf	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Associate investigator	Dr. Andrew Buldt	La Trobe Sport and Exercise Medicine Research Centre
Associate investigator	Prof. Nora Shields	Department of Physiotherapy, Podiatry, and Prosthetics and Orthotics
Research funder	This research receives in-kind support from La Trobe University.	

1. **What is the study about?**
We invite you to join our study. Kids with Down syndrome have a unique foot shape that can make it hard for them to find shoes that fit well. As many kids with Down syndrome have wider feet than other kids, they often wear shoes that don't fit well for their feet. They have to wear shoes that are too long so that they can be wide enough. This is a problem for these kids because shoes that don't fit well can cause pain and change the way they walk.
This project will ask kids **without** Down syndrome to join so that we may collect information about their foot dimensions and foot health. This information will be compared to information about kids with Down syndrome so that we can see the differences between the different feet.

2. **Do I have to participate?**
Joining our study is your choice. If you would like to join, please read the rest this information and ask us any questions.

You can read the information below and decide at the end if you do not want to join. If you decide not to join our study, this won't affect your relationship with La Trobe University, or any other group listed.

3. **Who is being asked to participate?**
You have been asked to participate because you:
- Aged between 5 to 17 years
- Do not have Down syndrome

You cannot join if you have:
- Foot pain
- A medical condition that may affect the way your foot works
- Surgery to your legs in the last 12 months (including amputation)
- Need to use a device that will help with walking (such as a cane or walker)
- Are not the same age or gender as the kids with Down syndrome

4. **What will I be asked to do?**

If you would like to join this study, we will ask your parents questions about your birthday, your medical history and existing medication list (if you have one). We will measure your height and weight and calculate your body mass index. Your foot will be checked through a number of ways that are described below.

Foot health assessment:
- Foot type: clinical scoring system and arch profile to determine foot posture

- Skin condition: visual assessment of the skin and nail to check for corns, calluses, infections, ingrown toe nails
- Presence of bony deformities: visual assessment for the presence of bunions and toe deformities
- Foot shape: a 3D foot scan will be taken of your foot. Your face will not be visible in these scans. These scans create a 3D shape of the foot which provides information on dimensions (i.e. length and width of the foot)
- Footwear characteristics: your shoes will be checked for the style of shoe, age, shoe fit. **You will be required to bring in two pairs of footwear for the appointment– your school shoe and your casual shoes worn frequently**.

Foot and ankle disability assessment:

- Your parents will answer some questions about how your child's feet or ankles may affect them.

It will take 30 minutes of your time to be part of this study.

5. What are the benefits?

The benefit of taking part in this study is that you will receive a free foot health assessment, which will tell you how healthy your feet are. If we find any issues with your feet, we will provide you a free appointment at the La Trobe University Podiatry Clinic. The La Trobe University Podiatry Clinic is a student-run clinic where students are exposed to a many patients and work under the help of Podiatrists (a foot expert). Please note, the cost of the consultation will be free but if you need certain treatments, your parents will need to pay for it. We will explain this to your parents beforehand.

The expected benefits to society in general is that it will improve our understanding of differences in foot measurements between kids with and without Down syndrome which is useful information for footwear companies. It is possible that this information can be used to guide footwear companies to make better-fitting shoes for kids with Down syndrome.

6. What are the risks?

With any study there are (1) risks we know about, (2) risks we don't know about and (3) risks we don't expect. If something happens that you aren't sure about, please let your parents know so that they can contact us immediately to manage any concern.

Name/Organisation	Position	Telephone	Email
A/Prof Shannon Munteanu	Chief investigator	9479 5866	s.munteanu@latrobe.edu.au
Nirmeen Hassan	Associate investigator	9479 6760	nmhassan@students.latrobe.edu.au
A/Prof Karl Landorf	Associate investigator	9479 5300	k.landorf@latrobe.edu.au
Dr. Andrew Buldt	Associate investigator	9479 6760	a.buldt@latrobe.edu.au
Prof. Nora Shields	Associate investigator	94795852	n.shields@latrobe.edu.au

We do not see any risks with this study.

