School of Physiotherapy and Exercise Science

Running in children and adolescents with cerebral palsy – a biomechanical analysis and intervention study

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Author's Declaration

I declare that this thesis is my own account of my research. To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made. This thesis contains no material which has been accepted for the award of any other degree or diploma in any university. This thesis contains four published papers and two papers prepared for submission. The statements of contribution of co-authors in these papers are presented in the Appendices. The version of each published manuscript is included in compliance with copyright requirements of the respective publishers and Curtin University. All references were standardised to the style output of Developmental Medicine and Child Neurology to maintain consistency in formatting throughout the thesis. All copyright material has been reproduced with permission and copies of all permissions appear as Appendices.

Human Ethics: The research presented and reported in this thesis was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. The proposed research studies received human research ethics approval from the Child and Adolescent Health Service Ethics Committee, Perth, Western Australia (201405SEP) and the Human Research Ethics Committee of Curtin University, Perth, Western Australia (HR 219/2014). The trial was prospectively registered with the Australian New Zealand Clinical Trials Notification ACTRN12614000467639.

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Abstract

Running is a fundamental movement skill which is important for participation in play, sport, and recreational activities. Running skill is a facilitator to an active lifestyle which promotes health and wellbeing. Children with cerebral palsy (CP) do not run as well as their typically developing peers and part of that limitation is likely to be due to the biomechanics of their running. This thesis investigated the effect of a task-specific, low-load plyometric running training program on the biomechanics of running in children and adolescents with CP in Gross Motor Function Classification Scale (GMFCS) Levels I and II. The aims of this thesis were:

1. To systematically review the existing literature on running in people with CP.

2. To describe lower limb power generation at a range of running speeds in children and adolescents with CP compared to the typically developing (TD) population.

3. To estimate the function of the stretch shortening cycle at a range of running speeds through an examination of leg stiffness in children and adolescents with CP, compared to the TD population.

4. To investigate the effect of a running training program developed for adult neurological rehabilitation on lower limb power generation and stretch-shortening cycle function in children and adolescents with CP.

To address the first aim, a systematic review of the literature was undertaken in 2016 according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines and updated in 2020. The review is presented in Chapter 2 of this thesis. The aims of the review were: 1) to review what is known about how people with CP run, and 2) to review what is known about the effect of interventions on improving running in people with CP. The review found that people with CP: 1) run more slowly than their able-bodied peers; 2) run with a shorter stride length; 3) run with a higher incidence of forefoot striking; and 4) that the characteristics of running in people with unilateral CP are different to those with bilateral CP. Included studies suggested that interventions that include running are feasible and safe in children and adolescents with CP and may result in functional improvements. However, there remained no information in the published literature regarding training of running skill in people with CP, nor the effect of intervention

on the biomechanics of running.

This body of work addressed two main types of questions and therefore employed two main types of research design. The primary question was “what is the effect of a running intervention in children with CP?” and to answer this question an RCT design was employed. Results of the RCT are presented in Chapters 7 and 8. However, prior to addressing this question, a more fundamental question had to be addressed - “what are the biomechanical differences during running between children with CP and those who are TD?” To answer this question, a cross-sectional design was employed, which utilised data from the baseline collection. Theses analyses are presented in Chapters 5 and 6. Prior to embarking on these analyses, there were some methodological considerations which needed to be addressed. An analysis of modelling techniques for three dimensional gait analysis (3DGA) was undertaken and is presented in Chapter 4.

A randomised controlled trial (RCT) assessing the effectiveness of a task-specific, low-load plyometric (i.e. rapid eccentric then concentric muscle action[1]) running training programme was undertaken. The control group was offered the intervention at the conclusion of the RCT. The intervention was conducted in a group setting but individualised to each participant and consisted of a hierarchy of activities focusing on ankle power generation, hip power generation and optimal running technique. Children and adolescents with CP, aged 9-18 years, who could walk 10m unaided, were recruited from a community service provider and underwent stratified randomisation into control (n=22, mean age=12 years 10 months) or intervention groups (n=21, mean age=13 years 0 months). Following baseline assessments, the intervention group received 12-weeks of a twice weekly intervention program with a home program, instead of usual care. The control group received usual care which in most cases consisted of the provision of a home exercise program. Appendix 1 presents a paper which reported that the training program was effective for running-related goal attainment and maintaining school participation.

In conjunction with this clinically based RCT, a concurrent biomechanical study was implemented, which forms the work of this thesis. Participants in the broader RCT who did not have a flight phase were excluded from biomechanical analysis. This included two participants in GMFCS level II and one participant in GMFCS level III who could walk 10m unaided. All participants attended a baseline assessment session at the motion analysis laboratory, where kinematic and kinetic data were collected for jogging, running, and sprinting. In addition, leg stiffness data, as measured by submaximal hopping, was collected on a custom-built sled. Baseline data were compared with data collected at a single time

point from a sample of typically developing children aged 10-12 years (n=21). At the conclusion of the 12-week intervention period participants returned to the motion analysis laboratory to repeat the assessments.In addressing the second aim of the thesis, analysis of baseline data revealed that maximum sprint speed and ankle power generation at push-off were reduced in children with CP GMFCS Level I compared to typically developing children and were further reduced in children with CP in GMFCS Level II. For all children with CP, ankle power generation was significantly reduced in affected legs compared to non-affected legs. In affected legs, hip power generation during swing was relatively increased. This work is presented in Chapter 5.

In addressing the third aim of this thesis, analysis of baseline data for stretch-shortening cycle (SSC) function revealed that leg stiffness was reduced in children with CP compared to TD children during jogging and running. Affected legs displayed reduced leg stiffness compared to non-affected legs only in GFMCS Level I during running and sprinting. This work is presented in Chapter 6.

Analysis of the RCT data was undertaken to address the fourth aim of this thesis. At follow-up, normalised speed of running had increased in the intervention group while the control group were slower. There was no change in sprint speed. In running, children in GMFCS Level II in the intervention group demonstrated increased ankle power generation while the control group had no change. In sprinting, children in both GMFCS Level I and Level II in the intervention group maintained ankle power generation, while ankle power generation was decreased in the control group. Most within-group differences did not translate to significant between-group differences at follow-up. At follow-up, the intervention group had greater leg stiffness compared to the control group during submaximal hopping. Participants in GMFCS Level I in the intervention group in had greater leg stiffness than the control group during jogging. These analyses are presented in Chapters 7 and 8.

Significance of the research: The contribution of ankle plantarflexor power generation to forward propulsion in running is reduced in young people with CP and is related to GMFCS level. This deficit appears to be compensated in part by increased hip flexor power generation during swing, but maximum sprinting speed is limited. Children with CP also have atypical leg stiffness profiles which implies that the function of the stretch-shortening cycle is sub-optimal. Leg stiffness profiles are different for GMFCS Level I and Level II. A low-load plyometric intervention to improve the skill of running in children and adolescents

with CP can change running biomechanics, specifically, improvements in ankle power generation for running in children in GMFCS Level II and improved leg stiffness in GMFCS Level I.

This body of work will form a foundation for further investigation of running training in people with CP, including an investigation of how plantarflexor spasticity and contracture relate to ankle power generation or SSC function, the optimal dose of intervention, how long improvements are maintained following intervention, and whether intervention should be differently tailored for children in GMFCS Levels I or II. This work could also be replicated in other neurodevelopmental diagnostic groups.

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I am grateful to the Lord God and to my family, who provide a firm foundation and a reminder of what is important.

Our project was born when Dr Noula Gibson approached Ability Centre to ask if we would be interested in working together to adapt and evaluate a running training program for children with CP. I subsequently embarked upon a PhD candidature which would add an examination of biomechanics to the measures of activity and participation already planned. Outcomes in the activity and participation domains were published with Dr. Gibson as first author and myself as second author; this article is included as Appendix 1. I am grateful for the considerable amount of work my supervisory team have done in raising me through this candidacy. Supervisors to this thesis were:

Professor Garry T Allison and Dr Susan Morris

Dr Gavin Williams and Dr Noula Gibson

The supervisory team have each brought their own perspective and strengths and have been responsive, dedicated, and professional while maintaining a high standard of integrity. It was a blessing to have supervisors that I also enjoyed as people. Thank you.

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Research outputs during the doctoral candidature

Published papers

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Chappell A, Allison GT, Williams G, Gibson N, Morris S. The effect of a running training intervention on ankle power generation in children and adolescents with cerebral palsy: A randomized controlled trial. Clinical Biomechanics. 2020 Jun 1;76:105024.https://doi.org/10.1016/j.clinbiomech.2020.105024.

Chappell A, Gibson N, Williams G, Allison GT, Morris S. Propulsion strategy in running in children and adolescents with cerebral palsy. Gait & posture. 2019 May 1;70:305-10.https://doi.org/10.1016/j.gaitpost.2019.02.018

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Gibson N, **Chappell A,** Blackmore AM, Morris S, Williams G, Bear N, Allison G. The effect of a running intervention on running ability and participation in children with cerebral palsy: a randomized controlled trial. Disability and rehabilitation. 2018 Dec 4;40(25):3041-9.https://doi.org/10.1080/09638288.2017.1367426

Manuscripts prepared for publication

Chappell, A., Allison, G.T., Gibson, N., Williams, G. and Morris, S. The effect of running intervention on leg stiffness in youth with cerebral palsy: a randomised controlled trial. Submitted for publication.

Platform presentations

Chappell A. The effect of a running training program on the skill of running in children and adolescents with cerebral palsy. Mark Liveris Seminar, Curtin University, 2020.

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Chappell, A., Gibson, N., Williams, G., Allison, G. T., & Morris, S. Propulsion strategy in running in children and adolescents with cerebral palsy. Paper presented at the AusACPDM conference, Auckland New Zealand, 2018.

Chappell, A., Williams, G., Gibson, N., Allison, G. T., & Morris, S. Rehabilitation of running in children. Workshop presented at the APA Conference, Sydney NSW, 2017.

Chappell, A., Gibson, N., Williams, G., Allison, G., Morris, S. Muscle power generation for running in young people with cerebral palsy. Poster presented to CAHS Research Symposium at Telethon Kids, Perth WA, 2017.

Invited lectures, courses and seminars

2020	Staff training, Western Kids Health, Perth
2020	Xcelerate staff training, Ability Centre, Perth
2017	APA course: Rehabilitation of running in children, Adelaide
2016	APA course: Rehabilitation of Gait and Running, Perth
2016	Rehabilitation of running in children workshop, AusACPDM Conference, Adelaide

Implementation

The running training program has been offered as a clinical service since the randomised controlled trial, as a program called Xcelerate. I continue to oversee Xcelerate and provide regular training to our physiotherapists to ensure treatment fidelity. The knowledge we have gained through this body of work therefore continues to be further evaluated, broadened and refined because of the close relationship between clinical research and clinical practice.

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List of Abbreviations

3DGA	Three-Dimensional Gait Analysis
ABI	Acquired Brain Injury
BMI	Body Mass Index
CERT	Consensus on Exercise Reporting Template
CoM	Centre of Mass
CP	Cerebral Palsy
DLAA	Developmental Level of the Arm Action
DoF	Degrees of Freedom
GAS	Goal Attainment Scaling
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
GRF	Ground reaction force
HELP	Hawaii Early Learning Profile
HiMAT	High-Level Mobility Assessment Tool
IK	Inverse Kinematics
LIC	Length at initial contact
MPST	Muscle Power Sprint Test
RoM	Range of Motion
RSA	Running Speed and Agility
SRT	Shuttle Run Test
SSC	Stretch Shortening Cycle
STA	Soft Tissue Artefact
TD	Typically Developing

Chapter 1 Background

1.1 Cerebral palsy: aetiology and classification

The term cerebral palsy (CP) is an umbrella term used to describe a group of conditions which result in disorders of movement and posture, due to a non-progressive defect or lesion in the developing brain[2]. In high-income countries such as Australia, about 1 in 500 children are diagnosed with CP[3]. Causes of CP include hypoxia, genetic defects, infections, congenital anomalies and inborn errors of metabolism[4]. Due to the diverse nature of conditions falling under the descriptor "CP", individuals with CP are a heterogenous population[2, 4]. When seeking to describe people with CP, clinicians usually consider topography, motor impairment, functional ability and comorbidities[5].

Depending on the site and extent of the central nervous system lesion, CP can be described topographically as affecting one or both sides of the body (hemiplegia/unilateral or bilateral), lower limbs only (diplegia), the whole body (quadriplegia), three limbs (triplegia) or one limb (monoplegia)[5]. Classification by topography is difficult because an "unaffected" limb often has subtle neurological signs and it can be difficult to distinguish between unilateral and bilateral, or diplegia and quadriplegia[5]. Again, according to the nature and site of central nervous system injury, CP can be described according to motor impairment. Muscle tone can be hypotonic, hypertonic (spasticity, dystonia, rigidity)[6], dyskinetic (chorea, athetosis), ataxic or mixed[5]. Spasticity is the most common motor disorder, followed by ataxia and then dyskinesia[7]. Classification by motor impairment is complicated by the co-existence of different types of muscle tone in different body regions, or even within the same body region. The presentation of motor impairment can emerge and evolve, particularly over the first two years of life[8].

Cerebral palsy can be described according to functional ability, most commonly by the Gross Motor Function Classification System (GMFCS)[9], the Manual Ability Classification System[10] and the Communication Function Classification System[11]. Classification by functional level assists in determining a person's needs, making management decisions and anticipating future motor function[12]. The validity, reliability and clinical utility of the GMFCS has been well established[9]. The GMFCS classifies children and adolescents with CP into one of five levels of gross motor function, with Level I the most able and Level V the least able. Just over 60% of all children with CP are classified as either GMFCS Level I or II[13]. In the

early 2000's the GMFCS was expanded and revised under the influence of the International Classification of Functioning, Disability and Health[14], to recognise "…that performance of gross motor function is influenced by the physical, social, and attitudinal environment and personal factors such as preferences, interests, and motivation…" (Palisano, Rosenbaum, Bartlett and Livingston, 2008 p744)[14]. The expansion included a 12 to 18 year-old band. Within this band, adolescents classified as Level I are described as able to run, although they may lack speed or coordination[9]. Within GMFCS Level II, over half in this band have been reported as able to run[15]. Children in GMFCS Level II take longer than children in GMFCS Level I to master the skill of running, which may emerge after the sixth birthday[9]. Children in GMFCS Level III walk using a hand-held mobility device in most indoor settings[14] and are not expected to attain running ability. Although almost half the classifications made in children under two years of age are revised[8], the GMFCS is more stable in childhood and adolescence, with 73% of children remaining in the same classification over time[16]. Reclassification occurs both down and up a level. This indicates that the role of intervention may be either improvement or maintenance. Children in GMFCS Level I are less likely to be reclassified than those in Level II[16] and the emergence of running skill is one event that may prompt reclassification up a level.

Gross motor development reference curves were developed to provide prognostic information according to GMFCS level[17]. Running ability in children with CP tends to decline after puberty, and this decline is associated with increasing Body Mass Index (BMI)[15] and decreasing participation in physical activity[18]. While youth in GMFCS Level I are likely to continue to be able to run, youth in GMFCS Level II often experience a decline in gross motor function and may lose the ability to run[14]. This sub-group of children with CP take longer to acquire running skill and lose it more quickly than children in GMFCS Level I. Youth in GMFCS Level II may eventually choose to use a hand-held mobility device at school or work as they get older[9].

In clinical practice, a person with CP is likely to be described multi-dimensionally, including topography, motor impairment, functional ability and comorbidities[5]. Over half of the CP population have at least one comorbidity; some of the more commonly occurring are epilepsy, digestive system disease, congenital malformation (excluding the nervous system) and respiratory disease[19]. Classification and standardised description of CP is important to provide families with prognostic information, facilitate communication between health care professionals and for goal setting and intervention planning[5, 14].

Classifications have also been developed for hemiplegic[20] and diplegic[21] walking gait, which have been useful to facilitate communication between health professionals. Gait deviations can be due to primary impairments such as spasticity, weakness or lack of motor control; secondary impairments such as contracture or skeletal mal-alignment; or compensations for primary or secondary impairments[22]. Gait deviations are commonly classified according to timing (stance or swing phase), plane (sagittal, frontal or transverse), and joint (ankle, knee, hip)[22]. However, it is recognised that accurate classification is limited by the heterogenous nature of the CP population[23, 24] and that walking gait classification is best used as a framework for individual analysis[22]. To date, there is no classification system for running in people with CP; however, it can be expected that any classification system for running would have similar limitations to walking classification, including limitations related to the heterogenous nature of the CP population.

1.2 Physical activity

The term physical activity refers to any movement produced by skeletal muscle which results in energy expenditure[25]. Physical activity is positively associated with metabolic and musculoskeletal health[26], executive function and academic performance[27]. In children with CP, physical activity has a positive correlation with happiness and quality of life, including health-related, physical, social and psychosocial domains[28]. While these relationships are complex, participation in physical activity has potential for change with intervention[29], while some other factors related to health and quality of life, such as GMFCS level, have limited potential for change[28].

Levels of physical activity decline as children enter adolescence, both in TD children[30] and children with CP[31]. Particularly for adolescents in GMFCS Level II, a decline in physical activity may be associated with the loss of running ability[14]. The majority of children with unilateral CP in GMFCS Levels I and II consistently fail to meet recommendations for healthy moderate to vigorous physical activity of 60 minutes per day[32]. In TD children, levels of motor coordination and/or aerobic capacity are predictive of level of physical activity[30]. Participation in running activities in adolescence has been linked to higher physical activity levels in adulthood[33], which in turn reduces morbidity and mortality[34]. Likewise in children and adolescents with CP, fitness, motor capacity and fundamental movement skill have been identified as facilitators to participation in physical activity, while lack of motor skill and lack of endurance are considered barriers[18, 35]. In this way, the ability to run can be

considered a facilitator of participation in physical activity in people with CP, with important positive impacts on health and wellbeing.

1.3 Running capacity, running skill and running performance

Running is itself a form of physical activity[25] and one of the most popular physical activities of childhood and adolescence[36]. It is also a fundamental movement skill[37] used briefly in everyday activities – to catch a bus, when it begins to rain, to get to class in time or in playing games such as "Chasey", "What's the time Mr Wolf?" or "Crocodile Crocodile". As a fundamental movement skill, running develops and matures throughout childhood[38], and can be improved through explicit teaching and practice, as has been reported in typically developing (TD) pre-school children[39]. In free-play, children typically run fast for short bursts of time lasting less than 15 seconds[40], therefore improving running skill can reasonably be expected to improve participation in playground games. Running has various aspects or domains that can be assessed in different ways. For the purposes of this thesis, it is necessary to define the terms that describe constructs of running, as there is some inconsistency in the existing literature. This section will define running capacity, running skill, and running performance.

1.3.1 Running capacity

Running capacity includes both aerobic capacity, which is related to steady-state sustained running velocity[41], and anaerobic capacity, which is related to power generation and sprinting[42]. Aerobic capacity is the ability of the cardiorespiratory system to deliver oxygen to the muscles for energy generation and can be assessed in people with CP using laboratory-based measures or field measures[43]. Laboratory-based measures include maximal exercise on a bicycle ergometer, arm-crank ergometer or treadmill[43]. The shuttle run test (SRT) is the only field-based measure which has been validated in people with CP[43].

Anaerobic capacity is the maximal anaerobic metabolism of adenosine triphosphate during bursts of high intensity exercise lasting less than 45 seconds[43]. In people with CP, anaerobic capacity can be assessed in the laboratory using the Wingate bicycle ergometer test or the Wingate arm-crank ergometer test. The muscle power sprint test (MPST) is the only field-based measure of anaerobic capacity that has been validated for people with CP[43]. Laboratory-based tests are standardised but expensive, and require more time and assessor training than field-based tests[43].

1.3.2 Running skill

Running skill refers to proficiency in the motor skill of running[44] and is related to technique. Motor proficiency is determined by the qualitative aspects of a motor task and can serve as a marker of childhood motor development[45]. Criteria and descriptors of "good" running technique will be discussed in section 1.5 of this chapter. Running technique can be quantified by biomechanics, and 3-Dimensional Gait Analysis (3DGA) is the gold standard for assessment of running biomechanics including kinematics and kinetics[46]. Two-dimensional video analysis is a much less expensive way to capture kinematics, but has limited reliability for some movements such as knee abduction[46].

1.3.3 Running performance

Running performance, or how a person performs in games or competition, is a complex interaction that includes skill, capacity, agility, cognition, vision, and emotion[47]. Running performance can be assessed by calculating linear velocity or acceleration[48, 49], and has also been examined during match play, for example in soccer[47], field hockey[50] and basketball[51].

Maximum running velocity requires high force production during a short period of ground contact[52]. Therefore, strength, power and plyometric (i.e. exercises which incorporate a rapid eccentric muscle action followed by a rapid concentric muscle action[1]) training programmes are commonly used to improve running performance[53]. Specificity of training, i.e. the proximity of a training exercise to the goal motor task, improves the transfer of gains made from training into performance[54]. As running requires rapid, cyclical force production in both the horizontal and vertical planes, these elements should be incorporated into training to maximise improvement in running performance[53].

1.4 Biomechanics of typical, mature running

Typical, mature running has received much attention in the research literature and will be outlined here in brief. Typical running will be summarised with respect to spatiotemporal characteristics; kinematics, which describe motion; and kinetics, which describe the moments of force related to motion.

1.4.1 Spatiotemporal characteristics

There are two key features of running that distinguish it from walking. Spatiotemporally,

the running stance phase is less than 50% of the gait cycle, with a flight phase between each single-limb support (Figure 1.1). In walking, the stance phase is more than 50% of the gait cycle, with a period of double-limb support between each single-limb support[55]. Secondly, in running, the centre of mass (CoM) is at its lowest point at mid-stance (in-phase), while in walking, the CoM is at its highest point at mid-stance (out-of-phase)[55]. The presence of a flight phase can be determined by using slow motion video, which is inexpensive but prone to inaccuracies, especially when the flight phase is momentary. Alternatively, 3DGA with force plates provides accurate information about the length of a flight phase and the trajectory of the CoM[46].