7. What will happen to information about me?

We will **collect** information about you in ways that will not tell who you are.

We will **store** information about you in ways that will not tell who you are.

We will **publish** information about you in ways that will not be identified in any type of publication from this study.

We will **keep** your information for 7 after the project is completed. After this time we will destroy all of your data.

The storage, transfer and destruction of your data will be undertaken in accordance with the Research Data Management Policy https://policies.latrobe.edu.au/document/view.php?id=106/.

The personal information you provide will be handled in accordance with applicable privacy laws, any health information collected will be handled in accordance with the Health Records Act 2001 (Vic). Subject to any exceptions in relevant laws, you have the right to access and correct your personal information by contacting the research team.

8. **Will I hear about the results of the study?**
We will let your parents know about the results of the foot health assessment within two weeks of the data collection session. Your parents will also receive the results of the study through their email once complete. As the tests tell us information about your foot health, your parents will be told if there are any problems that needs you to see a Podiatrist. In this case, a free appointment with student Podiatrists at the La Trobe University Podiatry Clinic will be organised.

9. **What if I change my mind?**
You can choose to stop taking part in the study at any time until [four weeks] after we have collected your information. You can let us know by:
1. Completing the 'Withdrawal of Consent Form' (provided at the end of this document);
2. Calling us; or
3. Emailing us

Your decision to stop taking part or having your information removed at any point will **not** affect your relationship with La Trobe University or any other group listed.

When you withdraw we will stop asking you for information. Any identifiable information about you will be withdrawn from the research study. However, once the results have been analysed we can only withdraw information, such as your name and contact details. If results haven't been analysed you can choose if we use those results or not.

10. **Who can I contact for questions or want more information?**
If you would like to speak to us, please use the contact details below:

Name/Organisation	Position	Telephone	Email
A/Prof Shannon Munteanu	Chief investigator	9479 5866	s.munteanu@latrobe.edu.au
Nirmeen Hassan	Associate investigator	9479 6760	nmhassan@students.latrobe.edu.au
A/Prof Karl Landorf	Associate investigator	9479 5300	k.landorf@latrobe.edu.au
Dr. Andrew Buldt	Associate investigator	9479 6760	a.buldt@latrobe.edu.au
Prof. Nora Shields	Associate investigator	94795852	n.shields@latrobe.edu.au

11. **What if I have a complaint?**
If you have a complaint about any part of this study, please contact:

Ethics Reference Number	Position	Telephone	Email
HEC19-290	Senior Research Ethics Officer	+61 3 9479 1443	humanethics@latrobe.edu.au

Consent Form – Declaration by Participant

I (the participant) have read (or, where appropriate, have had read to me) and understood the participant information statement, and any questions have been answered to my satisfaction. I agree to participate in the study, I know I can withdraw at any time until [four weeks] following the collection of my data. I agree information provided by me or with my permission during the project may be included in a thesis, presentation and published in journals on the condition that I cannot be identified.

I would like my information collected for this research study to be:

☐ Only used for this specific study;
☐ Used for future related studies;
☐ Used for any future studies
☐ I would like to receive a copy of the results via email or post. I have provided my details below and ask that they only be used for this purpose and not stored with my information or for future contact.

Name	Email (optional)	Postal address (optional)

Participant Signature

☐ I have received a signed copy of the Participant Information Statement and Consent Form to keep

Participant's printed name	
Participant's signature	
Date	

Declaration by Researcher

☐ I have given a verbal explanation of the study, what it involves, and the risks and I believe the participant has understood;
☐ I am a person qualified to explain the study, the risks and answer questions

Researcher's printed name	
Researcher's signature	
Date	

* All parties must sign and date their own signature

LA TROBE UNIVERSITY

College of Science Health and Engineering
School of Allied Health, Human Services and Sport

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

DIFFERENCES IN FOOT ANTHROPOMETRY AND FOOT HEALTH IN CHILDREN AND ADOLESCENTS WITH AND WITHOUT DOWN SYNDROME

Study identification:

Appointment date:

La Trobe University Human Ethics Committee application reference number HEC19-290

Investigators:

Nirmeen Hassan	**Associate professor Shannon Munteanu**	**Professor Nora Shields**
Associate professor Karl Landorf	**Dr Andrew Buldt**	

PARTICIPANT CHARACTERISTICS AND MEDICAL HISTORY

Sex: Male / Female	**Initials:**
DOB:	**Age (years):**
Medical history:	
Medication list: *Include medications bought from Pharmacy/OTC without prescription (i.e. tablets, capsules, mixtures, powders, injections, eye drops, supplements/vitamins, herbs etc).*	

EMAIL (to send foot health report)

Address:

PARTICIPANT ANTHROPOMETRY

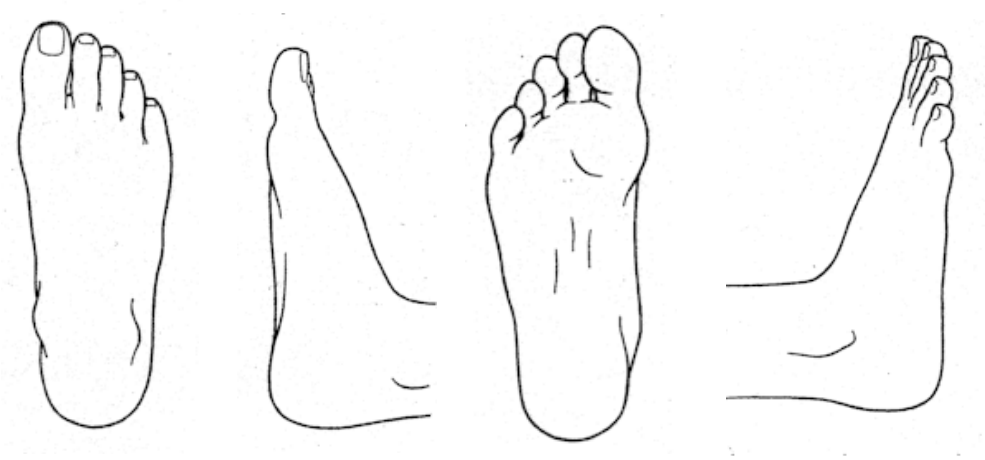
Weight (kg)	• Remove shoes • To one decimal point • Repeat twice • Use weighing scale	**Test 1**	**Test 2**
Height (cm)	• Remove shoes • To one decimal point • Repeat twice • Use flexible tape measure	**Test 1**	**Test 2**
BMI	Height/weight calculation		

FOOT HEALTH ASSESSMENT

Corn/Callus (C)	Bunionette (B)	Lesser toe deformity (L)	Wart (W)	Blister (B)
Onychomycosis (OM)	Onychocryptosis (OC)	ID maceration (IDM)	Tinea (T)	Other - specify

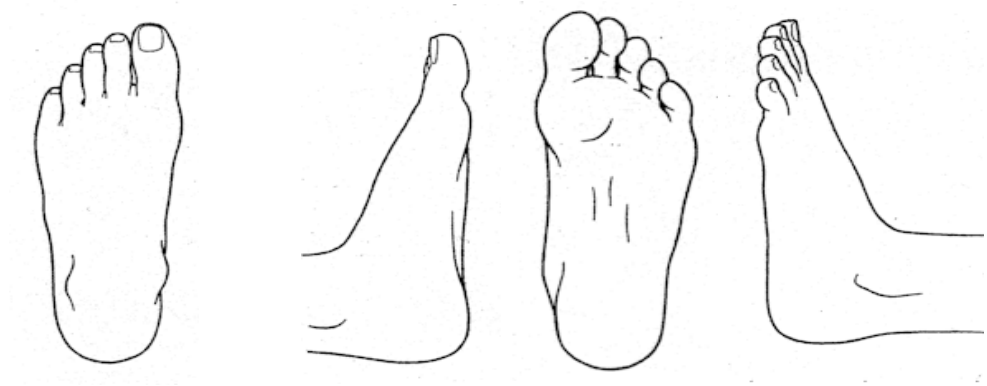
Right foot:

Xerosis scale: 0 1 2 3 4 5 6



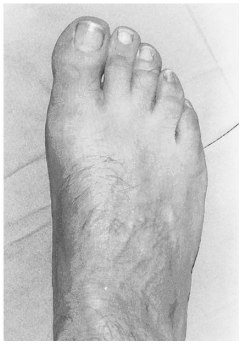
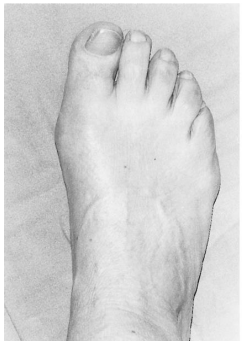
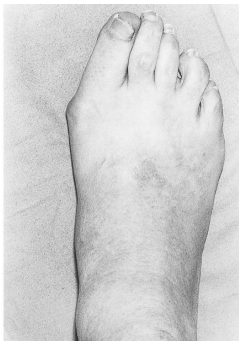
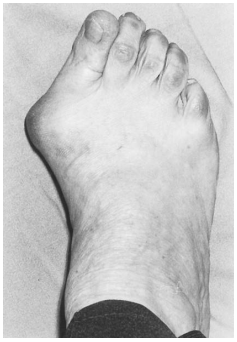
Left:

Xerosis scale: 0 1 2 3 4 5 6



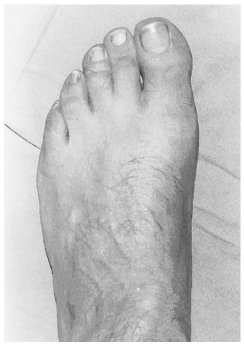
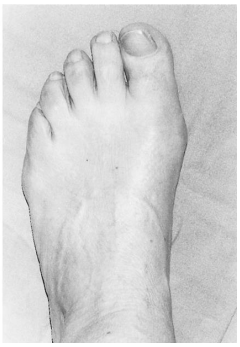
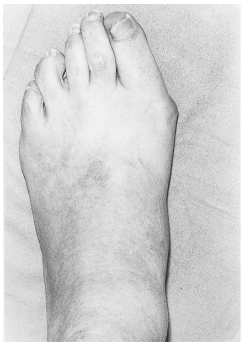
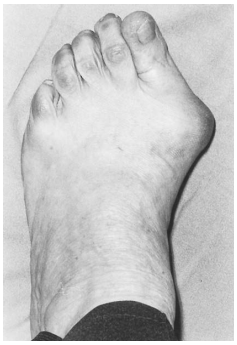
Hallux valgus – Manchester scale – right foot

_____ grade

0  1  2  3 

Hallux valgus – Manchester scale – left foot

_____ grade

0  1  2  3 

5

FOOT POSTURE INDEX

Foot Posture Index (FPI)	Left foot	Right foot
Talar head palpation		
Supra / Infra malleolar curve		
Inversion / eversion of calcaneus		
Prominence of TNJ		
Congruence of medial arch		
Abduction / Adduction of FF on RF		
Total		

ARCH INDEX:

Use carbon imprint paper to obtain **BOTH** foot prints. Trace around foot

	Left foot	Right foot
Arch index		

FOOT DIMENSIONS (MEASURE FROM CARBON PAPER IMPRINT)

	Left foot	Right foot
Length (mm):		
Width (mm):		

ASSESSMENT OF FOOTWEAR DIMENSIONS

	Left shoe:	Right shoe:
Length (mm):		
Width (mm):		

FOOTWEAR TRACING – CASUAL

Trace outline of **RIGHT** casual shoe (use inner lining if removable) here

FOOTWEAR TRACING – SCHOOL

Trace outline of **RIGHT** school shoe (use inner lining if removable) here

3D FOOT SCANS

- Label as follows: FA(studynumberInitials)_R – I.e. FA01AM_R
- Scan both feet
- Use very dark setting and switch off lights in room
- Have participant standing with feet at shoulder width apart
- FA= FOOT ANTRHOPOMETRY