Left initial contact **Mid-stance** **Toe-off** **Flight** **Mid-swing**

Figure 1.1: Phases of running gait with trajectory of the centre of mass

The running gait cycle begins when one foot strikes the ground, termed initial contact, and ends when the same foot strikes the ground again (Figure 1.1)[56]. The distance between two sequential contacts of the same foot is termed stride length, while the distance between initial contact of one foot and the initial contact of the contralateral limb is termed step length[57]. The time the foot is in contact with the ground is termed stance phase and it ends with toe-off[57]. In running, stance phase occupies less than half of the gait cycle, which creates two flight phases, one between toe-off and initial contact of the contralateral limb, and another at the end of swing, prior to initial contact[56]. As running velocity increases, the percentage of stance time decreases to as little as 22%[58] and the percentage of flight time increases correspondingly[56].

Running speed can be considered to fall broadly into three categories termed jogging, running, and sprinting. *Jogging* is the slowest category, and may be considered as speeds up to 60% of maximum running speed[59]. *Running* involves a primarily aerobic metabolism, allowing the pace to be maintained for longer distances than sprinting[56]. The goal of *sprinting* is to cover a short distance at top speed and it is primarily fuelled by anaerobic

power[56]. Transition to a forefoot strike is a marker of change from running to sprinting[56].

The velocity of the runner may be measured in metres per second or kilometres per hour. It may also be reported in terms of cadence, also termed step frequency, which is the number of steps per unit of time, either minutes or seconds. Cycle time is the time it takes to complete one gait cycle[58, 60]. In running, spatial and temporal parameters are interrelated[61]. Initial increases in velocity are achieved by increasing stride length, but at speeds above 5ms^{-1} the stride length is optimised and cadence is increased to attain higher velocities[62].

1.4.2 Kinematics

The range of motion (RoM) required for typical running is greater at all lower limb joints compared to walking[58] (Figure 1.2). In running, pronation of the foot is observed until mid-stance followed by supination to provide a rigid lever via the ankle windlass mechanism for the optimal action of the Achilles tendon at push-off[56]. As the runner approaches maximum velocity (sprinting) the ankle becomes more plantarflexed at initial contact and there is an increased incidence of forefoot strike pattern[56]. Knee RoM increases in swing and the flexion-extension wave associated with loading response becomes minimal at maximal speeds[58]. Peak hip flexion increases with higher velocities[58]. The hip is adducted during stance and abducted in swing, which minimises trunk and head movement in space[56], providing a stable base to the limbs.

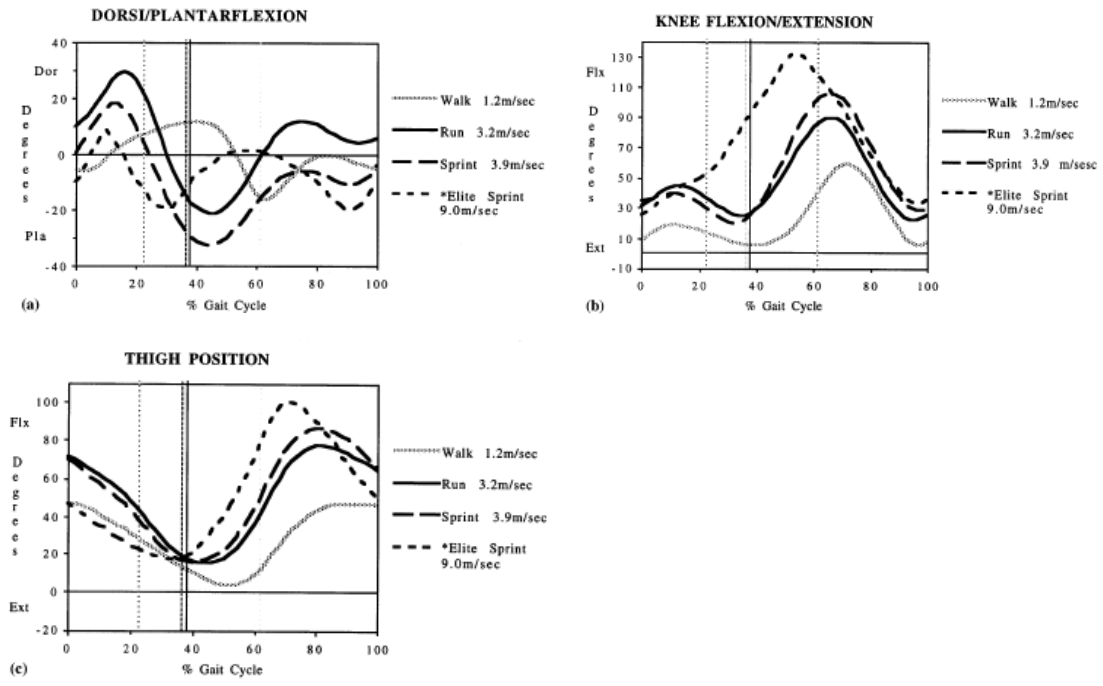


Figure 1.2: Sagittal plane kinematics of the ankle, knee and hip (reproduced with permission; Novacheck, 1998 p82 Figure 6)

Studies have proposed the following as markers of good running technique in adults:

1. Slight forward lean of the trunk[63] or upright posture[64]
2. Arms do not cross the midline and elbows are flexed at about 90°[63, 65]
3. Foot strike directly under the centre of mass and in line with the head[66, 67]
4. Some flexion of the hip and knee at initial contact[63] ("light and quiet"[64])
5. Internal knee and ankle progression angle in stance[65]
6. Greater hip extensor power generation at initial contact[65]
7. Greater knee flexion in swing[63]
8. Greater leg stiffness[68]
9. Minimal vertical oscillation of the centre of gravity[63, 66, 68, 69]
10. A cadence of at least 180 steps per minute[64, 65, 70]
11. Stride length may be self-selected[68], shorter[65-67, 70] or longer[63, 71] depending on the goal to be achieved

1.4.2.1 Foot strike pattern

Authors differ in their recommendations regarding foot strike (rearfoot initial contact[63], midfoot initial contact[66] or forefoot initial contact[64]) and indeed the recommendations should differ according to whether the goal is running economy or maximum speed. Nearly all sprinters use a forefoot strike, which typically eliminates the impact peak of the Ground Reaction Force (GRF)[72]. Forefoot striking requires strong eccentric activity of the plantarflexors to control ankle dorsiflexion following initial contact[73]. Only 1.5% of elite distance runners utilise a forefoot strike[74].

The incidence of forefoot strike during running or jogging decreases with age[75], from about 25% at age 3-4 years to about 5% at age 15-16 years while shod[75, 76] and 38% in adolescents when barefoot[76]. It has been suggested that a transition to a rearfoot strike may be influenced by using cushioned running shoes and/or a reduction in stride frequency with increasing age and leg length[75]. Rearfoot striking is associated with higher vertical loading rate than forefoot striking[73].

1.4.3 Kinetics

While kinematics describe the movement we observe, kinetics begin to answer the questions of 'how' and 'why'[56]. Kinetics are therefore important in understanding movement in children and adolescents with CP and will be the focus of this thesis.

The transition from walking to running typically occurs in adults at speeds of approximately 2ms^{-1} when peak vertical and anteroposterior GRFs during the propulsive phase start to decrease[77]. The observed decrease in GRFs are linked to a reduction in the force produced by the plantarflexors at push-off as they operate at shorter lengths and increased velocity[77, 78]. This requires a compensatory increase in hip power generation during stance and swing, above the requirements for running at the same velocity[78]. Transition to a running gait at this speed results in an increase in propulsive plantarflexor force, by improving the force-length-velocity relationships[77], a decrease in hip power generation and an overall decrease in muscular effort[78].

The modern nomenclature for power generation in gait is retained from early work in this field. During running, the CoM is accelerated forward only during the last 40% of stance phase, which constitutes the propulsive phase of stance[79]. The primary power generator for forward propulsion in running are the ankle plantarflexors during push-off (A2)[56, 80] (Figure 1.3). The ankle plantarflexors have short, pennate muscle fibres with a long, compliant tendon[81, 82] and are elongated in mid-stance before shortening at push-off[81]. These features make them ideally suited to the efficient storage and recycling of elastic energy utilising the stretch-shortening cycle, which reduces the work of the muscle fibres and therefore expenditure of metabolic energy[68, 81, 82]. The plantarflexors provide a large percentage of propulsive power at slower jogging speeds[83-85]. Maximum sprint velocity is essentially dictated by the time it takes to generate large forces to push against the ground in stance phase[52, 71]and the time it takes to recover the limb in swing[62]. At higher running speeds, ground contact time is brief and the ability of the plantarflexors to generate more power in less time becomes limited[62]. Further increases in velocity are achieved by increasing cadence[62], primarily by greater hip flexor power generation in swing, referred to as H3[83, 84]. The hip extensors contribute to forward propulsion by pulling the pelvis forward over the planted foot in the first half of stance phase (H1)[56, 80]. The knee extensors provide vertical power in stance to prevent the limb collapsing and contribute relatively little to forward propulsion[60, 80].

The relative contribution of ankle power to forward velocity has been termed propulsion strategy (PS) and is calculated using the formula A2/(A2+H3)[84, 85]. A higher PS indicates a relatively greater contribution of the plantarflexors to forward velocity and therefore higher metabolic efficiency due to use of the SSC[68, 81, 82]. In TD individuals, PS decreases with increasing velocity as the hip flexors generate power to increase cadence after plantarflexor power generation has reached a maximum[85]. In TD children, PS of 0.75-0.81 has been

reported in jogging and 0.69-0.75 in fast running[84, 85].

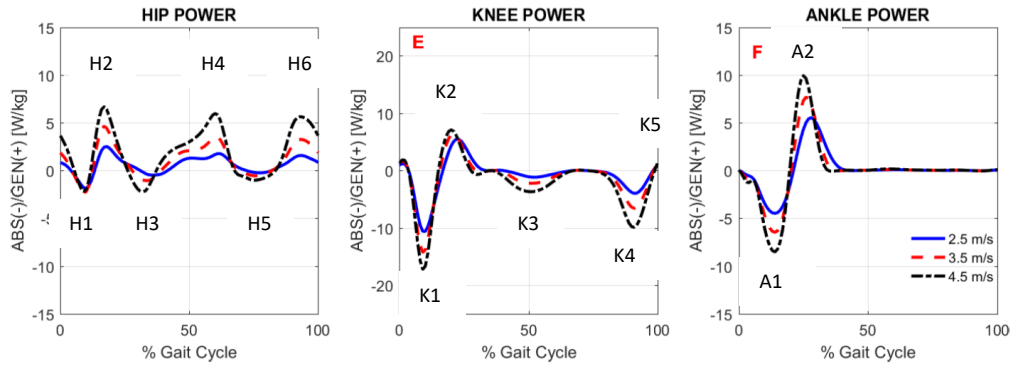


Figure 1.3: Hip, knee, and ankle powers in the sagittal plane at 2.5m/s, 3.5m/s, and 4.5m/s (Reproduced with permission; Fukuchi RK, Fukuchi CA, Duarte M. 2017. A public dataset of running biomechanics and the effects of running speed on lower extremity kinematics and kinetics. *PeerJ* 5:e3298 https://doi.org/10.7717/peerj.3298)[86]

Reflecting the SSC at each joint, there is a period of power absorption (A1, K1, H2) prior to power generation (A2, K2, H3)[56, 80, 82]. Maximising use of the SSC at the hip, knee and ankle optimises running function[82]. This requires a complex interaction of kinematics, connective tissue architecture[87], relative lengths of muscles and their tendons[87], neuromuscular control and reflex activity[88]. The gastrocnemius muscle has a long tendon and large pennation angle, which makes it more efficient than knee or hip musculature in performing mechanical work[89]. The Achilles tendon stores elastic energy during stretch and returns it on recoil, while also optimising muscle fascicle length and minimising mechanical energy expenditure[82]. When the SSC of the ankle plantarflexors is interrupted by changes in muscle reflex activity or muscle length, or a power deficit exists, mechanical work must be shifted proximally to compensate, typically to the hip[83]. Shifting mechanical work proximally to musculotendinous units with short, stiff tendons less suited to the SSC increases metabolic energy consumption[89].

1.5 The development of running skill

There is limited information in the research literature describing the early childhood developmental sequence which results in mastery of running skill. The Test of Gross Motor Development-2[90] describes four performance criteria for running, including the reciprocal swinging of bent arms, the presence of a flight phase, a narrow base of support with forefoot or rearfoot strike (but not a midfoot strike) and approximately 90° of knee flexion

in swing. As the Test of Gross Motor Development-2[90] is a criterion-referenced measure, there is no provision to describe the maturational process. The Developmental Level of the Arm Action (DLAA)[38] has been used to classify running skill according to maturational level[91]. It describes four levels of the arm action during running, with level 4 being the most mature.

Running as a motor skill emerges about six months after independent walking, between 18 and 24 months of age[92]. The ability to increase horizontal speed and "appear" to be running, or pre-running, occurs prior to the ability to generate sufficient vertical displacement for a consistent flight phase[93]. The emergence of running ability appears to be linked to the ability to generate sufficient ankle power at push-off to launch the body into a flight phase, and sufficient postural control to maintain stability of the trunk at higher velocities[93]. These abilities are developed incrementally as the young walker experiments with gradually reducing time spent in double support[94].

Initially, running is characterised by short stride length and high cadence with a short flight phase and midfoot initial contact[95, 96]. The movement of the CoM and the range of motion of the knee joint resemble walking rather than mature running, with the high point occurring in mid-stance[93]. The arms are held stiffly in high or medium guard with small movements in reaction to shifts in equilibrium[91]. The symmetry of both stride length and cycle time is established very early in running practice[97]. The ability to stop running without using an external support (such as a wall or a parent) is a distinct motor skill and emerges within six months of learning to run[92]. This phase corresponds to level 1 of the DLAA[38, 91].

The transition from level 1 to level 2 of the DLAA at about age three is associated with reduced step frequency[91]. The trajectory of the CoM now follows a mature pattern with the low point at mid-stance, the ankle and knee joints show increased range of motion with adult-like consistency[93]. Spinal rotation emerges and the arms swing to counterbalance the movement of the pelvis and leg swing[91]. At a given speed, children of this age do the same amount of internal work per unit of body mass as adults, despite differences in step frequency and body segment proportions[98]. However, the relative shortness of the lower limbs means the angle of the shank at initial contact is greater compared to adults at any given step length, which causes a greater braking force and therefore a greater amount of horizontal work per step[98] to move the CoM forward.

The transition to level 3 of the DLAA is marked by another decrease in step frequency and a

reduction in stance time[91]. Flight time increases, along with hip and knee flexion in swing and plantarflexion at initial contact[96], which may be linked to the observed reduction in vertical stiffness[91]. This phase generally occurs between the ages of four and six years, when complex multi-joint coordination emerges with increasing sensorimotor integration[99]. It is a phase when movements may be jerkier and less efficient[96]. Arm movement is initiated by spinal rotation, the elbows flex as they swing forward and extend as they swing backwards[91]. During this phase children master the ability to change direction quickly (agility)[92].

The transition to the last and fourth level of the DLAA is associated with a reduction in anteroposterior displacement, marking the acquisition of a smooth, efficient running gait[91]. Vertical stiffness decreases as shock absorption at initial contact is improved[91]. The elbow is maintained at 90 degrees of flexion and the arm action is driven by the shoulder[91]. This phase emerges at about age seven[91], but although walking gait matures at the age of seven years, running gait continues to mature until at least the age of nine years[90, 100]. Between the ages of seven and ten years, children master head stabilisation in space and the trunk becomes an efficient base of support for the limbs[99]. This phase is characterised by refinement of motor recruitment and coordination, with increased capacity to generate power using the stretch-shortening cycle through modulation of leg and vertical stiffness[101].

During adolescence, gender differences in running capacity become apparent due to hormonal influences on muscle mass and limb length[71]. Between the ages of 11 and 15 years there is increased compliance of the Achilles tendon which results in increased stance time, decreased cadence and the often observed “adolescent awkwardness”[102, 103]. Following peak height velocity (a biological marker for maturity and growth[104]), leg stiffness, cadence and step length increase, while stance time decreases. Changes in performance are related to increases in anaerobic power capacity[103].

1.6 Classification of mature, typical running gait

The particular spatiotemporal, kinematic and kinetic characteristics of each individual’s running gait results in a running pattern or style that is individual but may have characteristics in common with other runners[105]. Few authors have attempted classification of typical running gait, due to the many permutations of contributing factors and the changing nature of running patterns with changes in velocity. Two patterns are described in the literature with the goal of changing specific biomechanical parameters, ‘Pose

running'[106] and 'Groucho running'[107], and one other study proposes classifying running style as either 'aerial' or 'terrestrial' to facilitate coaching and training[105].

'Pose running' is a style which was developed to reduce GRF and peak patellofemoral compressive force, and was proposed to have a role in the rehabilitation of running injuries [106]. Pose running style is defined by a midfoot strike and a stance posture with forward lean of the trunk and vertical alignment of the shoulder, hip and heel on the stance side[106]. This style results in shorter step length, shorter contact time, reduced vertical impact peak and greater knee flexion at initial contact than a typical running style[106].

'Groucho running' is a style that was developed in order to examine how leg stiffness affects various aspects of running performance[107]. It is characterised by a large amount of knee flexion in stance phase, which results in reduced vertical stiffness, decreased flight time, increased contact time, increased step length and reduced transmission of shock to the skull[107]. It also requires up to 50% greater oxygen consumption than typical running[107].

The Volodalen® method classifies runners as either '*aerial'* or '*terrestrial'* based on five characteristics[105]. The aerial pattern is a more vertical, bouncing gait and is characterised by pronounced vertical oscillation of the head, arm-swing at the elbow, anterior pelvic tilt and a forefoot strike under the CoM[105]. In contrast, the terrestrial pattern is a more horizontal, gliding pattern characterised by low vertical oscillation of the head, arm-swing from the shoulder, posterior pelvic tilt and a rearfoot strike in front of the CoM[105].

The preferred running pattern of an individual is likely to be influenced by their neuromuscular architecture and function[105]. The aerial pattern is suited to maximising use of the stretch-shortening cycle which reduces metabolic energy requirements and maximises running speed. On the other hand, the terrestrial pattern minimises vertical displacement and thereby external work, which also minimises energy expenditure. Each runner's "optimal technique" will be informed by both their individual anatomy and their running goals, and therefore any training program should also account for these factors.

The validation of Groucho, Pose, aerial and terrestrial styles is preliminary and complicated by confirmation bias. Analysis has tended to focus on a pre-determined subset of biomechanical features. Further work is required using a large population to consider all the biomechanical features of running to determine if, in fact, these styles exist and what distinguishes these styles from one another. As mentioned previously, classifications have been developed for hemiplegic[20] and diplegic[21] walking gait, which have been useful to

facilitate communication between health professionals. To date, there is no classification system for running gaits in people with CP, therefore the potential clinical advantages that flow from the classification of walking are not available for running. Considering the paucity of published literature on the classification of typical running, and the heterogenous nature of the CP population[23, 24], it is expected that any future classification system for CP running gait would best be utilised as a starting point for individual analysis[22].

1.7 Running training

The principle of training specificity dictates that running training strategies will vary depending on whether the goal relates to running skill, capacity or performance[53]. Skill development is important among active individuals as motor competence is a motivating factor that results in increased participation in physical activity into adulthood[108]. In the neurotypical population, amateur runners have become increasingly interested in their running performance, and advancements in wearable technology (e.g. running watches) are now making possible the provision of feedback on running technique even to runners who do not work with a running coach[109]. In the research literature, verbal feedback[63], visual (video) feedback[63] and drills targeting running technique[64, 66, 67, 70, 110] are reported to be effective means of changing spatiotemporal variables[63, 66, 67, 70], kinematics[63, 66, 67] and leg stiffness[64] in novice adult runners[63, 67], sub-elite athletes[64, 66], military personnel with compartment syndrome[70] and 10 year old soccer players[110]. Education and running practice without feedback are reported to be ineffective in changing running technique in children[69].

In children and adolescents with CP, running capacity and agility can be improved by interventions including running[111]. Strength training and plyometric training programs have been shown to improve muscle power and running performance in TD children[112-114], including those who are obese[115]. Plyometric training has been reported to have a greater effect on running performance than traditional strength training[116]. Plyometric training incorporates exercises which require a rapid eccentric muscle action immediately followed by a rapid concentric contraction[117]. This cycle of activity is governed by the use of the stretch-shortening cycle (SSC)[117]. Running requires rapid force production with effective use of the SSC, so plyometric training is well suited to improve running performance[52, 53]. Such programs focus on building running capacity but not running technique/skill.

1.8 Improving physical literacy in children with cerebral palsy

Physical literacy is the competence and confidence to lead an active life[118]. Physical literacy is a concept which includes motor, social, cognitive and psychological domains[118] (Figure 1.4). Running skill fits into the motor domain of the physical literacy framework. According to this framework, as children with CP develop running skill, they also develop their confidence and motivation for physical activity[119]. However, consideration of other domains, such as context, environmental barriers and facilitators, individual knowledge and interest, is important for improvements in running skill in children with CP to be translated into increased physical activity and thereby improved health[35, 120].

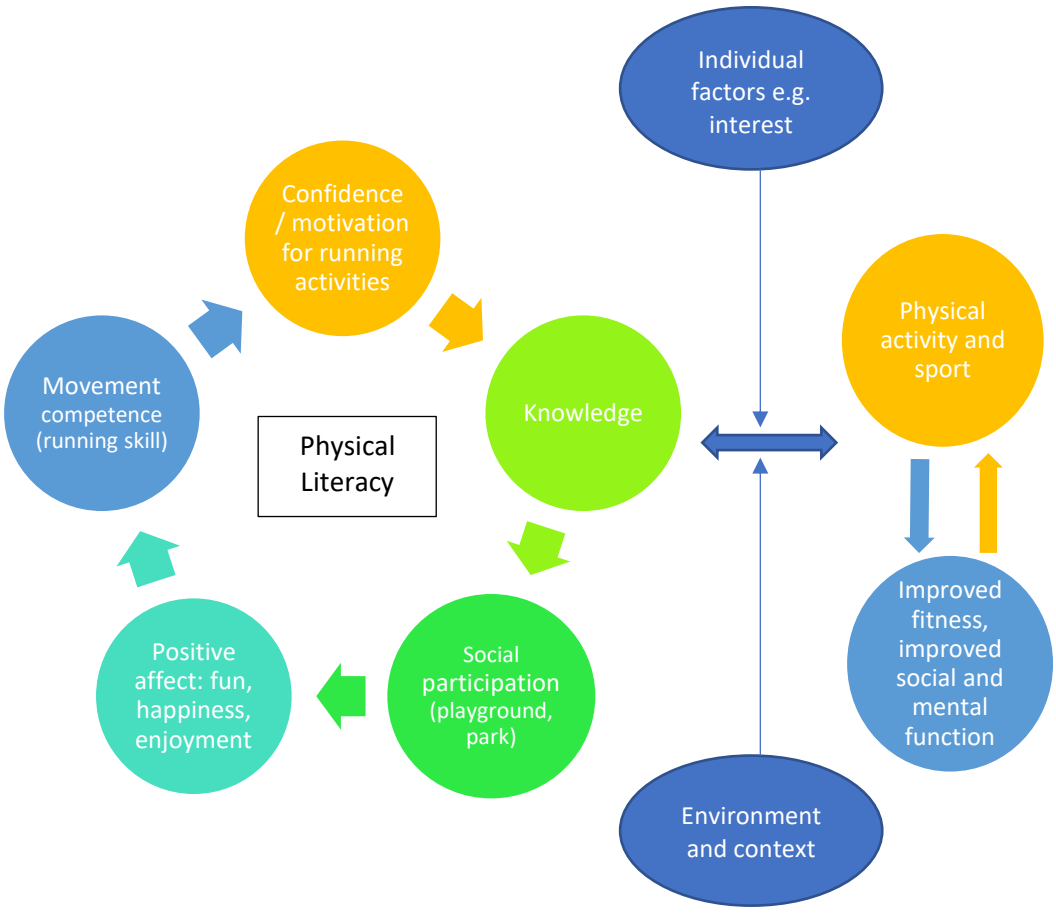


Figure 1.4: Model of physical literacy and physical activity (adapted from Cairney et al., 2019 p375[118])

1.9 Significance and aims of the thesis

Walking gait in people with CP is the focus of a large body of scientific research that attempts to understand the underlying causes of observed gait deviations, and guide management decisions[22], because it is our primary means of locomotion. However, for children and adolescents with CP in GMFCS Level I and II, walking may not be sufficient to maximise their participation in physical activity. These children often attend mainstream schools and have goals related to participation in physical education classes, sports days, playground games and community sport[121]. The ability to run with good technique is a facilitator to participation in physical activity both at school and for recreation, leading to important health and wellbeing benefits.

The fundamental requirement for running is the ability to generate enough power to launch the body into a flight phase, which is achieved primarily by the ankle plantarflexors[122]. Early increases in running speed are likewise achieved primarily through an increase in power generation by the ankle plantarflexors[83]. Therefore, ankle power generation is central to an examination of running skill. The ankle plantarflexors generate power through both muscle contraction and through the mechanism of the stretch shortening cycle, which improves efficiency[82]. Therefore, an examination of ankle power generation during running should include an examination of the function of the stretch shortening cycle.

The aims of this thesis were therefore:

1. To review systematically the existing literature on running in people with CP.
2. To describe power generation during jogging, running and sprinting in children and adolescents with CP and compare it to data from TD children.
3. To estimate the function of the SSC during jogging, running and sprinting in children and adolescents with CP through an examination of leg stiffness and compare it to data from TD children.
4. To investigate the effect of a running training program from adult neurological rehabilitation on the power generation and SSC function in children and adolescents with CP.

The knowledge gained in this body of work is expected to be of value to clinicians, especially physiotherapists, who work with young people with CP towards their high-level mobility goals.

Chapter 2 Running in people with cerebral palsy: a systematic review

2.1 Preface

This chapter describes a systematic review of the literature with two aims: 1) to review what is known about how people with CP run, and 2) to review what is known about the effect of interventions on improving running in people with CP. The research question was deliberately broad, as the body of literature relevant to CP was expected to be small. This systematic review was initially undertaken in 2016 and section 2.3 is reproduced as it appears in print: "Chappell A, Gibson N, Morris S, Williams G, Allison GT. Running in people with cerebral palsy: A systematic review. *Physiotherapy Theory and Practice* 2018; **35**: 15-30." https://doi.org/10.1080/09593985.2018.1434846. An update of the systematic review was undertaken in April 2020 and is presented at the end of the chapter. The new findings are discussed in relation to findings from the 2016 systematic review and includes discussion on how the knowledge pertains to the studies in this thesis.

2.2 Abstract

Background: Running is a fundamental motor skill which is important for participation in recreational activities throughout the lifespan. **Aim:** This review aims to address two questions: 1) how is running in people with cerebral palsy (CP) different to running in people who are neurotypical? and 2) does intervention improve running in people with CP? **Method:** A search of electronic databases was conducted in April 2016 and again in April 2020. Articles were reviewed by two assessors and had to meet the following criteria: 1. Population included people with CP; 2. Included information about running. Articles were assessed for quality using the Checklist for Measuring Study Quality by Downs and Black. **Results:** In 2016, 56 articles underwent full-text review and 17 were included. In 2020, a further 65 articles underwent full-text review and 15 were included. Quantitative analysis was not possible due to diverse study designs and populations. The quality of studies ranged from poor to good. Six of the eighteen descriptive studies reported kinematic and kinetic data. Fourteen intervention studies included running as an outcome measure, although running was not the focus of intervention. A few studies showed that sprint speed, agility and running efficiency are impaired in people with CP, but mechanisms

underlying these impairments have yet to be reported. **Interpretation:** Research on running in people with CP is limited, methodology and findings are inconsistent, and studies are generally fair quality. Further investigation is warranted.

2.3 Systematic review of the literature (2016)

INTRODUCTION

Cerebral palsy (CP) refers to a group of permanent, non-progressive disorders caused by an insult to the developing brain and resulting in disordered development of movement and posture[123]. People with CP experience functional and activity limitations due to these impairments [12, 123] and the level of functional limitation can be classified using the Gross Motor Function Classification System (GMFCS)[12]. Children and adolescents with mild CP who are classified as GMFCS Level I are able to run and jump independently, although many appear to lack speed or coordination. Children in Level II have a limited ability to run and jump although running is not part of the descriptor for 12-18 year olds. For children and adolescents with CP, GMFCS Level I and II, it is important to assess high level motor skills such as running, which are necessary for full participation with peers in physical activities[18, 124, 125]. In clinical practice, children with CP articulate goals related to improving running ability.

Whilst engagement is complex and multi-dimensional, one identified barrier to participation is motor function[126]. People with CP may demonstrate the ability to run in a clinical setting, but not have sufficient running skill to engage in running related community activities such as competitive sport[127]. For example, a child who has a slow running velocity might find it difficult to keep up in a game of chase at recess, and/or have reduced level of enjoyment. Low rates of participation in physical exercise are concerning because if these habits persist they can lead to secondary impairments and chronic health conditions[128]. Improving higher level mobility skills may reduce activity limitations and thereby reduce participation restrictions. Improved participation in physical activities may lead to lower rates of morbidity[129, 130]. Therapy targeting running skill may help children with CP reach their potential within the community.

A large body of research exists for walking in people with CP but very few studies investigate running. Running is a motor skill distinct from walking as the double-support phase is replaced by a flight phase[131]. Running can be subdivided into jogging, running and

sprinting by the speed at which the skill is undertaken[56]. During adolescence and adulthood running is a gateway into participation in many social, leisure and sporting activities and is important for daily functions such as dashing through the rain or catching a departing train[132]. For a person to successfully engage in an activity involving running requires competence in several domains. Children typically employ short bursts of sprint (or maximal) running lasting an average of three seconds during play or sport[40]. The ability to start, stop or change directions quickly are important for participation in games, and a minimum level of efficiency is needed to allow the participant to maintain the required level of intensity throughout the game. These factors may be affected by the primary neurological deficit associated with CP or by the resulting physical impairments.

It is necessary to define terms that will be used in this review as these have sometimes been used interchangeably in the literature. Running ability is the ability to generate a flight phase and therefore meet the requirements for running[131]. Running skill refers to proficiency in the motor skill of running[44] and is related to technique. Running capacity includes both aerobic capacity which is related to running distance[41] and anaerobic capacity which is related to power generation and therefore running velocity[42]. Agility is the ability to rapidly change direction or velocity in response to a stimulus and is influenced by physical factors such as strength and technique, as well as cognitive factors such as visual scanning and anticipation[133]. A person's running ability, skill, capacity and agility all contribute to their running performance in a given task.

This review has two aims: 1) to describe running skill, capacity and agility in people with CP and 2) to describe the impact of interventions on running skill, capacity and agility in people with CP. It is deliberately broad in scope to capture both descriptive and intervention studies.

METHOD

Study Identification and Selection

A systematic review was conducted following the PRISMA guidelines[134] (accessed at www.prisma-statement.org on 22/3/2016). A search was conducted of the electronic databases of Medline (26/4/2016), CINAHL (26/4/2016), Embase (27/4/2016), PEDRO (7/5/2016), Cochrane Database of Systematic Reviews (7/5/2016) and Cochrane Controlled Trials Register (7/5/2016). The search terms used when searching the Medline database are listed in Table 2.1 and these terms were adapted for other databases. The search was

limited to articles written in English and published since 1995. This date limit was chosen since a primary aim of this review was to describe running skill in people with CP and computerised motion analysis was in its infancy in the 1990's[135]. To be included articles needed to: 1. Include people with CP, and 2. Report information about running, that is, gait with a flight phase and no period of double support[56]. Articles were excluded if they were an abstract or general opinion article with no original data but there were no exclusion criteria for study design. The titles and abstracts of identified articles were reviewed by two assessors (AC and NG), who independently shortlisted articles by applying the inclusion and exclusion criteria to the title and abstract of identified articles. Where it was unclear from the title and abstract if an article met the inclusion criteria, the entire article was obtained and reviewed. Differences were resolved by consensus with a third reviewer (SM). Additional articles were then identified through a manual search of the reference lists of articles included in the review. Where data were not presented in a format available for analysis, i.e. data were presented in a figure, an attempt was made to contact the principal author for permission to obtain and use the data. Articles were excluded when it was not possible to extract data about running and authors did not provide data when requested.

Quality Assessment

The included articles were assessed for quality using a modified version of the Checklist for Measuring Study Quality developed by Downs and Black[136]. This checklist contains 27 questions under the categories: quality of reporting (10), external validity (3), internal validity-bias (7), internal validity-confounding (6) and power (1). The original checklist has high internal consistency (KR-20: 0.89), good test-retest and interrater reliability (r=0.88 and r=0.75), good face and criterion validity for both randomised and non-randomised studies and is suitable for use in health care studies. The external validity component has poor reliability (KR-20: 0.54) so it is recommended that care be taken when interpreting this section[136].

The following modifications were made to the checklist: 1. Articles were grouped into "Intervention" or "Descriptive" studies, as there were 10 questions in the checklist that were not applicable to any of the descriptive studies (shown in grey in Table 2.2), lowering the total possible score for the article by 10 points; 2. Total scores were converted to a percentage score resulting in standardised values which allowed comparison between intervention and descriptive studies, and 3. Question 27 (study power related to sample size), was simplified by assigning a score of 1 for adequate power or a score of 0 for low power based on whether the number of participants was sufficient to detect a difference

where one existed according to the stated aim of the study.

Table 2.1: Search terms

1. (cerebral adj1 palsy).mp*
2. diplegi$.mp*
3. quadr$.mp*
4. hemiplegi$.mp*
5. dyskine$.mp*
6. ataxi$.mp*
7. spastic$.mp*
8. 1 or 2 or 3 or 4 or 5 or 6 or 7
9. run$.mp*
10. Running/cl, ed, in, ph [Classification, Education, Injuries, Physiology]
11. 9 or 10
12. gait.mp* or Gait Disorders, Neurologic/ or Gait/
13. 11 or 12
14. 8 and 13
15. limit 14 to english language
16. limit 15 to yr="1995 -Current"
17. (kinetic$ or kinematic$ or intervention or training or propulsion or development).mp*
18. (temporospatial or temperospatial).mp*
19. (reference adj1 value$).mp*
20. 17 or 18 or 19
21. 16 and 20
*mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier

Using the modified checklist, articles were scored by three reviewers (AC, NG and SM). Agreement was reached by consensus. The Downs and Black checklist total score can be used to rate studies as good (total≥20), fair (total 15-19) or poor (≤14)[137]. Quality categories were converted to percentages to allow standardised description of both intervention and descriptive studies, good ≥ 71%, fair 51-70% and poor ≤ 50%.

Data Extraction

Data from the included studies were extracted and summarised using a customised proforma with the headings population, age range, study type, running factor of interest, results in CP group and results in typically developing (TD) group. For each variable of interest, the mean and standard deviation of results in the CP group, and the mean and standard deviation of results in the TD group were extracted.

Data Synthesis

For the purposes of data synthesis, articles were categorised into groups according to the primary aim of each study. A review of included studies revealed three main areas in which running had been reported; biomechanical analysis of running, the development of outcome measures based on running, or intervention with a running outcome measure. Data from included studies were compared and included visual inspection of graphs and figures when data were not reported or able to be obtained from the original authors of studies.

RESULTS

Study Selection

After removal of duplicates the initial search yielded 2603 studies to be considered for inclusion (Figure 2.1). A search of the reference lists of included studies identified a further three potentially relevant studies[138-140]. One further study[42] was identified incidentally by one of the reviewers (AC) bringing the total number of identified studies to 2607. The search terms failed to identify this study as running related terms were not included in the keywords of the article. Following screening of titles and abstracts, 56 studies were obtained for full-text review. After full text review, 28 studies were excluded as they were not about running, eight were excluded as it was not possible to extract data about running and two studies were excluded as they were an annotation or symposium extract and had no original data. Eighteen studies remained[42, 125, 131, 139-153]. Capio et al. (2011) and Capio et al. (2012) were two studies performed on the same group of children with CP using similar outcome measures. The 2011 study investigated inter-rater reliability and comparative validity using GMFCS levels, of process- and product-oriented assessment of fundamental movement skills, while the 2012 study compared the fundamental movement skills of the CP group with a TD group. The 2011 study was included as it reported the data about running and the 2012 study was excluded. At the end of the selection process 17 studies remained to be reviewed.

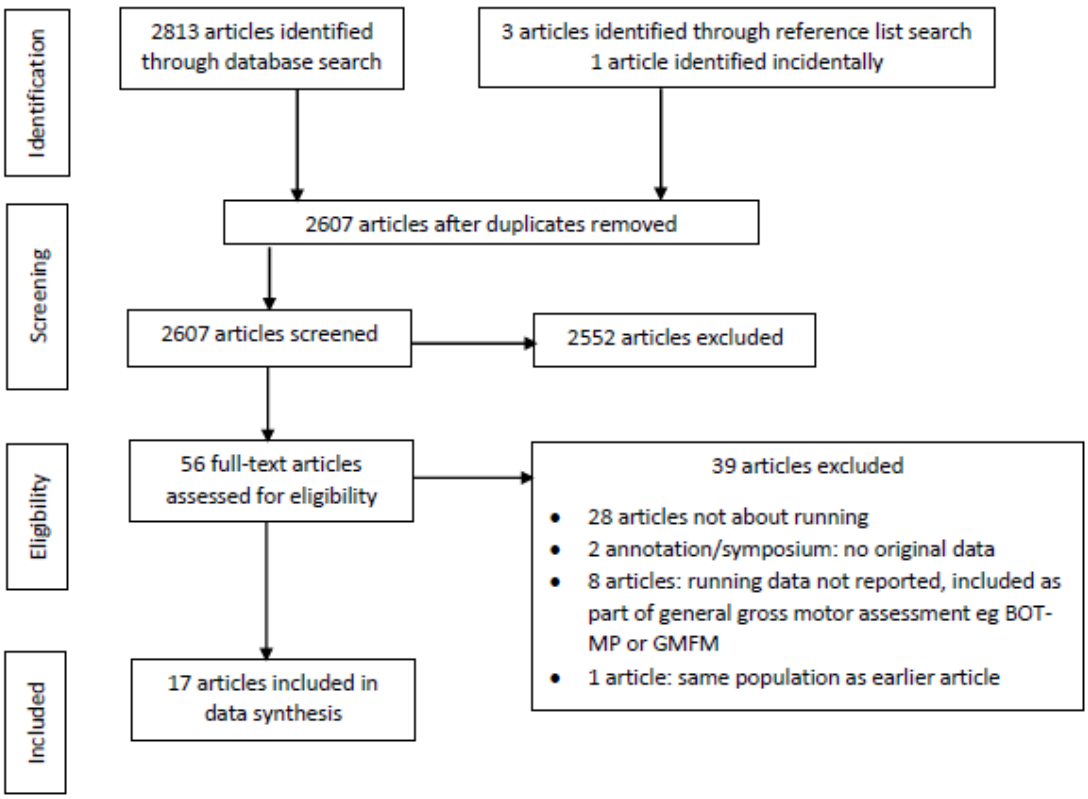


Figure 2.1: PRISMA flow diagram

Study Design

Twelve studies were observational while five studies reported on interventions. The observational studies were cross-sectional; three compared a CP group to a TD group[131, 150, 151]; one compared children with hemiplegia to children with diplegia [125] and the remaining eight studies had no group comparison. The intervention studies included: one randomised controlled trial[141]; one non-randomised experimental pilot study[145]; two pre-test post-test case series[148, 152] and one pre-test post-test case study[147].

Quality Assessment

The results of the Downs and Black Quality Assessment are shown in Table 2.2 and a summary of the sub-section totals is shown in
Table 2.3. The quality of the data was variable, descriptive studies had total scores ranging from 61% to 83%, while the scores for the intervention studies were more variable, ranging from 39% to 79%. In summary, the descriptive studies consistently demonstrated weak internal validity with a high risk of bias. The intervention studies demonstrated a stronger internal validity with a relatively lower risk of bias compared with the descriptive studies.

Table 2.2: Checklist for measuring study quality results

	Reporting										External validity			Internal validity – bias							Internal validity - confounding						P
Qu.	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27
Descriptive studies																											
Bohm & Doderlein 2012[125]	1	1	1	-	0	1	1	-	-	1	1	0	1	-	-	1	-	1	-	1	-	-	-	-	-	-	1
Capio et al 2011[143]	1	1	1	-	2	1	1	-	-	1	0	0	0	-	-	1	-	1	-	1	-	-	-	-	0	-	0
Davids et al 1998[131]	1	1	1	-	2	1	1	-	-	0	0	0	1	-	-	1	-	1	-	1	-	-	-	-	0	-	1
Ferland et al 2011[149]	1	1	1	-	1	1	1	-	-	1	1	1	1	-	-	1	-	1	-	1	-	-	-	-	0	-	1
Hong et al 2012[151]	1	1	1	-	1	1	1	-	-	1	1	0	1	-	-	1	-	1	-	1	-	-	-	-	0	-	1
Iosa et al 2013[150]	1	1	1	-	2	1	1	-	-	1	0	0	1	-	-	1	-	1	-	1	-	-	-	-	-	-	1
Kloyiam, 2011[146]	1	1	1	-	1	1	0	-	-	1	1	1	1	-	-	1	-	0	-	1	-	-	-	-	-	-	1
Ryan et al 2015[153]	1	1	1	-	2	1	1	-	-	1	1	0	1	-	-	1	-	1	-	1	-	-	-	-	-	-	1
Verschuren et al 2010a[142]	1	1	1	-	2	1	1	1	-	0	1	0	1	-	-	1	-	1	-	1	-	-	-	-	-	-	1
Verschuren et al 2010b[42]	1	1	1	-	1	0	1	0	-	0	1	1	1	-	-	1	-	-	-	1	-	-	-	-	1	-	1
Verschuren et al 2009[140]	1	1	1	1	2	1	1	-	-	1	1	0	1	-	-	1	-	1	-	1	-	-	-	-	-	-	1
Verschuren et al 2006[139]	1	1	0	-	2	1	1	0	-	0	0	0	1	-	-	1	-	1	-	1	-	-	-	-	0	-	1
Intervention studies																											
Jelsma et al 2013[152]	1	1	1	1	0	1	0	0	1	1	1	0	1	-	1	1	1	1	1	1	1	1	0	0	0	-	0
Capio et al 2015[145]	1	1	1	1	1	1	1	0	0	1	0	0	1	1	1	1	1	1	1	1	1	1	0	0	0	0	1
Kenyon et al 2010[147]	1	1	1	1	-	1	1	1	1	0	0	0	1	-	0	1	-	-	1	1	-	-	-	-	-	-	0
Verschuren et al 2007[141]	1	1	1	1	1	1	1	1	1	1	1	0	1	-	1	1	1	1	1	1	1	1	1	0	0	0	1
Johnson et al 2014[148]	1	1	1	1	-	1	0	1	1	0	1	0	1	-	0	1	-	-	0	1	-	-	-	-	-	-	0

-Not Applicable; Qu.=question; P=power

Table 2.3: Checklist for Measuring Study Quality Results – Section Totals

Section:		Reporting	External Validity	Internal Validity – Bias	Internal Validity – Confounding	Power	Overall Total	Downs and Black rating
Maximum possible score	**Number of subjects**	**11**	**3**	**7**	**6**	**1**	**/18 (Descriptive) /28 (Intervention)**	
Descriptive studies								
Bohm & Doderlein 2012[125]	49	6	2	3	N/A	1	12 (67%)	Fair
Capio et al 2011[143]	30	8	0	3	0	0	11 (61%)	Fair
Davids et al 1998[131]	19	7	1	3	0	1	12 (67%)	Fair
Ferland et al 2011[149]	50	7	3	3	0	1	14 (78%)	Good
Hong et al 2012[151]	33	7	2	3	0	1	13 (72%)	Good
Iosa et al 2013[150]	40	8	1	3	N/A	1	13 (72%)	Good
Kloyiam, 2011[146]	11	6	3	2	N/A	1	12 (67%)	Fair
Ryan et al 2015[153]	55	8	2	3	N/A	1	14 (78%)	Good
Verschuren et al 2010a[142]	306	8	2	3	N/A	1	14 (78%)	Good
Verschuren et al 2010b[42]	300	5	3	2	1	1	12 (67%)	Fair
Verschuren et al 2009[140]	68	9	2	3	N/A	1	15 (83%)	Good
Verschuren et al 2006[139]	25	6	1	3	0	1	11 (61%)	Fair
Intervention Studies								
Jelsma et al 2013[152]	14	7	2	6	2	0	17 (61%)	Fair
Capio et al 2015[145]	50	8	1	7	2	1	19 (68%)	Fair
Kenyon et al 2010[147]	1	8	1	3	0	0	12 (43%)	Poor
Verschuren et al 2007[141]	65	10	2	6	3	1	22 (79%)	Good
Johnson et al 2014[148]	3	7	2	2	0	0	11 (39%)	Poor

Data Synthesis

Although 17 studies were identified for inclusion, there were few common elements among studies and the data were unsuitable for meta-analysis. One study in the review by Davids, Bagley and Bryan (1998) described running in children with spastic diplegia compared to TD children, while two studies[125, 150] focussed on the symmetry of running in children with CP. In the remaining 13 studies data related to running were reported as an outcome measure or an intervention for cardiovascular fitness, rather than describing running impairment.