☐ RIGHT FOOT

☐ LEFT FOOT

Footwear assessment tool – Usual CASUAL shoes RIGHT

1. Shoe details, including size _____

2. Age of shoes: _____ (months)

3. Footwear style:

1 [ ] walking shoe	2 [ ] athletic shoe	3 [ ] oxford shoe	4 [ ] moccasin
5 [ ] boot	6 [ ] ugg boot	7 [ ] high heel	8 [ ] thongs
9 [ ] slipper	10 [ ] backless slipper	11 [ ] court shoe	12 [ ] mule
13 [ ] sandal	14 [ ] surgical/bespoke	15 [ ] other:	

4. Heel height:_____ (mm)

5. Forefoot height:_____ (mm)

6. Flexion point: 1[ ] at MTPJs 2[ ] proximal 3[ ] distal

7. Midfoot sole sagittal stability: 1[ ] minimal (>45º) 2[ ] moderate (≤ 45º) 3[ ] rigid (0-10º)

8. Fixation: 1[ ] none 2[ ] laces 3[ ] straps/buckles 4[ ] Velcro 5[ ] zips

Footwear assessment tool – Usual SCHOOL shoes RIGHT

1. Shoe details, including size _____

2. Age of shoes: _____ (months)

3. Footwear style:

1 [ ] walking shoe	2 [ ] athletic shoe	3 [ ] oxford shoe	4 [ ] moccasin
5 [ ] boot	6 [ ] ugg boot	7 [ ] high heel	8 [ ] thongs
9 [ ] slipper	10 [ ] backless slipper	11 [ ] court shoe	12 [ ] mule
13 [ ] sandal	14 [ ] surgical/bespoke	15 [ ] other:	

4. Heel height:_____ (mm)

5. Forefoot height:_____ (mm)

6. Flexion point: 1[ ] at MTPJs 2[ ] proximal 3[ ] distal

7. Midfoot sole sagittal stability: 1[ ] minimal (>45º) 2[ ] moderate (≤ 45º) 3[ ] rigid (0-10º)

8. Fixation: 1[ ] none 2[ ] laces 3[ ] straps/buckles 4[ ] Velcro 5[ ] zips

OXFORD ANKLE AND FOOT QUESTIONNAIRE FOR CHILDREN – PARENT VERSION

The following questions are based on how children and adolescents have previously expressed how they have been affected by a problem to their feet or ankles.

Please fill out this questionnaire to the best of your ability and place a tick under each answer that describes your child or adolescent best for each question.

Considering the previous week,

1. Has your child found walking difficult because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

2. Has your child found it difficult to run because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

3. Has it been difficult for your child to stand up for long periods?

never	rarely	sometimes	very often	always
○	○	○	○	○

4. Has your child had pain in their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

5. Have your child's legs been sore or ached after walking or running?

never	rarely	sometimes	very often	always
○	○	○	○	○

6. Has your child felt tired because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

7. Has your child's foot or ankle stopped them joining in with others in the playground?

never	rarely	sometimes	very often	always
○	○	○	○	○

8. Has your child's foot or ankle stopped them playing in the park or outside?

never	rarely	sometimes	very often	always
○	○	○	○	○

9. Has your child's foot or ankle stopped them taking part in physical education lessons?

never	rarely	sometimes	very often	always
○	○	○	○	○

10. Has your child's foot or ankle stopped them taking part in any other lessons at school?

never	rarely	sometimes	very often	always
○	○	○	○	○

11. Has your child been bothered by how their foot or ankle looks?

never	rarely	sometimes	very often	always
○	○	○	○	○

12. Has the way your child walks bothered them?

never	rarely	sometimes	very often	always
○	○	○	○	○

13. Has your child been embarrassed because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

14. Has anyone been unkind to your child because of their foot or ankle?

never	rarely	sometimes	very often	always
○	○	○	○	○

15. Has your child's foot or ankle stopped them wearing any shoes they wanted to wear?

never	rarely	sometimes	very often	always
○	○	○	○	○

Checklist for investigators

Tick	CHECKLIST
	Participant details obtained and accurate
	Informed consent obtained
	All items of assessment have been performed
	Scans have been checked for usability
	Carbon print clear and completed for both feet