Running Skill: Biomechanical studies

Participants

In the biomechanical studies the topographic distribution of CP of the participants were described as hemiplegia[150], diplegia[131], separately reported as hemiplegia, symmetric diplegia or asymmetric diplegia groups[125] or were a mixed group[146]. One study focussed on elite adult soccer athletes with CP[146] while three studies included children with CP but not adults[125, 131, 150] (Tables 2.4, 2.5 and 2.6).

Biomechanical outcomes

Biomechanical data was extracted where possible with spatiotemporal data reported in four studies (Table 2.4), kinematics from three studies (Table 2.5) and kinetics from two studies (Table 2.6). Three of the four studies reporting on spatio-temporal, kinematic or kinetic parameters had a fair quality grading[125, 131, 146] and one had a good rating[150]. The varied data collection methods, populations and reporting of biomechanical data precluded data pooling for an integrated analysis. Two studies reported that children with CP ran slower[131, 150] and had shorter stride lengths[131, 150] compared with peers who are TD. When transitioning from walking to running children with diplegia increased their cadence in contrast to peers who are TD, who increased step length as a strategy for increasing speed[131, 150].

Table 2.4: Spatio-Temporal Characteristics of Running

Paper:	**Bohm and Doderlien[125] Barefoot running**		**Davids et al.[131] Barefoot running**		**Iosa et al.[150] Shod running**		**Kloyiam[146] Shod running on treadmill at 12km/hr**
Spatio-temporal parameters	CP-hemiplegia Age: x=12 yrs SD=4; n=25	CP-diplegia Age: x=12.5 yrs SD=3.7 symmetric diplegia n=14 asymmetric diplegia n=10	CP-diplegia Age range: 4.2-12.4 yrs; n=19	TD Age range: 7.2-12.1 yrs; n=15	CP-hemiplegia Age: x=5.1 yrs SD=2.3; n=20	TD Age: x=5.9 yrs SD=2.6; n=20	Irish CP national soccer team Age range: 18-40 yrs; n=9
Velocity (m/s)	2.3m/s	2.3m/s	2.59 m/s* (SD=0.51)	3.72m/s (SD=0.56)	A little less than 2m/s*	Approx. 2.5m/s	
Cadence (steps/min)			238 steps/min (SD=34)	232 steps/min SD=34			
Stride length (m)			1.32m* (SD=0.27)	1.94m (SD=0.28)	Approx 1.2m*	Approx 1.6m	Average = 2.33m
Step time (s)	More symmetrical than walking	More symmetrical than walking	0.23s	0.22s	Approx 0.25s More symmetrical than walking	Approx 0.25s	
Stance %	More symmetrical than walking		44.4 (SD=6.7)	41.7 (SD=4.4)			
Double float %			6.9 (SD=4.0)	7.9 (SD=3.2)			
Gait symmetry					Less symmetrical than TD*		
Gait harmony (rhythmicity)					AP harmony no different to TD in running, although different in walking		

Gait smoothness as root mean square of acceleration					Running less smooth than walking	Running less smooth than walking	

*Significantly different to TD CP=cerebral palsy; TD=typically developing; yrs=years; n=number of participants; m/s=metres per second; Approx.=approximately; m=metres; min=minutes; steps/min=steps per minute; s=seconds; %=percentage

Table 2.5: Kinematics of Running

Paper:	**Bohm and Doderlien[125] Barefoot running**		**Davids et al.[131] Barefoot running**		**Kloyiam[146] Shod running on treadmill at 12km/hr**
Joint Kinematics Peak values	CP-hemiplegia Age: x=12 yrs SD=4 n=25	CP-diplegia Age: x=12.5 yrs SD=3.7 asymmetric diplegia n=10 symmetric diplegia n=14	CP-diplegia Age: 4.2-12.4 yrs n=19	TD Age: 7.2-12.1 yrs n=15	Irish CP national soccer team Age:18-40 yrs n=9
Ankle					
ROM (°)			34^{o} (SD=13^{o})	39^{o} (SD=7^{o})	
Initial contact	35% bilateral forefoot 64% unilateral forefoot	79% bilateral forefoot 21% unilateral forefoot			In plantarflexion bilaterally = 2/9 (22%) In plantarflexion unilaterally = 7/9 (78%)
Peak Plantarflexion (°)	Increased compared to TD	Slightly decreased compared to TD	24^{o} PF (SD=18^{o})	18^{o}PF (SD=11^{o})	
Peak Dorsiflexion (°)	Less than TD, less symmetrical than walking	Symmetric diplegic same as TD, asymmetric slightly less. Less symmetrical than walking.	11^{o}* (SD=12^{o})	20^{o} (SD=6^{o})	
Knee					
ROM (°)			43^{o}* (SD=8^{o})	73^{o} (SD=11^{o})	
Flexion at IC (°)	Slightly more than TD	Asymmetric same as TD, symmetric slightly less than TD	34^{o} (SD=8^{o})	28^{o} (SD=12^{o})	

Peak Flexion (°)	Slightly increased compared to TD	Slightly decreased compared to TD. More symmetrical than walking.	66°* (SD=8°)	90° (SD=12°)	
Peak flexion timing as % Gait Cycle	Similar to TD	Similar to TD	68% SD=4	67% SD=3	
Peak Extension (°)	Approx. 25°, similar to TD. More symmetrical than walking.	Approx. 25°, similar to TD.	23°* (SD=7°)	18° (SD=8°)	
Hip					
ROM (°)			50°* (SD=10°)	63° (SD=9°)	
Peak flexion (°)	Similar to TD. More symmetrical than walking	Similar to TD.	56° (SD=9°)	57° (SD=10°)	
Peak extension (°)	Less extension than TD. Less symmetrical than walking	Asymmetric less extension than TD, symmetric similar to TD. Less symmetrical than walking	6° flexion* (SD=7°)	6° extension (SD=5°)	
Pelvis					
Peak forward tilt	Significant differences to TD. Less symmetrical than walking	Significant differences to TD. Less symmetrical than walking			
Peak backward tilt	Significant differences to TD. Less symmetrical than walking	Significant differences to TD. Less symmetrical than walking			

*Significantly different to TD

X=mean; SD=standard deviation; yrs=years; n=number of participants; CP=cerebral palsy; TD=typically developing; ROM=range of motion; Approx.=approximately

Two studies identified that 99-100% of participants with CP were forefoot strikers on at least one side[125, 146]. Bohm and Doderlein (2012) reported that 25 children with hemiplegia were more plantarflexed throughout the gait cycle, more flexed at the knee in initial swing, terminal swing and initial contact, but less flexed at the knee during stance compared to their peers who are TD. The hip was also more flexed in swing but more extended at initial contact and during stance, compared to peers who were TD[125].

Davids, Bagley and Bryan (1998) reported that 19 children with diplegia were more plantarflexed throughout the gait cycle. Bohm and Doderlein (2012) divided their cohort with diplegia into symmetrical (n=14) and asymmetrical (n=10) groups, and reported that children with asymmetric diplegia demonstrated greater plantarflexion throughout the gait cycle except in initial swing, while children with symmetric diplegia were not significantly different to peers who are TD. Davids, Bagley and Bryan (1998) reported that children with diplegia were stiffer at the knee with less peak flexion in swing and less peak extension in stance compared to peers who are TD, and the hip was more flexed at terminal stance and initial swing. Bohm and Doderlein (2012) reported similar findings in children with asymmetric diplegia but found that children with symmetric diplegia were slightly more flexed at the knee and slightly more extended at the hip throughout the gait cycle compared to peers who are TD.

Only Bohm and Doderlein (2012) reported kinematics of the pelvis during running in children with CP. Children with hemiplegia had increased anterior tilt from mid-stance to mid-swing and reduced anterior tilt from mid-swing to initial contact. Children with asymmetric diplegia demonstrated more anterior tilt throughout the gait cycle compared to children with hemiplegia. Pelvic motion in children with symmetric diplegia was similar but with reduced total range of motion compared with the other 2 groups.

Kinetics were only reported in two studies[125, 131]. Davids, Bagley and Bryan (1998) compared children with diplegia to a TD group and found reduced power generation at the ankle and knee in the CP group, while power absorption at these joints was not significantly different. At the hip, however, power generation was not significantly different to the TD group, while power absorption was significantly reduced. The peak hip flexion moment was significantly less in the CP group while the peak hip extension moment was not significantly different to the TD group. Bohm et al (2012) reported that children with both hemiplegia and diplegia have less symmetrical power generation and absorption at the ankle, less symmetrical power generation at the knee and less symmetrical power absorption at the hip than during walking.

Bohm and Doderlein (2012) investigated the symmetry of walking and running in children with hemiplegia and diplegia using three-dimensional gait analysis (3DGA) and reported that asymmetries became more apparent in running compared with walking in both hemiplegia and diplegia. Asymmetry was defined as the absolute difference between left and right sides in each parameter measured, and 13 out of 22 parameters showed an increase in asymmetry, only step time was significantly more symmetrical in running than in walking. Iosa et al (2013), who used an inertial sensor device and a percentage ratio to define symmetry, also reported more symmetrical step time in running than in walking, but reported that both walking and running were significantly more asymmetrical in the hemiplegia group compared with the TD group.

Iosa et al (2013) investigated gait harmony, which reflects rhythmicity of gait and running smoothness, defined as the root mean square (RMS) of accelerations of the centre of mass (COM). Gait harmony was significantly reduced in the CP group compared to the TD group in both walking and running, but gait smoothness was not significantly different. Raw data were not published but represented graphically, an estimation of values is included in Tables 2.4, 2.5 and 2.6.

Running Capacity and Agility: Field measures of running

Four studies in this review reported on standardised assessments which used running to assess aerobic fitness, anaerobic power and agility. These assessments have been developed and standardised for children and adolescents with CP[42, 139, 140, 142]. The studies were all rated for quality as fair or good. Two of the papers reported the development of a modified shuttle run test (SRT) to test aerobic fitness specifically for GMFCS Level I, II and III with each group having unique parameters and reference curves[139, 142]. Performance on the SRT is positively correlated with height and males perform better than females[142]. Shuttle run test results were not significantly correlated to performance on the Gross Motor Function Measure (GMFM)[154] Part D or E[140], but were significantly related to hip abductor isometric muscle strength[149] and time spent in vigorous physical activity[153]. Shuttle run test results also have significant negative association with body mass index and systolic blood pressure[153], although the direction of causality has not been reported.

Reference curves have also been developed for children with CP GMFCS levels I and II for the muscle power sprint test (MPST, anaerobic power) and the 10x5m sprint test (agility)[42]. Performance was better on both these tests in males than in females and in GMFCS level I compared to level II. Mean power on the MPST increased in curvilinear fashion with

height[42] while there was a negative linear relationship between 10x5m sprint time and height. Both tests were significantly correlated with the GMFM Part D and E[140], indicating that sprint ability and agility are important factors for high level mobility.

Capio, Sit & Abernethy (2011) reported that both running process and running product-oriented scores on the Test of Gross Motor Development – 2nd edition (TGMII)[90] were significantly correlated with GMFCS level. Hong et al. (2012) reported that the Running Speed and Agility (RSA) scale of the Bruininiks-Oseretsky Test of Motor Proficiency (BOTMP)[155] was significantly correlated with peak torque of knee flexors at all velocities, peak torque of knee extensors at highest velocity (120^{o}/s) and curl up scores (trunk muscular endurance) in children with CP, GMFCS Levels I and II, indicating that high velocity force development and trunk stability are important for running skill and agility.

Kloyiam (2011) analysed the running of the Irish CP soccer team using 3DGA on a treadmill set at 12 km/hr. This study aimed to identify differences in running endurance and economy of elite soccer players with CP compared to elite soccer players without CP. Distances on the Yo-Yo IRL1 assessment of soccer specific endurance[156] were significantly less in the athletes with CP compared to athletes without CP. Nine players with CP were assessed for running economy with variable scores, sometimes similar or better than athletes without CP, which did not enable any relationship or associations to be established.

Intervention Studies

There were five studies[141, 145, 147, 148, 152] which reported an intervention with at least one outcome measure related to running. Two of the studies were rated poor for study quality[147, 148], two were rated fair[145, 152] and one was rated good[141]. While four of the interventions included running activities[141, 143, 147, 148], none of these activities reported training the motor skill of running, or running gait education. These studies used various assessments as pre- and post-intervention measures and different intervention protocols, as summarised in Table 2.7. Interventions were reported to effect improvements in running speed over 15m [145], aerobic fitness[141, 147], anaerobic power[141, 147] and agility[141, 147]. Gross motor function measure and BOTMP-2 scores did not change with improvements in running[141, 147]. One study[152] did not observe a significant improvement in a running related outcome (running speed and agility sub-test of the BOTMP-2). In this study the participants undertook a WiiFit intervention which did not include any running activities.

The assessments used in these intervention studies reflected changes in running capacity or

agility but gave no information about spatio-temporal, kinematic or kinetic changes in running. The effect of intervention on the motor control strategy or skill of running, the strategy chosen to increase velocity, symmetry or rhythmicity of movement was not reported.

Table 2.6: Kinetics of Running

Paper:	Bohm and Doderlien[125]		Davids et al.[131]	
Joint kinetics: peak values	**CP-hemiplegia Age: x=12 yrs SD 4 n=25**	**CP-diplegia Age: x=12.5 yrs SD 3.7 asymmetric diplegia n=10 symmetric diplegia n=14**	**CP-diplegia Age range: 4.2-12.4 yrs n=19**	**TD Age range: 7.2-12.1 yrs n=15**
Ankle				
Peak plantarflexion moment (Nm)			1.61Nm* (SD=0.39)	1.98Nm (SD=0.37)
Peak dorsiflexion moment (Nm)			-0.06Nm (SD=0.07)	-0.08Nm (SD=0.04)
Peak power generation (Nm/kg)	Approx. 3 Nm/kg Less symmetrical than walking	Approx. 4 Nm/kg Less symmetrical than walking	4.77* (SD=1.89)	11.23 (SD=2.66)
Peak power absorption (Nm/kg)	Approx. -4 Nm/kg Less symmetrical than walking	Asymmetric approx. -3.5Nm/kg; symmetric approx. -5Nm/kg. Less symmetrical than walking	-3.10 (SD=1.08)	-4.28 (SD=2.22)
Knee				
Peak extension moment (Nm)			1.49 (SD=0.49)	-1.81 (SD=0.59)
Peak flexion moment (Nm)			-0.52 (SD=0.14)	1.09 (SD=1.13)
Peak power generation (W/kg)	Approx 1.5 Nm/kg. Less symmetrical than walking	Approx. 2 Nm/kg. Less symmetrical than walking	3.59* (SD=2.58)	6.91 (SD=3.74)
Peak power absorption (W/kg)	Approx. -1.5 Nm/kg	Asymmetric Approx. -3Nm/kg, symmetric approx. -1 Nm/kg	-2.66 (SD=1.12)	-8.51 (SD=10.84)
Hip				
Peak extension moment (Nm)			1.13 (SD=0.30)	2.13 (SD=2.01)
Peak flexion moment (Nm)			-1.47* (SD=0.57)	-2.45 SD=0.96
Peak power generation (W/kg)	Approx. 4 Nm/kg	Asymmetric approx. 5 Nm/kg, symmetric approx. 4Nm/kg	3.96 (SD=1.61)	7.65 (SD=6.93)
Peak power absorption (W/kg)	Approx. -1 Nm/kg Less symmetrical than walking	Approx. -1 Nm/kg Less symmetrical than walking	-6.94* (SD=3.97)	-16.31 (SD=7.89)

*Significantly different to TD; X=mean; SD=standard deviation; yrs=years; n=number of participants; Nm=newton metres; Nm/kg=newton metres per kilogram; Approx.=approximately; W=watts

Table 2.7: Summary of intervention studies

Author	Downs and Black rating	Population	Frequency and Duration of Intervention	Description of Intervention	Outcome
Capio et al.[145]	Fair	Children with CP GMFCS Level I (n=6), II (n=14) and III (n=4); TD children n=26	Once a week for 45 minutes for 4 weeks	Skill specific practice including running for 5 minutes in an open space without obstacles. No further instruction given	Significant effect of training on running speed over 15m (no group effect). Both CP and TD training groups spent significantly more time in moderate-vigorous activity on weekend days (not weekdays)
Jelsma et al.[152]	Fair	N=14 CP spastic hemiplegia GMFCS I and II; Age 7-14	Four times per week for 25 minutes for 3 weeks	WiiFit Games on balance board	No significant difference in Running Speed and Agility (RSA) subtest of BOT-2
Johnson et al.[148]	Poor	n=3; Age=8 yrs 9m - 10 yrs; R hemiplegia GMFCS Level I	Twice weekly for 35-45 minutes, duration 8-10 weeks	Plyometric training, 3 sets of 5 repetitions of: - 4 jumping or hopping exercises - 4 throwing exercises	Change in agility (10x5m sprint) less than MCID. Two participants improved running speed, not maintained after 6 weeks
Kenyon etl al.[147]	Poor	n=1; age=16 yrs; spastic diplegia GMFCS Level I	Twice weekly for 10 weeks	Interval training with focus on wrestling, building to 2 minutes per station: - Repetitive jumps - Repetitive 2 handed ball toss - Side shuffle - Supine-prone-standing transitions - High knee jogging - Sit ups - Squat thrusts - Push ups	Improvement greater than SEM in mean power (MPST change=318.81W) and agility (10x5m sprint change=9.32sec). Improvement greater than MCID in aerobic fitness (SRT-I change=3.5 levels). BOT-2 and GMFM-66 scores did not improve more than SEM
Verschuren et al.[141]	Good	n=86 CP GMFCS I and II; Age 7-18	Twice weekly for 45 minutes for 8 months	Functionally based exercise program:	Improvement in aerobic capacity (SRT $p<.001$), X=2.4min SD=1.9min;

				- 8 aerobic exercises of 3-6 minutes - 8 anaerobic exercises of 20-30 sec - Functional activities (running, changing directions abruptly, step-ups, stairs)	Improvement in mean anaerobic capacity (MPST p=.004) x=20.4W SD=38.0W; Improvement in agility (10x5m sprint p<.001) x = -4.5sec SD=4.1sec GMFM Dimension E (walking, running and jumping) did not improve significantly

Abbreviations: CP=cerebral palsy; TD=typically developing; BOT-2=Bruiniks-Oteretski Test of Motor Proficiency 2nd edition; MCID=minimum clinically important difference; SEM=standard error of measurement; W=watts; SRT=Shuttle Run Test; GMFM-66=Gross Motor Function Measure – 66 items; MPST=muscle power sprint test; x=mean; SD=standard deviation.

DISCUSSION

The primary finding of this review is that studies describing running gait in people with CP are rare and there are insufficient data to adequately describe the impairments associated with running gait in CP, particularly in the adult population. Of the four biomechanical studies, three focussed on children with only one article describing a small number of biomechanical parameters of running gait in elite adults with CP. There were no studies of running for non-elite adults with CP. It may that running is under-investigated as the majority of interventions such as surgery or botulinum toxin injection have the goal of improving walking gait[157] and therefore clinical research has focussed on walking gait as the functional outcome.

Three broad categories of assessments have been used to describe running in people with CP. Running ability and skill have been assessed using motion analysis[125, 131, 146, 150], running capacity and agility have been assessed using field measures[42, 139, 140, 142] and running performance has been assessed as part of more generalised tests of motor development[143, 145, 147, 152]. Classifications of walking gait exist for both hemiplegic-type and diplegic-type CP[20, 21] but this review did not identify any methods for classifying running gait.

The biomechanical studies reporting on running skill in this review employed different methodologies, making comparison of studies and generalisation of findings difficult. Two of the studies that included 3DGA of running in children with diplegia reported different kinematic findings. One study separated symmetric and asymmetric diplegia[125] while the other did not[131]. The differences in findings suggest that it is important to operationally define symmetric and asymmetric diplegia for the purposes of biomechanical analysis of movement.

The second main finding of this review is that running capacity, agility and running efficiency, which are important for engagement in running activities, are all affected by the impairments associated with CP (see Table 2.8). However, the mechanisms underlying these relationships have not yet been reported in the literature. Studies of field measures of running capacity and agility report that boys with CP perform better on the SRT (aerobic fitness), MPST (anaerobic power) and 10x5m sprint (agility) than girls with CP[42, 142], which has also been reported in children who are TD[158, 159]. Both anaerobic power and aerobic fitness reported in children with CP are lower than that reported in children who are TD[41, 158] and lower in GMFCS Level II than in Level I[42]. Children and adolescents with CP achieve

maximum heart rates at slower speeds than their peers who are TD, which is why a modified SRT has been developed to test aerobic fitness in this population[139].

Table 2.8: Summary of factors affecting successful engagement in running activities in children with CP

Running skill	Running capacity		Agility
	Aerobic	Anaerobic	
Spatiotemporal Characteristics • Velocity is decreased in children with CP compared to children who are TD [131, 150] • Stride length is decreased in children with CP compared to children who are TD [131, 150] • Running is less symmetrical in children with CP than children who are TD [150] • Step time is more symmetrical than in walking [125, 150] • Running is less smooth than walking [150]	Shuttle Run Test • Aerobic capacity is higher in individuals in GMFCS Level I than Level II [41] • Aerobic capacity is higher in males than females [41] • Aerobic capacity increases with height [41]	Muscle Power Sprint Test • Anaerobic power is higher in individuals with GMFCS Level I than Level II [42] • Anaerobic power is higher in males than females [42] • Anaerobic power increases with height [42]	10 x 5m Test • Individuals who are GMFCS Level I have better agility than those with GMFCS Level II [42] • Agility is better in males than females [42] • Agility decreases with height [42]
Kinematics • Barefoot running is more asymmetrical than walking [125] • Peak dorsiflexion is significantly reduced in children with CP compared to children who are TD [131] • Peak knee flexion and peak knee extension are both significantly reduced in children with CP compared to children who are TD [131] • Peak hip extension is significantly reduced in children with CP compared to children who are TD [131]			
Kinetics • Ankle and knee power generation is significantly reduced in children with CP compared to children who are TD [131] • Hip power absorption is significantly reduced in children with CP compared to children who are TD [131]			

• Power generation and absorption is more asymmetrical than in walking [125]			

GMFCS=Gross Motor Function Classification Scale; CP = Cerebral Palsy; TD = Typically Developing

In children who are TD, running gait is achieved utilising a different motor control strategy to walking[93, 122, 160]. Running requires the generation of a flight phase involving greater vertical displacement of the centre of mass than in walking[93]. The ability to generate a flight phase is influenced by physical maturation and the development of motor skills, power and endurance[93, 101, 161]. The few biomechanical studies identified in this review[125, 131, 146, 150] report that people with CP run more slowly than their peers who are TD, with a shorter stride length. This may be due to compromised distal power generation and a compensatory proximal power generation strategy[125, 131] but the interaction between power generation and strategies to achieve propulsion in the CP population requires further exploration.

Leg stiffness and the SSC function are variables which have been linked to sprint running performance and have been shown to change with maturation[95, 101, 161]. Leg stiffness during loading response and mid-stance of walking has been shown to be reduced in children with CP compared to children who are TD[162], but it is unknown if it is also reduced during running in people with CP. It is possible that the presence of lower limb spasticity and hypertonicity lead to greater leg stiffness and impact on the stretch shortening cycle during running as it is a higher velocity movement than walking. The interactions of these positive and negative features of the upper motor neurone syndrome and their impact on running skill, capacity and agility have not been reported but may be important for developing effective running training programs for people with CP.

In the TD population rear-foot striking is more common, although its prevalence decreases in barefoot runners compared to shod runners[163, 164]. In pre-adolescent children who are TD, 45% are forefoot strikers when barefoot, while only 24% are forefoot strikers when shod[165]. This systematic review shows that people with CP have a higher than usual incidence of forefoot striking independent of barefoot or shod condition[125, 146]. The relationship between spasticity and foot-strike pattern has not been reported, but is an important question as a forefoot strike pattern has been postulated to reduce injury risk in the TD population[166]. More research is needed on the biomechanics of CP running gait to better understand impairments which commonly have a negative or perhaps positive

impact on running skill.

A previous study has suggested that bilateral motor impairment is one factor which increases the risk of decline in walking gait function in adults with CP[167]. This review has shown that characteristics of running in people with unilateral or asymmetrical motor impairment are different to those with symmetrical motor impairment, but the relationship between these characteristics and running skill, capacity and agility is unknown.

Running is sometimes assessed as an item of a more generalised test of motor development. Studies in this review which included running as part of a generalised motor assessment such as the GMFM[141, 147] or the BOTMP-2[147] reported significant changes in specific measures of running performance without significant changes in the generalised motor assessment score, despite anaerobic power and agility being significantly correlated to the GMFM part D and E[141]. The authors therefore recommend that clinicians interested in assessing running choose measures which assess pertinent aspects of running capacity specifically, such as the modified shuttle for aerobic capacity or the 10x5m sprint test for agility, rather than a more generalised scale of motor development or function.

The third main finding of this review was that few studies have used an intervention to improve running ability, and those that have did not report spatio-temporal, kinematic or kinetic parameters. The intervention studies in this review indicate that interventions that include running can change running capacity or agility[141, 145, 147, 148]. None of the interventions specifically trained the motor skill or motor pattern of running. One study identified muscle power generation as a target area, but this was reported as anaerobic power using the MPST while kinetic data during running were not reported[147]. None of the studies investigated the mechanisms of improvement in running capacity or agility, so it is not known whether the children with CP learned to use abnormal strategies more effectively, or normalised the kinematics and kinetics of running. This could be a question for future research. It is not known whether intervention changes the motor control strategy or skill of running, symmetry, the strategy chosen to increase velocity or rhythmicity of movement. It is also unknown how factors such as dosage (frequency, duration), tasks included in the interventions, baseline performance and maturity all interact to affect outcomes. While running capacity and agility are trainable in this population, the heterogeneity of the outcome variables and the absence of kinematic and kinetic data means that the underlying mechanisms that led to these improvements are largely unknown. It would be helpful for future intervention studies to focus on specific

variables related to running skill, including biomechanics and physiology.

Limitations of this review are the small and disparate number of studies identified in the literature and range of study quality. In order to keep the review broad in scope we chose to include single case studies and studies utilising small numbers. Two of the five intervention studies had sample sizes of n=1 and n=3[147, 148] with scores of 0 for internal validity on the Downs and Black Assessment[136]. The scope of this review was limited to English language peer-reviewed articles. It is possible that further information about running gait in people with CP could be found in the grey literature or published in other languages.

CONCLUSIONS

There is yet to be a comprehensive analysis of running gait in people with CP and running is under-reported in the literature compared to walking. There were no studies identified in this review which sought to report the gait deviations associated with adult CP running gait. Running gaits differ between people with hemiplegia, asymmetric diplegia and symmetric diplegia and further research is required to better define and describe these groups. In addition, no studies utilised GMFCS levels to determine if functional level affects running skill. Running in CP is an area which deserves further attention, as the ability to run is likely to contribute positively to physical activity levels, general health and participation.

2.4 Systematic review update (2020)

Methods

The methodology employed for the systematic review update was the same as that employed for the original systematic review described earlier in this chapter, with two exceptions: only studies published between 2015 and April 2020 were included; and any articles included previously, as well as any publications resulting from this thesis, were excluded from the new analysis. The research questions were the same, the same search terms were used, the same databases were searched, and the same study selection process followed. Titles and abstracts were again screened by two reviewers (AC and NG) and by a third reviewer (SM) where there was no consensus. The same analysis of study quality was undertaken using the Checklist for Measuring Study Quality developed by Downs and Black[136]. Once again, studies were considered as either descriptive or intervention studies.

Results

Study selection

The study flow is presented in Figure 2.2. After screening and exclusion, 17 articles remained for data synthesis, however two pairs of articles reported different outcomes for the same group of participants:

Runciman et al (2016)[168] and Runciman et al (2016)[169]

Bohm et al (2018)[15] and Kratschmer et al (2019)[170]

For the purposes of data extraction, each pair was analysed as a single publication (15 studies in total).

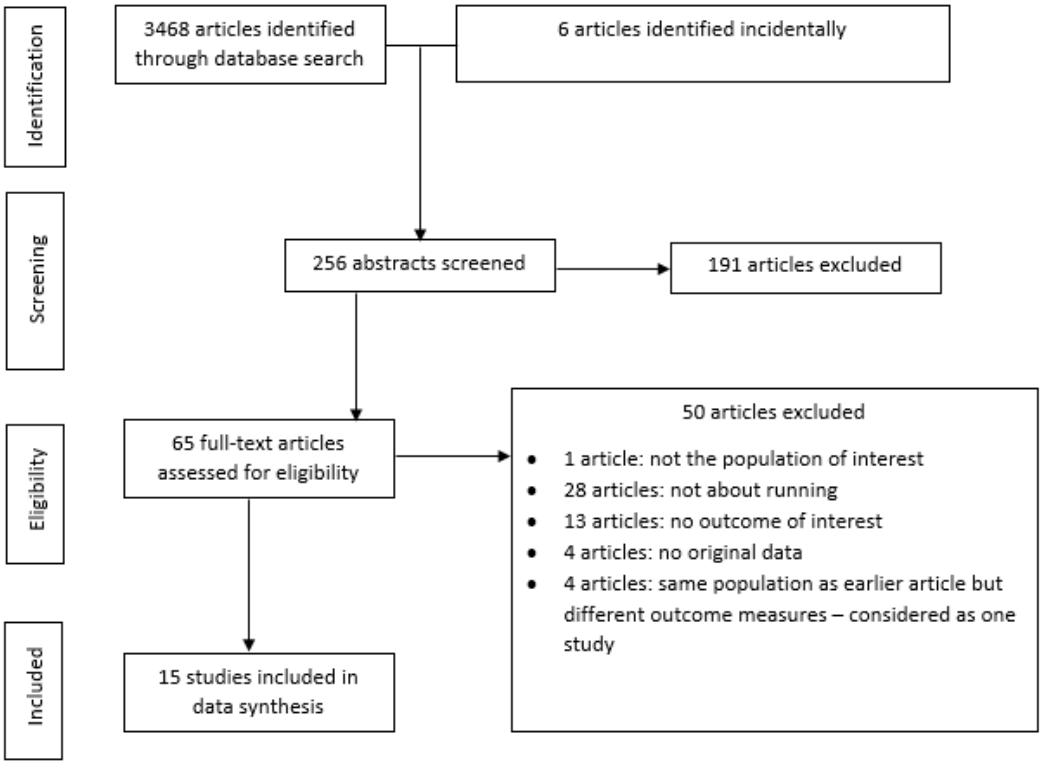


Figure 2.2: Systematic review update PRISMA flow diagram

Study Design

Six studies were descriptive while nine studies reported on interventions. The descriptive studies were cross-sectional; three compared an adult Paralympic CP group to neurotypical athletes[168, 169, 171, 172]; one compared four adult Paralympic athletes from different classes[173]; one described a large cohort of German children with CP, GMFCS Level II[15, 170]; and one compared matched samples of Dutch children with CP in 2004 and 2014[174]. The intervention studies included: four randomised controlled trials (RCTs)[175-178]; a randomised

prospective controlled pilot study comparing progressive resistance training and high intensity interval training[179]; a double-baseline study[180]; two case studies of adolescents[181, 182] and a case study of a Paralympic sprinter from Brazil[183]. The interventions are described in Table 2.14.

Quality Assessment

The results of the Downs and Black Quality Assessment are shown in Table 2.9 and a summary of the sub-section totals is shown in

Table 2.10. As in the previous review, the quality of the data was variable. Descriptive studies had total scores ranging from 39% to 67%, while the scores for the intervention studies ranged from 29% to 86%. With two exceptions[172, 174], the descriptive studies demonstrated weak external validity and all descriptive studies had high risk of bias and confounding. Only one descriptive study had sufficient power to detect a clinically important effect[15, 170]. Likewise, only two of the RCTs and the double-baseline designed study[177, 178, 180] had sufficient power to detect a clinically important effect. As in the previous review, the single case studies[181-183] had weak external and internal validity, while the remaining intervention studies had variable external validity, risk of bias and risk of confounding.

Table 2.9: Checklist for measuring study quality results

Section	Reporting										External validity			Internal Validity - Bias							Internal validity - confounding						Power
Question	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25	26	27
Descriptive studies																											
Antunes et al[173]	1	1	0	-	1	1	0	-	-	0	0	0	1	-	-	1	-	0	-	1	-	-	-	-	0	-	0
Bezodis et al[171]	1	1	0	-	0	1	1	-	-	0	0	0	0	-	-	1	-	1	-	1	-	-	-	-	0	-	0
Bohm et al[15]	1	1	1	-	1	1	1	-	-	1	0	0	0	-	-	1	-	0	-	1	-	-	-	-	0	-	1
Kratschmer et al[170]	1	1	1	-	1	1	1	-	-	1	0	0	1	-	-	1	-	0	-	0	-	-	-	-	0	-	1
Reina et al[172]	1	1	0	-	1	1	1	-	-	1	1	1	1	-	-	1	-	1	-	0	-	-	-	-	0	-	1
Runciman et al [169]	1	1	1	-	0	1	1	-	-	0	0	0	1	-	-	1	-	1	-	1	-	-	-	-	0	-	0
Runciman et al [168]	1	1	1	-	1	1	1	-	-	0	0	0	1	-	-	1	-	1	-	1	-	-	-	-	0	-	0
Zwinkels et al[174]	1	1	0	-	1	1	1	-	-	1	1	1	1	-	-	1	-	1	-	1	-	-	-	-	0	-	0
Intervention studies																											
Cleary [175]	1	1	1	1	1	1	1	1	1	1	1	1	1	0	0	1	1	1	1	1	1	1	1	1	1	1	0
Fisher-Pipher et al[182]	1	1	1	1	0	1	0	0	1	0	0	0	1	0	0	1	0	0	1	1	0	0	0	0	0	0	0
Gillett et al[176]	1	1	1	1	2	1	1	1	1	1	1	0	0	0	0	1	1	1	1	1	1	1	1	1	0	1	0
Hedgecock et al[181]	0	1	1	1	0	1	0	0	1	0	0	0	0	0	0	1	0	1	0	1	0	0	0	0	0	0	0
Kara et al[177]	1	1	1	1	1	1	1	1	0	1	0	0	1	0	1	1	1	0	0	1	1	1	1	0	0	0	1
Kara et al[178]	1	1	1	1	1	0	1	1	0	1	0	0	0	0	1	1	1	1	1	1	1	1	1	0	0	1	1
Salvador et	1	1	1	1	1	1	0	0	1	0	0	0	1	0	0	1	0	0	0	1	0	0	0	0	0	1	0

al[183]																											
Schranz et al[179]	1	1	1	1	1	1	1	0	0	1	1	1	1	0	0	1	1	1	0	1	1	1	1	0	0	0	0
Van Vulpen et al[180]	1	1	1	1	1	1	1	1	1	1	1	1	1	0	0	1	0	1	1	1	0	0	0	0	1	1	1

-Not Applicable

Table 2.10: Checklist for measuring study quality results – section totals

Section:		**Reporting**	**External Validity**	**Internal Validity – Bias**	**Internal Validity – Confounding**	**Power**	**Overall Total**	**Downs and Black rating**
Maximum possible score	**Number of subjects**	**11**	**3**	**7**	**6**	**1**	**/18 (Descriptive) /28 (Intervention)**	
Descriptive studies								
Antunes et al[173]	T35, T36, T37 and T38 athletes n=4	4	1	2	0	0	7 (39%)	Poor
Bezodis et al[171]	T36 n=1 NT n=16	4	0	3	0	0	7 (39%)	Poor
Bohm et al[15] Kratschmer et al[170]	n= 280 n=64	7 7	0 1	2 1	0 0	1 1	10 (56%) 10 (56%)	Fair Fair
Reina et al[172]	Paralympic footballers n=82; NT=31	6	3	2	0	1	12 (67%)	Fair
Runciman et al[169] Runciman et al[168]	Paralympic athlete n=6; NT=12	5 6	1 1	3 3	0 0	0 0	9 (50%) 10 (56%)	Poor Fair
Zwinkels et al[174]	2004 n=25 2014 n=25	6	3	3	0	0	12 (67%)	Fair

Intervention Studies								
Cleary et al[175]	Intervention=10 Control=10	10	3	5	6	0	24 (86%)	Good
Fisher-Pipher et al[182]	n=1	6	1	3	0	0	10 (36%)	Poor
Gillett et al[176]	Intervention=8 Control=9	11	1	5	5	0	22 (79%)	Good
Hedgecock et al[181]	n=1	5	0	3	0	0	8 (29%)	Poor
Kara et al[177]	Intervention=15 Control=15	9	1	4	3	1	18 (64%)	Fair
Kara et al[178]	Intervention=25 Control=15	8	0	6	4	1	19 (68%)	Fair
Salvador et al[183]	n=1	7	1	2	1	0	11 (39%)	Poor
Schranz et al[179]	PRT=11 HICT=11	8	3	4	3	0	18 (64%)	Fair
Van Vulpen et al[180]	n=22	10	3	4	2	1	20 (71%)	Good

NT=neurotypical; PRT=progressive resistance training; HICT=high intensity circuit training

Data synthesis

Descriptive studies

The systematic review update identified 15 additional studies. Five studies focused on elite adult track athletes with CP[168, 169, 171-173]. These studies reported that athletes with unilateral[168, 169, 173] or bilateral[171-173] CP had increased asymmetry[173], decreased anaerobic capacity[173], decreased aerobic capacity[168, 169], slower maximum sprint speed[168, 169, 172] and were less agile[172] compared to elite neurotypical adults (Table 2.11Table 2.12, Table 2.14 and Table 2.15), in keeping with our original findings[111]. In elite adults with unilateral CP, vertical hop height was decreased on the affected side but not on the non-affected side, and bilateral vertical jump height was decreased compared to the TD group[169]. The authors concluded that performance of bilateral anaerobic tasks in athletes with CP is determined by the affected side, rather than the non-affected side[169]. Paralympic athletes with CP were reported to show sprint times affected by fatigue in the same magnitude as their elite peers without CP[169] and to display conservative pacing strategies over 1600 metres with relative underperformance compared to other neurotypical athletes[168]. A single case study reporting initial sprint acceleration in an athlete with hemiplegia described a reduction in step length, lower limb range of movement, lower limb angular joint angular velocity and therefore power generation compared to neurotypical sprinters (Table 2.12 and Table 2.13)[171].

A cross-sectional study of children with CP in GMFCS Level II, reported that only 67% of children with unilateral CP and 55% of children with bilateral CP could run[15]. Running ability was more likely in the absence of rectus femoris spasticity and the presence of gastrocnemius spasticity. The authors hypothesised that gastrocnemius spasticity contributes to the rapid development of ankle joint moments during running. Hopping ability was positively correlated with running ability, while muscle strength was not.

Another cross-sectional study of children with unilateral spastic CP, GMFCS Levels I-II reported more typical running kinematics than walking kinematics[170]. When running, children with CP had more typical ankle, knee and hip range of motion in swing phase compared to walking, which the authors attributed to the greater influence of central pattern generators in running compared to walking. The incidence of increased ankle plantarflexion in stance was greater in running than walking. The authors recommended the inclusion of running analysis with walking analysis for treatment planning in this population.

Zwinkels and colleagues[174] compared a cohort of children with CP, GMFCS Levels I and II, with a comparable cohort examined ten years previously. From 2004 to 2014 they reported a trend towards increased BMI and body mass, increased aerobic and anaerobic capacity (as measured by the MPST and SRT, respectively) and increased agility as measured by the 10x5m sprint. The authors suggested that these trends could reflect an increasing focus on exercise programs for children with CP and that increased BMI could reflect increased percentage of lean muscle mass. It is unknown whether these trends are associated with an increased ability to run in this population.

Intervention studies

The nine intervention studies published since 2015 included a variety of strength, functional and plyometric activities, but none reported training the motor skill of running, or running gait education. Intervention studies used various assessments as pre- and post-intervention measures, although the muscle power sprint test (MPST) and 10x5m sprint test have become more popular since the last review. The different intervention protocols are summarised in Table 2.14.

Like the original review, the updated review identified intervention studies which reported changes in running capacity or agility but data on spatio-temporal, kinematic or kinetic changes in running were lacking. Combined PRT and functional aerobic training was reported to increase muscle volume of gastrocnemius, soleus and tibialis anterior, which is one possible mechanism for reported improvements in anaerobic power, functional strength and agility[176]. The effect of intervention on the motor control strategy or skill of running was not reported.