LA TROBE UNIVERSITY
College of Science Health and Engineering
School of Allied Health, Human Services and Sport

Mailing address
La Trobe University
Victoria 3086 Australia
T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

Date

Recipient title, first name, last name

Address, postal or email

Suburb, State, post code

Dear Enter name of parent

Thank you for participating in our research project titled *Differences in foot anthropometry and foot health in children and adolescents with and without Down syndrome.* On the day of the assessment, I carried out a foot health assessment and have summarised the results below.

RESULTS	
Presence of toe deformity:	Choose an item.
Bunionette:	Choose an item.
Skin and nail assessment for the presence of:	
• Corns:	Choose an item.
• Callus:	Choose an item.
• Ingrown toenails:	Choose an item.
• Maceration between toes:	Choose an item.
• Wart(s):	Choose an item.
• Tinea of the skin:	Choose an item.
• Blisters:	Choose an item.
• Other:	Choose an item.
Footwear assessment:	
• Style/characteristics of footwear:	Choose an item.
• Fit of footwear:	Choose an item.

Comments: Click here to enter text.

College of Science Health and Engineering
School of Allied Health, Human Services and Sport

Mailing address
La Trobe University
Victoria 3086 Australia

T + 61 3 9479 5815
F + 61 3 9479 5737
E health@latrobe.edu.au
latrobe.edu.au/health

Recommend follow-up by a podiatrist: Choose an item.

Kind regards,

Nirmeen Hassan

PhD student
Discipline of Podiatry
School of Allied Health, Human Services and Sport, College of Science, Health and Engineering
La Trobe University, Victoria, 3086, Australia
HS3 room 523a

Discipline of Podiatry
School of Allied Health, Human Services and Sport
COLLEGE OF SHE (SCIENCE, HEALTH & ENGINEERING)

Student Podiatry Clinic
Health Sciences Clinic
La Trobe University
Bundoora Victoria 3086
T +61 3 9479 5831
F +61 3 9479 5036
E podiatry@latrobe.edu.au
www.latrobe.edu.au/podiatry

30/4/19

To whom it may concern,

This is a letter to confirm that I have read the ethics for Nirmeen Hassan's research project titled 'Differences in foot anthropometry and foot health in children and adolescents with and without Down Syndrome'.

As clinic manager I approve the use of the clinic for Nirmeen to undertake her project.

Kind regards,

Kim Holmes
Clinic Manager, Health Sciences Clinic
External Clinical and 4th Year Coordinator, Podiatry Lecturer.
Discipline of Podiatry
School of Allied Health, Human Services and Sport
College of Science, Health and Engineering
La Trobe University | Bundoora, Victoria 3086, Australia
T: 03 9479 3364 | E: kim.holmes@latrobe.edu.au | W: www.latrobe.edu.au

Appendix 9. Publication acceptance correspondence

From: Journal of Intellectual Disability Research <onbehalfof@manuscriptcentral.com>
Sent: Tuesday, October 2, 2018 5:33:33 AM
To: Nirmeen Hassan <N.Hassan@latrobe.edu.au>; Shannon Munteanu <S.Munteanu@latrobe.edu.au>; Karl Landorf <K.Landorf@latrobe.edu.au>; Nora Shields <N.Shields@latrobe.edu.au>
Cc: jidr.editorialoffice@wiley.com <jidr.editorialoffice@wiley.com>
Subject: Journal of Intellectual Disability Research - Decision on Manuscript JIDR-11-2017-0251-OM.R1

01-Oct-2018

Dear Ms Hassan,

I am pleased to accept your manuscript entitled "Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials" for publication in the Journal of Intellectual Disability Research.

Your article cannot be published until you have signed the appropriate license agreement. Within the next few days you will receive an email from Wiley's Author Services system which will ask you to log in and will present you with the appropriate licence for completion.