Table 2.11: Spatiotemporal-temporal characteristics of running

Paper:	Bezodis et al[171]		Bohm et al[15]		Kratschmer et al[170]		Runciman et al[168]
Spatio-temporal parameters	Elite male T36 athlete n=1	NT elite adults n=8	Bilateral CP Age= 6-17 years n=115	Unilateral CP age= 6-17 years n=48	Unilateral CP, GMFCS Level I-II n=64	TD n=30 age=9.7 yrs (SD=2.7 yrs)	Paralympic athletes with unilateral CP n=6, NT adult national hockey players n=12
Normalised velocity			0.88	0.91	0.96 (SD=0.18)	1.03 (SD=0.21)	CP sprint time over 100m=11.9sec (SD=0.9sec) 40m=5.6sec (SD=0.3sec)* NT sprint time over 40m=5.3sec (SD=0.2sec)
Normalised step length (m)	First=0.75* Second=0.93*	First=1.06 Second=1.09			104 (SD=17)	120 (SD=19.2)	
Normalised step time					1.15 (SD=1.12)	1.15 (SD=0.10)	
Flight time (s)	First=0.04* Second=0.04	First=0.10 Second=0.05					

*Significantly different to NT; %=percentage; Approx.=approximately; CP=cerebral palsy; m/s=metres per second; m=metres; min=minutes; n=number of participants; NT=Neurotypical; TD=typically developing; steps/min=steps per minute; s=seconds; yrs=years;

Table 2.12: Kinematics of running

Paper:	**Bohm et al[15]**		**Kratschmer et al[170]**	
Joint Kinematics Peak values	Bilateral CP GMFCS II 6-17 years n=115	Unilateral CP GMFCS II 6-17 years n=48	Unilateral CP, GMFCS Level I-II n=64	TD age= 9.7 years (SD=2.7 years) n=30
Ankle				
ROM (°)	34°	35°	Affected 35° Non-affected 37°	Dominant 47° Non-dominant 50°
Initial contact	0°	10° PF	Affected 5° PF Non-affected 5°DF	Dominant 0° Non-dominant 0°
Peak Plantarflexion (°)	17°	25°	Affected 18° Non-affected 10°	Dominant 20° Non-dominant 22°
Peak Dorsiflexion (°)	17°	10°	Affected 17° Non-affected 27°	Dominant 27° Non-dominant 28°
Knee				
ROM (°)	45°	55°	Affected 64° Non-affected 60°	Dominant 73° Non-dominant 73°
Flexion at IC (°)	37°	30°	Affected 30° Non-affected 30°	Dominant 25° Non-dominant 25°
Peak Flexion (°)	75°	80°	Affected 80° Non-affected 79°	Dominant 87° Non-dominant 87°
Peak flexion timing as % Gait Cycle	70%	70%	Affected 70% Non-affected 70%	Dominant 68% Non-dominant 65%
Peak Extension (°)	30° flexion	25° flexion	Affected 25° flexion Non-affected 30° flexion	Dominant 20° flexion Non-dominant 23° flexion
Hip				
ROM (°)	50°	50°	Affected 54° Non-affected 60°	Dominant 58° Non-dominant 58°
Peak flexion (°)	60°	55°	Affected 55° Non-affected 57°	Dominant 55° Non-dominant 53°
Peak extension (°)	10° flexion	5° flexion	Affected 5° flexion Non-affected 0°	Dominant 3° extension Non-dominant 5° extension

Pelvis				
Peak anterior tilt (°)	22°	19°		
Peak posterior tilt (°)	17° anterior	12° anterior		

PF=plantarflexion; DF=dorsiflexion; n=number of participants; CP=cerebral palsy; TD=typically developing; ROM=range of motion

Table 2.13: Kinetics of running

Paper:	**Bezodis et al[171]**		**Bohm et al[15]**	
Joint kinetics: peak values	Elite male T36 athlete n=1	NT elite adults n=8	Bilateral CP 6-17 years n=115	Unilateral CP 6-17 years n=48
Metatarsophalangeal joint				
Normalised negative work	-0.021	-0.024		
Normalised positive work	0.005	0.010		
Ankle				
Peak plantarflexion moment (Nm/kg)			1.6	1.55
Peak dorsiflexion moment (Nm/kg)			0.6	0.5
Normalised negative work	-0.073*	-0.043		
Normalised positive work	0.149	0.164		
Knee				
Peak extension moment (Nm/kg)			1.4	1.1
Peak flexion moment (Nm/kg)			0.5	0.5
Normalised negative work	-0.002	-0.004		
Normalised positive work	0.075*	0.110		
Hip				
Peak extension moment (Nm/kg)			1.25	1.25
Peak flexion moment (Nm/kg)			1	1
Normalised negative work	-0.088	-0.082		
Normalised positive work	0.064*	0.133		
Normalised horizontal external power	0.51*	0.82		

*Significantly different to TD; n=number of participants; Nm=newton metres; Nm/kg=newton metres per

Table 2.14: Summary of intervention studies

Author	Downs and Black Rating	Population	Frequency and Duration of Intervention	Description of Intervention	Outcome
Cleary[175]	Good	Unilateral and bilateral CP attending specialist schools; GMFCS I-III; aged 8-18 years; intervention n=10, control n=9	30 min, 3 times per week for 9 weeks	10 min warm up Aerobic exercise at 60-70% of maximum, progressing to 70-80% of maximum, according to interest (eg bike riding, walking, running, football, basketball)	No change in 6MWT Improvement > MPST at week 10, not maintained at week 20. Improved submaximal treadmill test at week 10, not maintained at week 20. No change in participation, school function or quality of life.
Fisher-Pipher et al[182]	Poor	A 15-year-old girl with bilateral spastic CP, GMFCS II	60-70 minutes, twice weekly for 11 weeks plus HEP 2-3 times per week	5-10 min warm up Side-stepping, forward-diagonal step-to, 2.4m shuttle run, 30 sec step-up and down, dynamic balance activities HEP: 15-20min treadmill walking and modified yoga poses.	Improvement > MCID on 6MWT No change in MPST Improvement > MCID on 10x5m sprint (agility) No change in functional strength Improvement in GMFM part D and E Improved performance and satisfaction for 4 goals on the COPM
Gillett et al[176]	Good	Unilateral and bilateral CP; GMFCS I-II; aged 15-30 years; intervention n=8, control n=9	3 sessions per week for 12 weeks	PRT: seated bent- and straight-knee calf raise, leg press, seated tibialis anterior raise, standing calf raise Functional aerobic training: stair climbing, bending, changing direction, stepping over obstacles.	Improvement > MCID in functional strength (p<0.001) Improvement > MCID in MPST (peak power p=0.026, mean power p=0.085) Improvement > MCID in 10x5m sprint (p=0.016) Increased muscle volume (gastrocnemius, soleus and tibialis anterior) Increased strength of plantarflexors, no change in dorsiflexors No change in timed up-stairs or timed down-stairs No change in intramuscular fat or passive muscle properties

Hedgecock et al[181]	Poor	An 18-year-old male with bilateral spastic CP, GMFCS I	One hour, twice per week for 12 weeks, with a twice-weekly HEP	10 min BWSTT Leg press, seated knee extension, resisted dorsiflexion, resisted plantarflexion, half-kneel to stand, curb steps, reverse curb steps, jumping, cone agility drills. HEP: side-lying leg raise, squats, bridges, supine leg lifts, 20-30min run/walk, supine hamstring stretch, standing plantarflexor stretch	No change in MPST No change in 10x5m sprint (agility) No change in functional strength Timed up and go worse No change in 6MWT Achieved 3/3 short-term goals and 1/5 long-term goals
Kara et al[177]	Fair	Unilateral spastic CP; GMFCS I-II; aged 7-14 years; intervention n=18, control n=17	Control and taping groups both received twice-weekly NDT intervention for 12 weeks	Taped 6 days per week with Kinesio® Tape on upper and lower extremities	Taping group: Functional strength improved > MCID; improvement with medium effect size on the WeeFIM Both groups: no change in 10x5m sprint (agility); change < MCID on MPST
Kara et al[178]	Fair	Unilateral spastic CP; GMFCS I; aged 7-16 years; intervention n=17, control n=16	Control: 60 minutes, 3 times per week for 12 weeks Intervention: 90 minutes, 3 times per week for 12 weeks	Control: active therapy (locomotor training, improving symmetry of weight bearing, stretching) Intervention: 5-10 min warm up (e.g. jogging) Leg press, jumping, hopping, balance training on BOSU ball 5-10 min dynamic stretching	Intervention group: Increased lower limb muscle strength with large effect size Both groups: change < MCID on MPST; change < MCID on TUG
Salvador et al[183]	Poor	An adult T37 Paralympic sprinter	3 times per week for 4 weeks	5 min warm up 4x 5 min bouts of walking with	Improved 400m time

		with unilateral CP		pneumatic cuff inflation over the upper thighs, progressing to 7 bouts with increased pressure	Improved running economy Increased muscle strength
Schranz et al[179]	Fair	Unilateral and bilateral CP; GMFCS I-II; aged 8-16 years; PRT n=11, HICT n=11	3 times per week for 8 weeks	Both groups: warm up, 3 circuits (sit-to-stand, heel raise, forward lunge, bridging, lateral step-up), cool down PRT: progressed using a weight vest HICT: maximal intensity for 30 sec with 30 sec recovery	HICT: increased total isometric strength and improvement > MCID on MPST, neither maintained at follow-up PRT: improved TUG and timed stairs test; TUG improvement maintained at follow-up. No change in 6MWT, GPS, energy expenditure index, ASKp or PODCI
Van Vulpen et al[180]	Good	Unilateral and bilateral CP; GMFCS I-II; aged 4-10 years; n=22	One hour 3 times per week for 14 weeks	10 min warm up 35 min power exercises (loaded running, loaded walking, loaded chair push, loaded stair climbing, loaded scooter, loaded side-steps) 15 min game	Improvement > MCID on MPST Improvement >MCID on SRT Increased plantarflexor, knee extensor and hip abductor strength

Abbreviations: 6MWT=6 minute walk test; ASKp=Activity Scale for Kids Performance version; BWSTT=body weight supported treadmill training; COPM=Canadian Occupational Performance Measure; CP=cerebral palsy; GMFCS=gross motor function classification scale; GMFM=Gross motor function measure; GPS=Gait Profile Score; HEP=home exercise program; HICT=high intensity circuit training; MCID=minimum clinically important difference; min=minute; NDT=neurodevelopmental therapy; PODCI=Pediatric Outcome Data Collection Instrument; PRT=progressive resistance training; sec=second; SRT=Shuttle Run Test;

One randomised controlled trial[175] used running as an aerobic activity, although not for all participants, and reported improved aerobic and anaerobic capacity at the conclusion of the intervention; improvements were not maintained at 10-week follow-up. Another randomised controlled trial compared high intensity interval training to progressive resistance training in children with CP and included the muscle power sprint test (MPST) as an outcome measure, although the intervention did not include running activities[179]. Only the high intensity interval training group reported improvements on the MPST. A third randomised controlled trial compared "active therapy" which included locomotor training to progressive functional strength and power training, which did not include running activities, except jogging in the warm-up[177]. Neither group reported clinically significant improvements on the MPST. A fourth randomised controlled trial compared upper and lower body Kinesio® Taping with no taping, both groups received neurodevelopmental therapy twice weekly for 12 weeks[177]. Neither group had clinically significant improvements in the MPST (anaerobic power) or 10x5m sprint (agility).

A randomised prospective controlled pilot study reported that high intensity circuit training was more effective than progressive resistance training in improving anaerobic power as measured by the MPST[179]. A double-baseline study reported that functional power training was effective at improving both aerobic and anaerobic power (as measured by the SRT and MPST respectively) in young children with CP[180].

Two case-studies[181, 182] included one agility drill in the intervention. The dosage was similar, but the participant in GMFCS Level II had significant improvement in agility, while the participant in GMFCS Level I did not have clinically significant change. Neither study reported change in the MPST[181, 182] which may be due to a lack of statistical power. The participant in GMFCS Level II had significant improvements in the GMFM Sections D and E[182] following intervention. A third case study of a Paralympic sprinter reported that walk training with restricted blood flow through the use of pneumatic cuffs was effective at improving 400m sprint times and economy[183].

Table 2.15: Summary of factors affecting running activities in people with CP: summary of findings from the 2016 and 2020 reviews. Grey text indicates items from the 2016 review only, while black text indicates new or extended knowledge.

Running skill	Running capacity		Agility
	Aerobic	Anaerobic	
<u>Spatiotemporal Characteristics</u> Velocity is decreased in children with CP compared to children who are TD[131, 150, 170] and in Paralympic sprinters compared to elite NT adult athletes[168, 169] Step length is decreased in children with CP compared to children who are TD[131, 150, 170] and during the acceleration phase of Paralympic sprinters compared to elite NT athletes[171] Step time is the same in children with CP and TD children[170] Running is less symmetrical in children with CP than children who are TD[150] Step time is more symmetrical than in walking[125, 150] Running is less smooth than walking[150]	<u>Shuttle Run Test</u> Aerobic capacity is higher in individuals in GMFCS Level I than Level II [41] Aerobic capacity is higher in males than females [41] Aerobic capacity increases with height [41] Aerobic capacity of Dutch children with CP, GMFCS I and II, in special education schools improved between 2004 and 2014 (p<0.01)[174]	<u>Muscle Power Sprint Test</u> Anaerobic power is higher in individuals with GMFCS Level I than Level II [42] Anaerobic power is higher in males than females [42] Anaerobic power increases with height [42] Anaerobic capacity of Dutch children with CP, GMFCS I and II, in special education schools improved between 2004 and 2014 (p<0.01)[174]	<u>10 x 5m Test</u> Individuals who are GMFCS Level I have better agility than those with GMFCS Level II [42] Agility is better in males than females [42] Agility decreases with height [42] Agility of Dutch children with CP, GMFCS I and II, in special education schools improved between 2004 and 2014 (p<0.01)[174]
<u>Kinematics</u> Ankle ROM and peak dorsiflexion are reduced in children with CP compared to children who are TD and peak plantarflexion is reduced in children with bilateral CP compared to unilateral CP[15, 131, 170] Children with unilateral CP are more plantarflexed at initial contact on the affected side compared to children with bilateral CP or children who are TD[15, 170] Peak knee flexion and peak knee extension are reduced in children with CP compared to children who are TD and peak knee extension is more reduced in children with bilateral CP than those with unilateral CP[15, 131, 170]			

Peak hip extension is reduced in affected legs of children with CP compared to children who are TD[15, 131, 170] Children with bilateral CP have greater range of movement of the pelvis in the sagittal plane than those with unilateral CP[15, 170] Barefoot running is more asymmetrical than walking [125]			
<u>Kinetics</u> Ankle and knee power generation is significantly reduced in children with CP compared to children who are TD [131] Negative work at the ankle was reduced in a Paralympic sprinter with CP compared to elite NT athletes[171] Peak knee extension moment is greater in children with bilateral CP than children with unilateral CP[15] Positive work at the knee and hip were reduced in a Paralympic sprinter with CP compared to elite NT athletes[171] Hip power absorption is significantly reduced in children with CP compared to children who are TD[131] Power generation and absorption are more asymmetrical than in walking[125]			

CP = Cerebral Palsy; GMFCS=Gross Motor Function Classification Scale; NT=neurotypical; TD = Typically Developing

Discussion

The initial systematic review identified 17 studies published in the 20 years prior which were relevant to running in people with CP. The identification of a further 15 studies in four years highlight the increasing focus on running in people with CP. Areas of focus over the last four years include Paralympic sprinting, the difference in running between unilateral and bilateral CP, running kinematics in children with CP and the change in running capacity and agility in children with CP over time (Table 2.15). Increased attention to running in people with CP reflects an increasing interest in the broader area of physical literacy in

youth with neurodevelopmental disabilities[119, 184]. Within this framework, competence in the skill of running is expected to improve confidence and motivation, which in turn increases participation in physical activity and positive affect[118]. These factors are considered important for improving physical, mental and social health[118].

The updated review supports the findings of the initial review that people with CP: 1) run more slowly than their able-bodied peers; 2) run with a shorter stride length; 3) run with a higher incidence of forefoot striking; and 4) that the characteristics of running in people with unilateral CP are different to those with bilateral CP.

One study hypothesised that muscle strength was not correlated with running ability in children with CP in GMFCS Level II because the running velocity in the study was slow (jogging) and therefore the strength demand was not as high as for sprinting[15]. However, running is a motor task which is performed quickly, the entire stance phase takes less than 0.25 seconds[62]. Measures of muscle strength are undertaken at a much slower velocity and therefore are not task-specific for walking and running. In contrast, hopping (i.e. vertical single leg jumps), which was shown to be correlated to running ability[15], requires a rapid generation of ankle power for push-off.

As in the previous review, various outcome measure were used to assess running in the intervention studies. The MPST, SRT and 10x5m sprint continue to be used appropriately to assess anaerobic power, agility and aerobic power respectively. The GMFM contains only one item of 66 (or 88) that involves running[185], while the BOT-2 contains one running speed and agility item out of 47 items[155]. Neither assessment considers movement quality during running and are probably not the measure of choice for intervention studies focussed on running. The Challenge Module[186], Test of Gross Motor Development – third edition [90, 187] or the HiMATmay be more appropriate[188, 189], although only the TGMD-3 has any assessment of running quality, and this over a short distance (15 metres). A new measure may be needed for intervention studies that want to measure change in running in children with CP.

Together, studies published to date suggest that interventions including running are feasible and safe in children and adolescents with CP and may result in functional improvements. In general, intervention protocols which included power or plyometric activities had positive outcomes on the MPST, protocols which included progressive resistance training (PRT) had positive outcomes for muscle strength, while protocols which included agility exercises had positive outcomes for the 10x5m sprint test. These findings

reinforce the importance of the principle of training specificity for people with CP. However, there remains no information in the published literature regarding training running skill in people with CP, nor the effect of intervention on the kinematics and kinetics of running. The systematic review update identified only one study that considered increased muscle volume in the shank as a mechanism of improvement in running capacity with no outcome measure of running skill. Mechanisms for improving running skill therefore remain unexplored. It is unknown whether people with CP learn to use abnormal strategies more effectively or can normalise the kinematics and kinetics of running.

Chapter 3 Research methodology

3.1 Preface

This chapter will outline the methodology of the studies in this thesis, which has also been presented in the following research outputs:

"Chappell, A., Gibson, N., Williams, G., Allison, G.T. and Morris, S., 2019. Propulsion strategy in running in children and adolescents with cerebral palsy. Gait & Posture, 70, pp.305-310." https://doi.org/10.1080/09638288.2017.1367426

"Chappell, A., Allison, G. T., Williams, G., Gibson, N., & Morris, S. (2020). The effect of a running training intervention on ankle power generation in children and adolescents with cerebral palsy: A randomized controlled trial. Clinical Biomechanics, 76." https://doi.org/10.1016/j.clinbiomech.2020.105024.

This chapter includes details and rationales for the research instrumentation, data collection and processing workflow. There were some methodological challenges to overcome, which resulted in work which will be presented in Chapter 4 of this thesis and was published as:

"Chappell, A., Liew, B., Murphy, A.T., Gibson, N., Allison, G.T., Williams, G. and Morris, S.L., 2019. The effect of joint translation constraint on within-participant variability of kinematics and kinetics during running in cerebral palsy. Clinical Biomechanics, 63, pp.54-62." https://doi.org/10.1016/j.clinbiomech.2019.02.003

3.2 Systematic review

A systematic review was undertaken to synthesise research describing running and running interventions in people with CP. The review was published and subsequently updated. The systematic review and systematic review update methodology is reported in detail in Chapter 2.

3.3 Evaluation of joint constraint boundaries

A comparison was undertaken of lower limb joint powers calculated with three different Inverse Kinematic (IK) models with global optimisation. The study is reported in Chapter 4. One stride of each participant was processed three times using three different models. All three models used inverse kinematics with global optimisation. The IK3 model had three mobilisers at each joint, the IK6 model had six mobilisers and the IK6C model had six mobilisers with custom constraints derived from the literature. Hip, knee and ankle joint angles, moments and powers were calculated and compared. The outcome of this study informed the data processing protocol employed in the experimental studies.

3.4 Experimental studies

Four experimental studies were designed. Two studies investigated power for forward propulsion during running in children and adolescents with CP and compared this to a sample of typically developing children. Two further studies investigated the effect of a task-specific, low-load plyometric running training intervention on power for forward propulsion in children and adolescents with CP.

3.4.1 Participants

Participants were recruited through a community service provider. Participants had to be aged 9-18 years, have cerebral palsy or like condition, GMFCS level I or II and have a goal related to running. Phone calls were made to eligible participants explaining the study and inviting them to participate. Participants had to be willing and able to attend two training sessions weekly after school and complete two additional exercise sessions per week at home. Participants were excluded if they had undergone orthopaedic surgery in the previous six months; had a medical condition which contraindicated strenuous exercise; were unable to complete assessments due to a cognitive impairment; or had cognitive or behavioural impairments which precluded intervention in a group setting. Parents gave written consent and participants gave assent where applicable. All study forms are included as Appendices. A stratified random allocation to control or intervention group was conducted by an independent team member using computer-generated numbers[190]. Allocation was stratified by age and High-Level Mobility Assessment Tool (HiMAT)[191] score to keep groups equivalent. Participants were notified of their allocation after the baseline assessments were completed.

3.4.2 Ethics

All studies in the thesis were approved by the Child and Adolescent Health Service Ethics Committee, Perth, Western Australia (201405SEP) and the Human Research Ethics Committee of Curtin University, Perth, Western Australia (HR 219/2014). The trial was prospectively registered with the Australian New Zealand Clinical Trials Notification ACTRN12614000467639. Informed consent was given by the parent/guardian and assent by the participant where applicable.

3.4.3 Funding support

This work was supported by an Australian Government Research Training Program Scholarship, a Non-Government Centre Support Grant and Perth Children's Hospital Foundation Grant ID 9632.

3.4.4 Study flow

Participants attended four assessment sessions at Curtin University, Bentley. Immediately following the baseline assessment, participants in the intervention group attended 12 weeks of the running training program. All participants were then re-tested. Following the second assessment session, participants in the control group were offered the 12-week running training program. All participants then attended a third assessment session. Participants in the control group then attended a fourth assessment session 12 weeks following the cessation of the running training program. The study plan is presented in Figure 3.1.

3.4.5 Derived variables

The main biomechanical variables of interest were:

1. Leg stiffness during a sub-maximal hopping task
2. Ankle power generation in stance (A2)
3. Hip power generation in swing (H3)
4. Leg stiffness in the stance phase of running

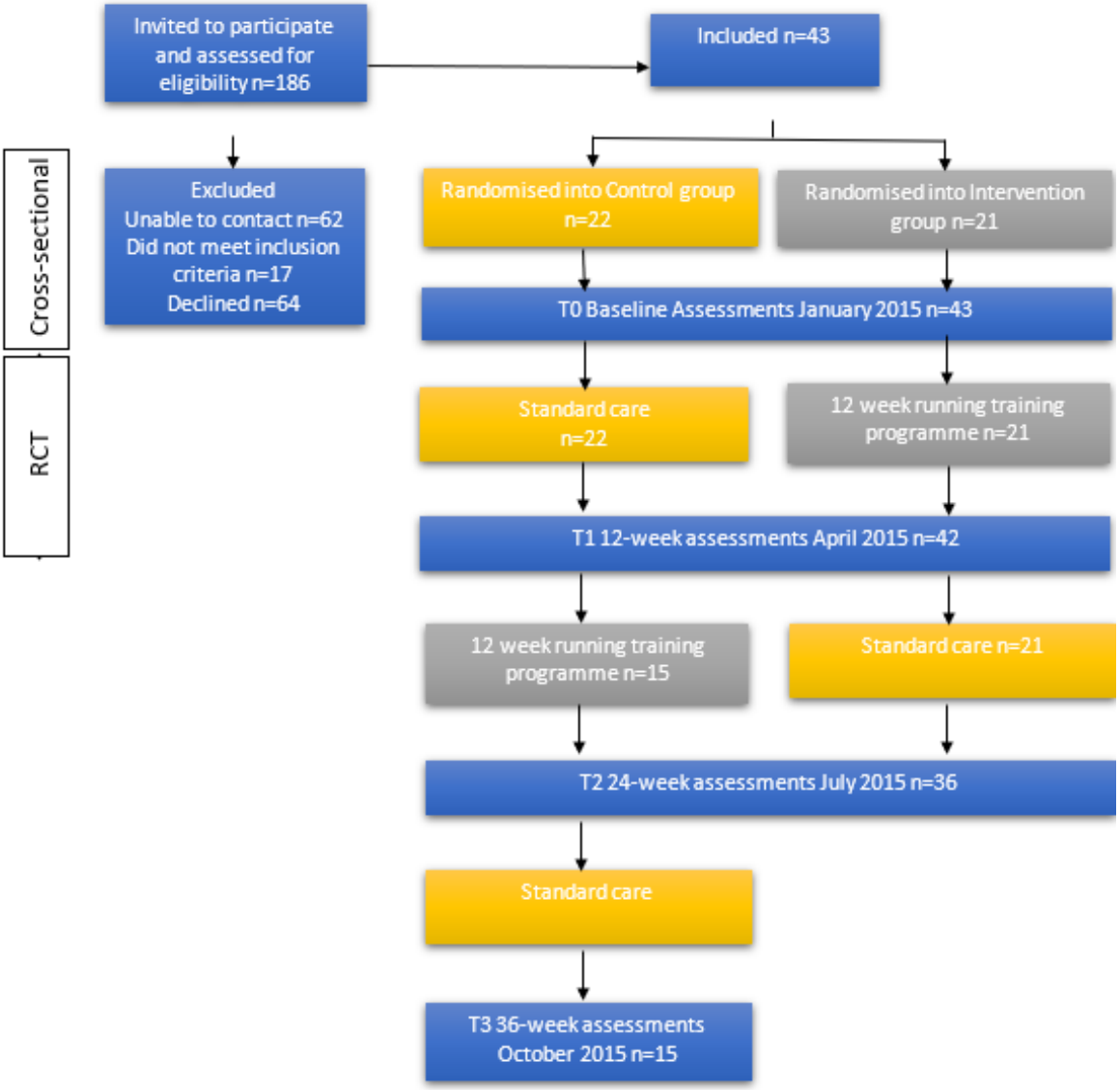


Figure 3.1: Study plan

3.4.6 Instrumentation

3.4.6.1 Sleigh

Leg stiffness was estimated during a sub-maximal hopping task which was performed on a custom-built leg sled. The leg sled was used to optimise reliability and facilitate the task by unloading the limb[192]. An AMTI© force plate was installed at 90° to the base of the sled (Figure 3.2) and connected to a laptop with Labview© program which recorded flight time and contact time. A counterweight was available to nullify the weight of the sled if required

to successfully complete the task. Participants performed 20 consecutive one-legged hops at a self-selected frequency. Data were collected from both right and left legs. Ten consecutive hops in the middle of the 20 were chosen for analysis. Leg stiffness was calculated using Dalleau's equation[193]:

$$K_{leg} = \frac{m\pi(t_f + t_c)}{t_c^2(\frac{t_f + t_c}{\pi} - \frac{t_c}{4})}$$

where m=body mass, t_f=flight time and t_c=contact time. This method has been shown to be valid when assessing leg stiffness during submaximal hopping[194] and related to repeated fast-SSC function, such as in running, in male youths[195].

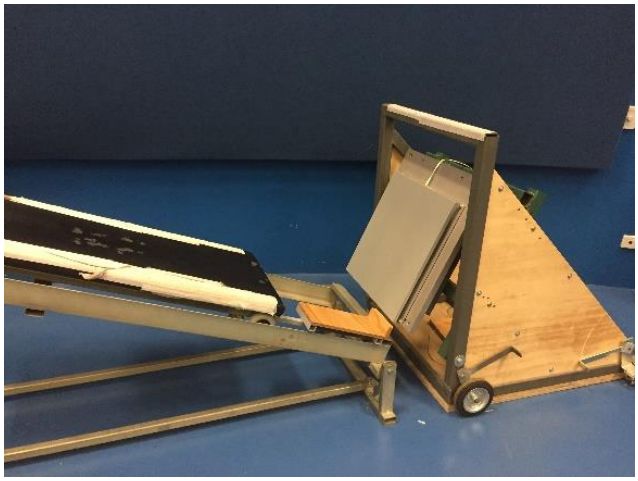


Figure 3.2: Custom-built sleigh for assessment of leg stiffness during hopping

Two-legged hopping on a force platform or contact mat has been shown to be modestly reliable in both male[195] and female[196] youth. Previous studies have reported differences in leg stiffness between dominant and non-dominant legs[197], so a one-legged hopping task was employed in this body of work. Children with CP often have difficulties with balance and control so that one-legged hopping is difficult. The use of a sleigh (Figure 3.2) could be expected to improve reliability and repeatability by reducing balance demands, and has been shown to be a valid measure of leg stiffness[198]. One legged hopping on a sleigh has been reported to be highly reliable in healthy adults[199] although reliability may be reduced if the upper body is restrained[200]. No upper body restraint was used in this body of work.

A variety of testing protocols have been employed in the research literature, either using a specified number of hops ranging from 5 to 30[195, 196], or a series of 10-20 second trials[201, 202]. Some protocols have selected 10 consecutive jumps for analysis[203]. This protocol was adopted primarily to capture steady-state hopping, but also to minimise "settling into the

task" during the first hops and fatigue during the last hops.

3.4.6.2 Three-dimensional gait analysis

Three-dimensional marker tracking systems allow computer simulation of movement which is used to estimate joint kinematics and kinetics. Reflective markers are attached to the skin and tracked by infra-red cameras, while the ground reaction force (GRF) is measured by force plates embedded in the floor (Figure 3.3). Marker placement correlates with a biomechanical model utilised by computer software to simulate the movement.

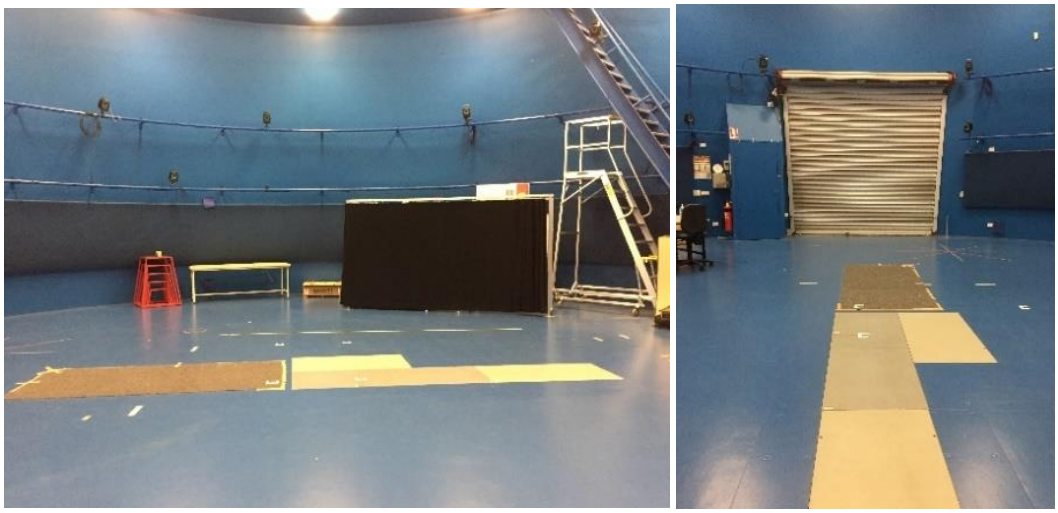


Figure 3.3: The motion analysis laboratory

Spatiotemporal variables, kinematics and kinetics measured with three dimensional gait analysis (3DGA) are reliable and repeatable during walking in children with CP I[204-207]. Reliability is greater in children classified as GMFCS level I than in GMFCS level II[205]. Specifically, for kinematic reliability ≥ 0.9, four walking strides are required for children in GFMCS level I, but 8 strides for children in GMFCS level II[205].

In neurotypical adult runners, kinematic variables have excellent reliability (> 0.93) over five trials[208]. Kinetic variables have been reported to have excellent reliability (>0.93) over 10 trials and good reliability (>0.8) over 3 trials[208]. Repeatability is greater within-day than between-days, and running at a self-selected speed is not more or less repeatable than running at a specified speed[209]. For the present study it was considered that constraining running speed to a standardized speed may change the natural running style of the participants as they were untrained and potentially had mild intellectual disability.

Therefore, to make it as simple as possible, it was decided to allow self-selected speed within three defined speed categories – "jogging", "running" and "sprinting". To maximise reliability, each participant was asked to complete at least five trials in each speed group. It was recognised that this might be difficult for sprinting, where a participant was visibly fatigued, three sprinting trials were considered sufficient.

Inherent in the use of computer modelling is the introduction of error, as a mathematical model cannot exactly replicate the complex and individual movement of each human body[210]. Sources of error include:

1. Accuracy of marker placement[209, 211, 212]
2. Modelling of the foot as a single segment[213, 214]
3. The movement of skin markers with respect to the underlying bone, termed soft tissue artefact (STA)[215-217]
4. Mislocation of the joint centre of rotation[218-220]

Measures adopted to minimise these sources of error during the present work are discussed in the following paragraphs.

Accuracy of Marker Placement

Inaccurate marker placement can result in large inter-participant variation[221], which in turn reduces statistical power making it difficult to detect true changes in gait due to intervention, pathology or the passing of time[222]. Thigh and shank clusters may reduce error from marker placement, compared to using single markers[221] and this method was adopted for the present work. To further maximise accuracy and reliability, all marker placement was undertaken by the principal investigator, who is an experienced physiotherapist.

Foot modelling

The foot and ankle comprise a complex segment composed of many joints which move in all three planes. While it is possible to model the foot as multiple segments, it is not possible to compute kinetics when more than one segment is in contact with a force plate[223]. As a large proportion of power for propulsion in running is generated in stance phase; limiting data to swing phase only was not an option for this work. Modelling the foot as a single segment assumes rigidity of the segment[223], an assumption which is not met in the barefoot condition, especially in children with CP[224]. The shod condition better meets

the assumption of rigidity but the foot can move inside the shoe[225, 226], introducing another inaccuracy. As this work required data collection over the course of a year, it was also possible that participants would change their running shoes, which would affect kinematic and kinetic data[227]. It was therefore decided to collect both barefoot and shod data, requesting that participants use the same shoes at each data collection if it were possible. In the barefoot condition a marker set was used which could allowed processing both as a multi-segment foot or as a rigid foot.

Soft Tissue Artefact

In the lower limb, STA is greatest at the thigh[228] due to a larger amount of adipose and muscle tissue between the skin and the bone, compared to the shin or the foot. STA can be attenuated by using a thigh cluster of markers[228], either with a wand or without a wand[229]. The decision was made to use a thigh cluster without a wand, as it was thought the presence of the wand could potentially alter natural movement.

Inverse kinematic (IK) modelling techniques utilizing joint constraints are reported to minimise the effect of STA[228, 230, 231]. An analysis was undertaken to compare the effect of three different joint constraint protocols on intra-subject variability, with the aim of minimising error due to STA. This analysis is reported in Chapter 4. Based on this analysis, an IK model with customised joint constraints developed from the research literature for children with CP was utilised for data processing.

Marker set employed in the experimental studies

Reflective markers 10mm in diameter were placed on the skin of participants by an experienced physiotherapist using a modified Cleveland Clinic Foundation marker protocol[232] which is routinely adopted in the Curtin University Motion Analysis Laboratory. Details of marker placement are provided in Table 3.1 and illustrated in Figure 3.4. Anatomical markers not used for tracking were removed after the static and dynamic calibration trials.

Table 3.1: Marker set

SEGMENT	ABBREVIATION	PLACEMENT/FULL NAME	Anatomical	Tracking
Foot	RMT1, LMT1	Right/Left 1st metatarsal head	✓	✓
	RMT5, LMT5	Right/Left 5th metatarsal head	✓	✓
	RCAL, LCAL	Right/Left calcaneus (middle of posterior aspect)	✓	✓
Shank/leg	RMMAL, LMMAL	Distal end of tibia, medial malleolus	✓	
	RLMAL, LLMAL	Distal end of fibula, lateral malleolus	✓	
	RTB1, LTB1 RTB2, LTB2 RTB3, LTB3	Right/Left Tibia: Long bar runs along tibia axis and the short bar wraps laterally		✓
Thigh	RMFC, LMFC	Right/Left Medial Femoral Condyle	✓	
	RLFC, LLFC	Right/Left Lateral Femoral Condyle	✓	
	RTH1, LTH1 RTH2, LTH2 RTH3, LTH3	Right/Left Thigh: Long bar runs along Iliotibial band and short bar wraps anteriorly		✓
Pelvis	RASI, LASI	Right/Left Anterior Superior Iliac Spine	✓	✓
	RPSI, LPSI	Right/Left Posterior Superior Iliac Spine	✓	✓

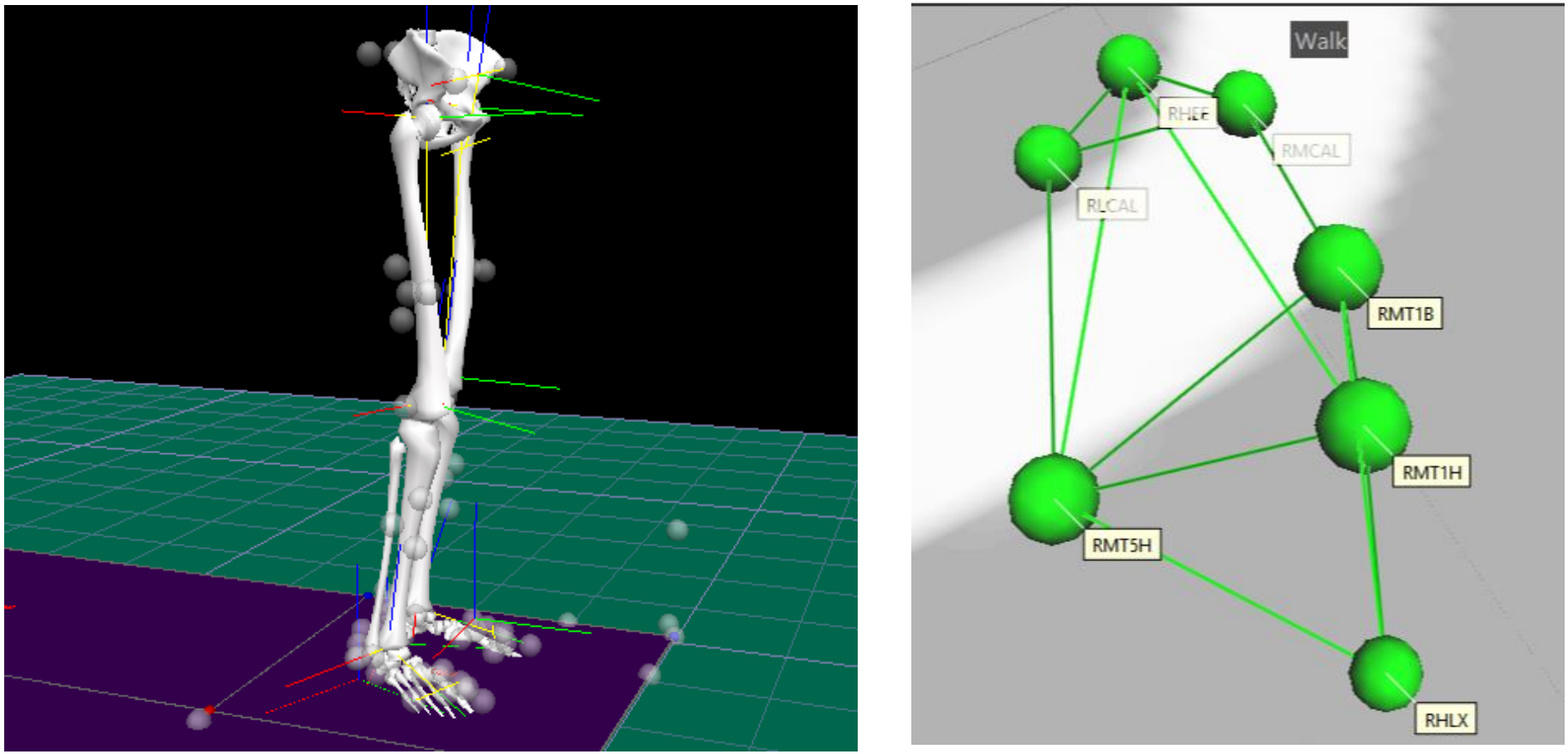


Figure 3.4: Marker set of the lower limb and foot

Data collection procedure

The order of barefoot and shod gait was randomised by a coin toss at the first data collection and the order maintained at subsequent testing sessions. Static and dynamic calibration trials were collected for each condition. Following the collection of static and dynamic calibration trials participants were asked to walk back and forth across the force plates 10 times to warm up and get used to moving with the markers on. Participants were then asked to perform at least five trials at these speeds:

1) jog "like a warm up or like a jog around the oval at school"
2) run "faster than jogging, but not your fastest" and
3) sprint "like you are in a race"

Ten metres were available before and after the force plates to allow for acceleration and deceleration. A two-minute sitting break was permitted between speeds if required. Kinematic data were recorded by an 18-camera motion capture system at 250Hz (Vicon T-series, Oxford Metrics, UK). Synchronised ground reaction forces (GRF) were collected at 1000Hz using three in-ground force platforms in series (AMTI, Watertown, MA). For the shod condition, participants wore their usual sport shoes; calcaneus and metatarsal markers were placed on the shoes over the relevant landmark. In-shoe orthotics were permitted, while any orthotics extending above the malleoli were not. Marker trajectories were labelled and filled using Vicon Nexus 2.5 (Vicon Motion Systems, Oxford, UK).

Data Processing and Conditioning

Trials were cleaned and labelled in Vicon Nexus 2.5 and exported to Visual 3D™ version 6 (C-Motion, Inc.) for processing. When analysing 3DGA data it is necessary to decide whether to select a single representative trial or to average data from several trials once errorneous trials have been excluded[233]. For the present work, non-representative trials were excluded by visual inspection of the principle investigator[234]. Non-representative trials were identified by examination of outlying data points and subsequent visual confirmation that:

- the participant decelerated over the force plates,
- the participant included an action other than running, for example galloping or skipping, or
- the participant lost balance during the trial.

Where derived variables of interest were peaks, for example A2 or H3, a mean was

calculated across included trials to improve reliability[208]. Where a maximum value was of interest, for example, maximum speed, a single representative trial was included.

Kinematic data were filtered at 18 Hz using a zero-lag 4th order Butterworth filter. Force plate data were unfiltered. A regression equation was used to calculate hip joint centres[235]. Knee joint centres were calculated as the midpoint between the medial and lateral femoral condyles[236]. Ankle joint centres were calculated as the midpoint between the medial and lateral malleoli[236]. Inertial and geometric properties of the segments were based on previously published models[237, 238]. Derived variables (A2, H3 and leg stiffness during the stance phase of running) were calculated in Visual 3D™.