When you receive your proofs please check them carefully especially author names and affiliations.

You will be pleased to know that the journal now has 'online early', which means that, as soon as your corrected proofs are accepted and have been laid out, your paper will be available to view on the JIDR website and can be cited.

Thank you for your contribution. We look forward to your continued contributions to the Journal.

Yours sincerely,

Professor Craig Melville
Editor in Chief, Journal of Intellectual Disability Research
craig.melville@glasgow.ac.uk

From: Disability and Rehabilitation <onbehalfof@manuscriptcentral.com>
Sent: Monday, November 11, 2019 1:03:37 AM
To: Nirmeen Hassan <N.Hassan@latrobe.edu.au>; NIRMEEN HASSAN <nmhassan@students.latrobe.edu.au>
Cc: Nirmeen Hassan <N.Hassan@latrobe.edu.au>; NIRMEEN HASSAN <nmhassan@students.latrobe.edu.au>; Nora Shields <N.Shields@latrobe.edu.au>; Karl Landorf <K.Landorf@latrobe.edu.au>; Andrew Buldt <A.Buldt@latrobe.edu.au>; Nicholas Taylor <N.Taylor@latrobe.edu.au>; Angela Evans <Angela.Evans@latrobe.edu.au>; Cylie.williams@monash.edu <Cylie.williams@monash.edu>; Hylton Menz <H.Menz@latrobe.edu.au>; Shannon Munteanu <S.Munteanu@latrobe.edu.au>
Subject: Disability and Rehabilitation - Decision on Manuscript ID TIDS-06-2019-038.R2

10-Nov-2019

Dear Ms Hassan:

Ref: Efficacy of custom-fitted footwear to increase physical activity in children and adolescents with Down syndrome (ShoeFIT): randomised pilot study

Our referees have now considered your paper and have recommended publication in Disability and Rehabilitation. We are pleased to accept your paper in its current form which will now be forwarded to the publisher for copy editing and typesetting.

You will receive proofs for checking, and instructions for transfer of copyright in due course.

The publisher also requests that proofs are checked and returned within 48 hours of receipt.

Thank you for your contribution to Disability and Rehabilitation and we look forward to receiving further submissions from you.

Sincerely,
Professor Muller
Editor in Chief, Disability and Rehabilitation
davemuller01@btinternet.com

From: em.jfar.0.6b8248.829ba0b7@editorialmanager.com <em.jfar.0.6b8248.829ba0b7@editorialmanager.com> on behalf of Journal of Foot and Ankle Research Editorial Office <em@editorialmanager.com>
Sent: Monday, May 25, 2020 7:54:06 PM
To: Nirmeen Hassan <N.Hassan@latrobe.edu.au>
Subject: Decision has been reached on your submission to Journal of Foot and Ankle Research - JFAR-D-20-00043R1

JFAR-D-20-00043R1
Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome
Nirmeen Hassan; Andrew K Buldt, PhD; Nora Shields, PhD; Karl B Landorf, PhD; Hylton B Menz, PhD; Shannon E Munteanu, PhD
Journal of Foot and Ankle Research

Dear Mrs Hassan,

I am pleased to inform you that your manuscript "Reproducibility of foot dimensions measured from 3-dimensional foot scans in children and adolescents with Down syndrome" (JFAR-D-20-00043R1) has been accepted for publication in Journal of Foot and Ankle Research.

Before publication, our production team will check the format of your manuscript to ensure that it conforms to the standards of the journal. They will be in touch shortly to request any necessary changes, or to confirm that none are needed.

Articles in this journal may be held for a short period of time prior to publication. If you have any concerns please contact the journal.

Any final comments from our reviewers or editors can be found, below. Please quote your manuscript number, JFAR-D-20-00043R1, when inquiring about this submission.

We look forward to publishing your manuscript and I do hope you will consider Journal of Foot and Ankle Research again in the future.

Best wishes,

Gordon J Hendry, PhD, BSc (hons), PgC
Journal of Foot and Ankle Research
https://jfootankleres.biomedcentral.com/

Comments:

Reviewer #2: Much improved and thank you for taking on board my comments. Great to see more research in the field of Podiatry and Down syndrome.

--
Please also take a moment to check our website at https://www.editorialmanager.com/jfar/l.asp?i=40077&l=BH1PYJD0 for any additional comments that were saved as attachments. Please note that as Journal of Foot and Ankle Research has a policy of open peer review, you will be able to see the names of the reviewers.

As a result of the significant disruption that is being caused by the COVID-19 pandemic we are very aware that many researchers will have difficulty in meeting the timelines associated with our peer review process during normal times. Please do let us know if you need additional time. Our systems will continue to remind you of the original timelines but we intend to be highly flexible at this time.

This letter contains confidential information, is for your own use, and should not be forwarded to third parties.

Recipients of this email are registered users within the Editorial Manager database for this journal. We will keep your information on file to use in the process of submitting, evaluating and publishing a manuscript. For more information on how we use your personal details please see our privacy policy at https://www.springernature.com/production-privacy-policy. If you no longer wish to receive messages from this journal or you have questions regarding database management, please contact the Publication Office at the link below.

_____

In compliance with data protection regulations, you may request that we remove your personal registration details at any time. (Use the following URL: https://www.editorialmanager.com/jfar/login.asp?a=r). Please contact the publication office if you have any questions.

Appendix 10. Copyright permissions

Items subject to copyright:

1. Chapter 2. Hassan et al. 2019. (licence permission included for the Journal of Intellectual Disability Research)
2. Chapter 3. Hassan et al. 2019. Permission not required unless thesis is commercially published.
3. Chapter 4. Hassan et al. 2020. Permission not required as per Creative Commons Attribution (CC – BY). Further information available at: https://www.biomedcentral.com/about/policies/reprints-and-permissions
4. Chapter 5. Hassan et al. 2020. Manuscript submitted for publication. Permission not required unless thesis is commercially published.

Type of use	Dissertation/Thesis
Requestor type	Author of this Wiley article
Format	Electronic
Portion	Full article
Will you be translating?	No
Title	Footwear, foot dimensions and physical activity in children with Down syndrome
Institution name	La Trobe University
Expected presentation date	Sep 2020
Order reference number	01040920
Requestor Location	Ms. Nirmeen Hassan La Trobe University Plenty rd and Kingsbury Drive Bundoora Melbourne, Victoria 3086 Australia Attn: Ms. Nirmeen Hassan
Publisher Tax ID	EU826007151
Total	0.00 AUD

Terms and Conditions

TERMS AND CONDITIONS

This copyrighted material is owned by or exclusively licensed to John Wiley & Sons, Inc. or one of its group companies (each a"Wiley Company") or handled on behalf of a society with which a Wiley Company has exclusive publishing rights in relation to a particular work (collectively "WILEY"). By clicking "accept" in connection with completing this licensing transaction, you agree that the following terms and conditions apply to this transaction (along with the billing and payment terms and conditions established by the Copyright Clearance Center Inc., ("CCC's Billing and Payment terms and conditions"), at the time that you opened your RightsLink account (these are available at any time at http://myaccount.copyright.com).

JOHN WILEY AND SONS LICENSE
TERMS AND CONDITIONS

Sep 03, 2020

This Agreement between La Trobe University -- Nirmeen Hassan ("You") and John Wiley and Sons ("John Wiley and Sons") consists of your license details and the terms and conditions provided by John Wiley and Sons and Copyright Clearance Center.

License Number	4901630113219
License date	Sep 03, 2020
Licensed Content Publisher	John Wiley and Sons
Licensed Content Publication	Journal of Intellectual Disability Research
Licensed Content Title	Effectiveness of interventions to increase physical activity in individuals with intellectual disabilities: a systematic review of randomised controlled trials
Licensed Content Author	N. M. Hassan, K. B. Landorf, N. Shields, et al
Licensed Content Date	Nov 8, 2018
Licensed Content Volume	63
Licensed Content Issue	2
Licensed Content Pages	24