3.4.7 Training program

Following baseline data collection, children in the intervention group underwent 12 weeks of twice-weekly, individualised intervention in a group setting, with a home program. The intervention will be described in this section according to the Consensus on Exercise Reporting Template (CERT)[239]. The description has been published in the research output included as Appendix 1: "Gibson, N., Chappell, A., Blackmore, A. M., Morris, S., Williams, G., Bear, N., & Allison, G. (2018). The effect of a running intervention on running ability and participation in children with cerebral palsy: a randomized controlled trial. *Disability and rehabilitation*, *40*(25), 3041-3049." This paper is included in the Appendices because the focus of the paper was activity and participation, while this thesis is concerned with the biomechanics of running.

3.4.7.1 Overview of the intervention

Although most people with CP at GMFCS Levels I and II can run[15], a training program specifically for the skill of running could not be identified in the published literature nor in clinical CP networks. However, a running training program for adults with acquired brain injury (ABI)[240], demonstrated to have been effective in improving running ability and skill[241], was identified. Adults who sustain an ABI present with muscle weakness, spasticity and deficits of motor control[240], which while not identical, are not unlike that of people with CP. In both ABI and CP populations, gains in muscular strength through strength training programs frequently fail to translate into functional gait improvement[240, 242] as the velocity of muscular contraction is not task-specific for gait[62]. It was not known whether the program would be effective in improving running skill in people with CP.

The running training programme for adults following ABI was modified to make it engaging

for the age of the participants, but the core philosophy and set of hierarchical exercises was the same. The programme was able to be delivered safely in the community with good adherence and resulted in 86% of participants achieving or exceeding their individual running goals[190]. The intervention was provided in a group setting but the activities were individualised for each participant. Participants were expected to attend twice per week and to carry out a home program another two times per week. Participants could choose to attend either of two community locations, located about 30 minutes' drive from each other. The group was staffed by six physiotherapists who were specifically employed to carry out the intervention. All six physiotherapists, including the doctoral candidate, received the same training prior to commencing the study. The fidelity of the intervention was overseen by a study supervisor who was not involved in the collection or analysis of biomechanical data.

3.4.7.2 Description of the intervention according to CERT guidelines

1. Exercise Equipment
    a. Warm up only: exercise bike, elliptical

Intervention: Total Gym® leg sled or similar, trampette, stairs, resistance bands, single step, cones, agility ladder (Figure 3.5)

Figure 3.5: Examples of equipment used - Total Gym®, trampette, stairs and agility ladder

2. Qualifications and training

    Six base-grade physiotherapists with experience in working with school-aged children with CP delivered the program. All therapists involved in delivering the program had undertaken 15 hours training in rehabilitation of running for people with neurological impairment. All physiotherapists also undertook eight hours training in motivational interviewing delivered by a clinical psychologist.

3. Individual/Group

   Intervention was delivered in a group setting. Individualised programs were established and progressed at individual rates. Participants attended the group twice per week and an individualised home programme was provided to be performed twice per week.

4. Supervision

   Participants were supervised in a ratio of one physiotherapist per three participants. Each group was comprised of ten participants. The participants were taught the exercises individually and once performing the exercise correctly, allowed to practice independently within the group setting or as part of a home programme.

5. Adherence to exercise

   Progress notes were completed at the end of each session, including the level of difficulty of each exercise and number of repetitions, or time spent doing the exercise. Attendance was recorded for each participant as the number of sessions attended out of 24 possible sessions. Home program exercises were prescribed weekly and home exercise diary sheets were collected at the end of each week.

6. Motivation strategies

   Participants were encouraged with verbal feedback about their technique, both what was done well and what changes needed to be made. Participants were given a time or number of repetitions to aim for. Exercises were incorporated into games or challenges where practicable. Participants were also encouraged to 'buddy up' with another group participant to encourage each other. Each participant had a home program, with diary sheets issued and collected weekly to encourage adherence. Participants received a weekly newsletter from a study supervisor that highlighted the rationale and purpose of different aspects of the exercises, for example, what an exercise was trying to achieve related to the skill of running, the importance of frequency of practice, the importance of practicing the exercises properly, the importance of getting the correct 'dose' of the exercise prescribed etc. Participants who did not attend a session without informing physiotherapists of the reason were called by one of the physiotherapists the next day to encourage attendance. The physiotherapists used motivational interviewing techniques to encourage participants to explore options for adhering to home prescribed exercises and for exploring and promoting physical activity in the community.

7. Decision rule(s) for determining progression

   Once the participant was consistently able to perform the exercise with good technique they were progressed to the next level. Speed and quality of movement were

prioritised over-load as the focus was on ballistic movement necessary for running. The program incorporated a series of hierarchically challenging 'pre-runner' and 'runner' activities[240, 243]. Activities early in the hierarchy included single joint activities at loads less than body weight and/or at slower velocities. Activities later in the hierarchy included multi-joint activities with full body weight loading at higher velocities. The final stage of the hierarchy is comprised of complex tasks including agility exercises, the introduction of ball skills, or game play. Participants were prescribed relevant activities to address the running gait impairments demonstrated for that individual. Within the hierarchy protocol, individually tailored exercises were derived by the therapist viewing slow motion observational sagittal and frontal video footage of the participant's running gait and determining the abnormalities affecting the acquisition of typical running skill. The exercises/activities targeted the three main muscle groups responsible for forward progression when walking and running, i.e. the ankle plantar flexors, hip flexors and hip extensors. Exercises were progressed once good form/technique was demonstrated on the starting activity. Good technique was defined as movement with sufficient force through full range with minimal accessory movement and correct timing.

For pre-runners, progression of propulsion and running exercises occurred on the Total Gym® leg sled until the highest level was reached. The exercises were then performed on a small trampoline until the participant was able to perform the exercises overground. For simulation of leg turnover and appropriate foot contact alignment an activity termed the "claw" exercise was used (see below)[240, 243]. For participants whose motor control did not enable good technique, activities such as the "claw" were either broken down into components or facilitated with therapist handling until the participant was able to perform a cycle with good technique. This was progressed by decreasing therapist facilitation and eventually adding resistance (for example with resistance bands). Once the participant progressed from a pre-runner to a runner and could run with good technique overground, slopes were added and the distance or speed increased depending on the individual goal. From slopes, participants progressed to agility exercises. These began with simple cutting/side-stepping exercises and progressed in complexity. Once a reasonable level of agility was attained, sport-specific skills relevant to the individual participant's interest were introduced.

8. Exercises

   Each session followed the following structure:

   i. Warm up (usually on a stationary bike, elliptical or cross trainer): 5 minutes

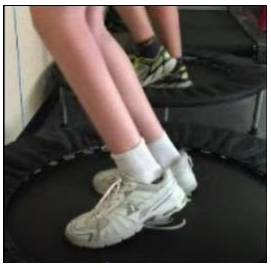
ii. Individualised skill development activities: 50 minutes

iii. Passive and active stretches: 5 minutes

Activities were grouped into three main areas of focus and progressed as follows:

a) Ankle power generation

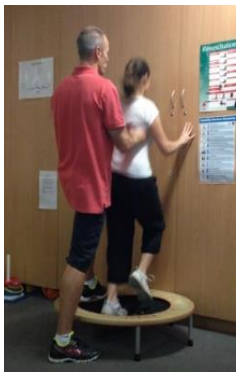
a. Leg sled



b. Trampoline

c. Overground

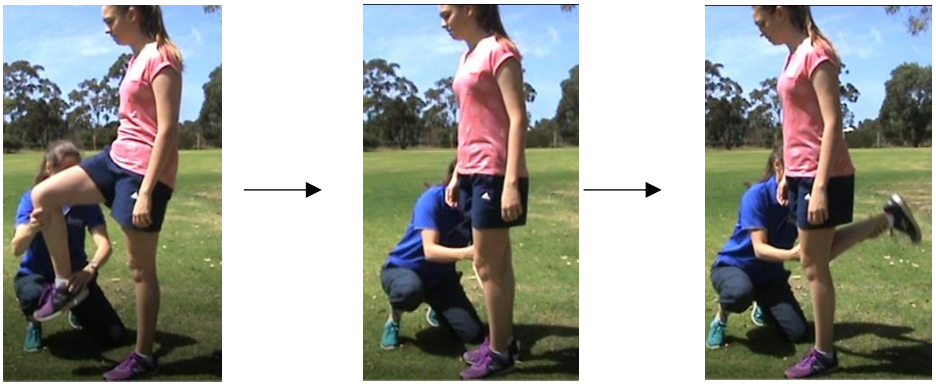
b) Hip power generation



a. Trampoline

b. Overground

c) 'Claw' for running motor pattern



9. Home program

All participants received a home program to be performed twice weekly, which contained individually tailored exercises that had been learned with the

physiotherapists and which they could perform independently with good technique. Participants were asked to complete the home program on two additional days to the two training days that they attended.

10. Non-exercise components

    None.

11. Type and number of adverse events

    One participant sustained a sprained ankle, which required first aid and resolved within one week with full return to activity.

12. Setting

    Two locations were utilised:

    - A public oval with sports clubroom
    - A community service provider with an adjacent public reserve

13. Exercise intervention

    Participants were asked to attend two one hour sessions per week with the home program performed another two times per week, for a total of 12 weeks.

14. a) Generic/Tailored

    Each participant received an individually tailored program based on a core group of hierarchical exercises, delivered in a group setting and as a home exercise programme.

    b) How the exercises were tailored

    The exercises were tailored for each participant according to the identified impairments impacting their ability to run and by level of difficulty. The physiotherapists progressed the exercises according to the participant's response. Adjunct exercises were added by the physiotherapists if necessary, for example hip abductor strengthening exercises were added if the participant could not stabilise the pelvis while performing the exercises.

15. Decision rule for starting level

    Participants were started at the most challenging level they could perform with good technique.

16. a) Adherence/Fidelity

    To minimise bias the fidelity of the intervention was overseen by a study supervisor who was not involved in the collection of the biomechanical data. Physiotherapists met for 10 minutes following each session to discuss issues experienced by individuals in the group and find solutions. A study supervisor visited each site where the group program was being delivered to ensure the integrity of the program was being maintained, and to troubleshoot any intervention queries from the administering therapists. This

procedure reinforced similar administration of the intervention between physiotherapists at each site and adherence to the program. Additionally, all physiotherapists delivering the program met on two further occasions and presented a total of six case presentations of intervention delivery and progression of the activities. These case presentations were chaired by NG and aimed to reinforce and ensure the intervention strategies and principles were being adhered to.

b) Was the intervention delivered as planned?

The intervention was delivered as planned. Median number of sessions attended out of a possible 24 training sessions in the 12-week intervention was 16 (66.7%), with minimum four (16.7%) and maximum 21 (87.5%).

3.5 Statistical analysis

Biomechanical data were conditioned in IBM SPSS® Statistics for Windows and imported to Statistical Analysis System (SAS®) software. Where data were skewed, Box-Cox transformations were applied. Mixed models were then used to examine between-group differences. This method accounted for random effects from the inclusion of multiple data points belonging to both legs of each participant and did not require equal numbers of data points from each subject[244].

As this body of work considered multiple variables for statistical analysis, it was necessary to consider whether it was appropriate to adjust the *p*-value of significance. This adjustment is often recommended to reduce the likelihood of a Type I error, that is, finding a significant difference between groups which is, in fact, due to chance[245]. The statistician at the School of Physiotherapy and Exercise Science was consulted, and she indicated that such an adjustment was not necessary. Firstly, Bonferroni adjustments apply when asking whether two groups are the same *in all respects* [245]. This body of work asked specific questions about derived variables which were considered independently, so the Bonferroni adjustment does not apply. Secondly, making a Bonferroni adjustment simultaneously increases the likelihood of a Type II error, that is, not finding a significant difference between groups which are, in fact, different [245]. In this body of work, given n=43, the likelihood of a Type II error was a concern, therefore making a Bonferroni adjustment may make it more likely that the findings would be misinterpreted. It has been recommended that analysis of kinematic and kinetic variables during running be undertaken with a minimum of 25 neurotypical adult participants and a minimum of 25 steps per participant

at a self-self selected speed [246]. The RCT in this body of work included 21 subjects in each group, who ran at three different speeds, with varying numbers of trials before fatigue. Therefore, the chance of a Type I error was deemed less likely than the chance of a Type II error. The *p*-value of significance for all analyses was set at 0.05.

Examples of mixed models employed in this thesis are:

Baseline leg stiffness

```
proc mixed data=work.tk;
class Subject Descriptor Side GMFCS;
model KlegDL = Descriptor SPS Descriptor*SPS / solution CL DDFM=KR;
random Subject Subject*Side;
lsmeans descriptor / at sps=1 pdiff CL;
run;
```

Randomised controlled trial: A2

```
proc mixed data=work.Ps_a2trans;
class Subject Affected Side Group Time GMFCS trial;
where Affected = 'A' and GMFCS = '1';
model Mean_A2=Time Group statures_per_sec Group*Time
Time*statures_per_sec Group*statures_per_sec
Time*Group*statures_per_sec / solution residual;
repeated Time / type=AR(1) subject=Subject*Side*trial R RCorr;
lsmeans Time Group Group*Time / pdiff;
run;
```

Linear mixed modelling allows post-hoc comparison of fixed effects at specified values of covariates. In this body of work, least-squares means were compared at normalized speeds of 1, 2 and 3 to reflect jogging, running and sprinting respectively. These comparisons are reported as *t*-values.

Statistical analyses were performed based on each study hypotheses. These are detailed in the methods sections of the relevant studies reported in Chapters 4, 5, 6, 7 and 8.

Chapter 4 The effect of joint translation constraint on within-participant variability of kinematics and kinetics during running in cerebral palsy

4.1 Preface

This chapter begins with an examination of the effect of including joint constraint boundaries in a biomechanical model on the estimation of joint powers at the ankle, knee and hip across the jogging gait cycle. As the CP population is a heterogenous one, there is a high degree of variability in CP gait data[12, 23]. Use of modelling techniques which minimise extrinsic variability, that is, variability introduced by the data processing workflow, is desirable in order to both improve precision and to avoid failing to detect a true difference between groups (i.e. improve statistical power).

A known-groups analysis was undertaken to determine whether the use of specified joint constraint boundaries would result in less variability in biomechanical data. A preliminary examination of the effect of joint constraint modelling on joint power was undertaken. Further analysis to identify how joint angles and joint moments were affected by modelling joint constraint boundaries, during jogging, running and sprinting, was then undertaken and is presented as a published paper in section 4.3.

The resulting analysis, which is presented as section 4.3 of this chapter, was published as "Chappell, A., Liew, B., Murphy, A.T., Gibson, N., Allison, G.T., Williams, G. and Morris, S.L., 2019. The effect of joint translation constraint on within-participant variability of kinematics and kinetics during running in cerebral palsy. Clinical Biomechanics, 63, pp.54-62." https://doi.org/10.1016/j.clinbiomech.2019.02.003

4.2 Inverse Kinematic modelling with joint constraint boundaries

Historically, clinical gait analysis used direct kinematics to estimate the position of each segment directly from tracking markers[131, 247]. In direct kinematics, the movement of one segment is considered independently to the movement of any another[248]. While this avoids the propagation of modelling errors distally[249], considering segmental movement

independently does not manage error from STA which may be severe enough to result in apparent joint dislocations[230, 231, 248]. To counter this, the Inverse Kinematic (IK) method was developed, initially using segmental optimisation to estimate the position of each segment from tracking markers using a "best fit" approach, minimising the weighted sum of squared distances between measured and model-based marker positions[230, 248]. Later, IK with global optimisation went a step further, estimating the position of the entire segment chain from tracking markers with the assumption of joint constraint. The imposition of joint constraint has been effective in substantially reducing STA in comparison with segmental optimisation methods[228, 230, 231].

When using global optimisation, the degrees of freedom (DoF) permitted at each joint, termed mobilisers, are specified *a priori*[248]. Constraining the lower limb joints to three mobilisers (i.e. flexion/extension; ab/adduction and medial/lateral rotation) is a potential source of error, particularly at the knee, as physiological translation is ignored[250, 251]. During gait, normal physiological translation at the knee of up to 35mm has been reported[252], with smaller normal translations also reported at the hip[253] and ankle[254]. Ignoring translation can result in under-estimating positive joint work during walking[255].

Using six mobilisers reduces error due to hip joint centre mislocation compared to using three mobilisers[256]. However, permitting six mobilisers at each lower limb joint can result in over-estimation of translation at the hip and ankle[250]. Joint cartilage compression, mislocation of the instantaneous centre of rotation and STA are potential sources of error in joint translation data[255]. The number of mobilizers used is reported to have a greater effect on kinematic data than the computation method (direct or inverse) applied[250].

It is possible in Visual 3D™ to specify boundaries of joint translation using a Limited-memory Broyden–Fletcher–Goldfarb–Shanno (LBFGSB) optimiser[257]. The use of this method is thought to minimise errors from ignoring joint translation while avoiding over-estimation of translation movement. However, the effect of joint constraint boundaries on kinematic results has yet to be reported in the research literature.

Differences in kinematic results are magnified when calculating joint powers, which are the product of joint moments and joint angular velocities[258]. Joint powers are of interest to clinicians because they reflect the activity of muscle groups in producing and controlling movement[258]. Even if the effect of joint constraint boundaries on kinematics were small, the effect on joint powers could be significant and therefore clinically important. Thus, it is

of interest to know how joint constraint boundaries affect joint power calculations. A comparison was undertaken of lower limb joint powers calculated with three different Inverse Kinematic (IK) models with global optimisation: IK3 (3 mobilisers); IK6 (6 mobilisers) and IK6C (6 mobilisers with custom constraint).

4.2.1 Methods

4.2.1.1 Participants

Data for this analysis were taken from the baseline data collected as part of the larger RCT investigating the effect of a running training program on the biomechanics of running in children with CP. Forty-three children aged 9-18 were enrolled in the study. Three participants were excluded from this analysis as they did not have a flight phase. For futher details please refer to the published study presented in section 4.3.

4.2.1.2 Data processing

All data were processed using Visual 3D™ version 6 (C-Motion, Inc.). A single representative jog trial which included a flight phase was selected for each participant. Kinematic data were filtered at 18 Hz using a zero-lag 4th order Butterworth filter. Force plate data were unfiltered. The same stride was processed three times using the models described in Table 4.1. All three models used inverse kinematics with global optimisation. The IK3 model had three mobilisers at each joint, namely flexion/extension, abduction/adduction and internal/external rotation. The IK6 model had these three mobilisers with an additional three mobilisers, namely medial/lateral translation, anterior/posterior translation and superior/inferior translation, nominally constrained to 1m of movement in each direction. The IK6C model had the same six mobilisers but with custom constraints derived from the literature.

The custom constraints derived from the research literature were as follows. Femoral head translation in healthy adults has been measured at 4mm in any direction [253, 259]. Children and adolescents with CP, Gross Motor Function Classification Scale (GMFCS) Level I or II who are able to run, are at low risk of hip displacement [260, 261]. Hip joint constraint was therefore set at 5mm in any direction. There was less agreement on knee translation measures in the literature, although these were consistently greater in the anterior-posterior direction than medial-lateral direction [252, 262-264], which was reflected in the IK6 model. There was diversity in the reporting of ankle joint translations in the research

literature, including the movement assessed and the segmental relationship; talocrural, talus relative to tibia or calcaneus relative to tibia [254, 265, 266]. Ankle translations measured during walking gait [254] were chosen for incorporation into the IK6 model.

Scalar power and translation at the right hip, knee and ankle joints were calculated and then extracted for the first full right stride (initial contact to initial contact) from each jogging trial. Statistical parametric mapping (SPM), specifically a one-way repeated measures ANOVA (α=0.05) was used to compare time-normalised (1D) scalar power of the hip, knee and ankle joints.

Table 4.1: Inverse kinematic models with three, six and six constrained mobilisers

Model	POSE filter frequency	Optimiser	Segment	Weight factor	Translations	Translation Constraints (proximal end of segment with respect to proximal segment)			
IK3	6Hz	Levenberg Marquardt	Pelvis RTH RSK RFT	4 2 3 4	X Y Z 0 0 0 0 0 0 0 0 0				
IK6	6Hz	Levenberg Marquardt	Pelvis RTH RSK RFT	4 2 3 4	X Y Z X Y Z X Y Z X Y Z	None			
IK6C	6Hz	LBFGSB	Pelvis RTH RSK RFT 	4 2 3 4 	X Y Z X Y Z X Y Z X Y Z 	Hip Knee Ankle 	X Y Z X Y Z X Y Z	-0.005m (lateral) -0.005m (posterior) -0.005m (inferior) -0.015m (lateral) -0.015m (posterior) -0.02m (inferior) -0.005m (lateral) -0.006m (anterior) -0.003m (inferior)	0.005m (medial) 0.005m (anterior) 0.005m (superior) 0.01m (medial) 0.015m (anterior) 0.00m (superior) 0.003m (medial) 0.004m (anterior) 0.005m (superior)

IK6C=IK6Constrained; LBFGSB= Limited-memory Broyden-Fletcher–Goldfarb–Shanno; POSE= Position and Orientation of a Segment; RTH=Right Thigh; RSK=Right Shank; RFT=Right Foot; m=metres;

4.2.2 Results

While the IK3 model attempts to keep joint translation to nil, it did result in very small translations which were largest at the ankle with a mean of 0.017mm in the posterior direction (Figure 4.1). The IK6 model resulted in large ranges of joint translation over the gait cycle at all three joints. Using the IK6 model, anterior translation of up to 14cm was estimated at the ankle, 15cm at the knee and 17cm at the hip, and posterior translation of up to 10.6cm was estimated at the ankle, 15cm at the knee and 20cm at the hip. These estimates are clearly outside the range of physiological probability and the IK6 model was therefore excluded from further analysis. The IK6C model estimated reasonable translations over the gait cycle at all three joints, which were largest at the knee. Maximum estimated translation was 8mm anterior and 7mm posterior at the ankle, 15mm anterior and 23mm posterior at the knee, and 8mm anterior and 6mm posterior at the hip.

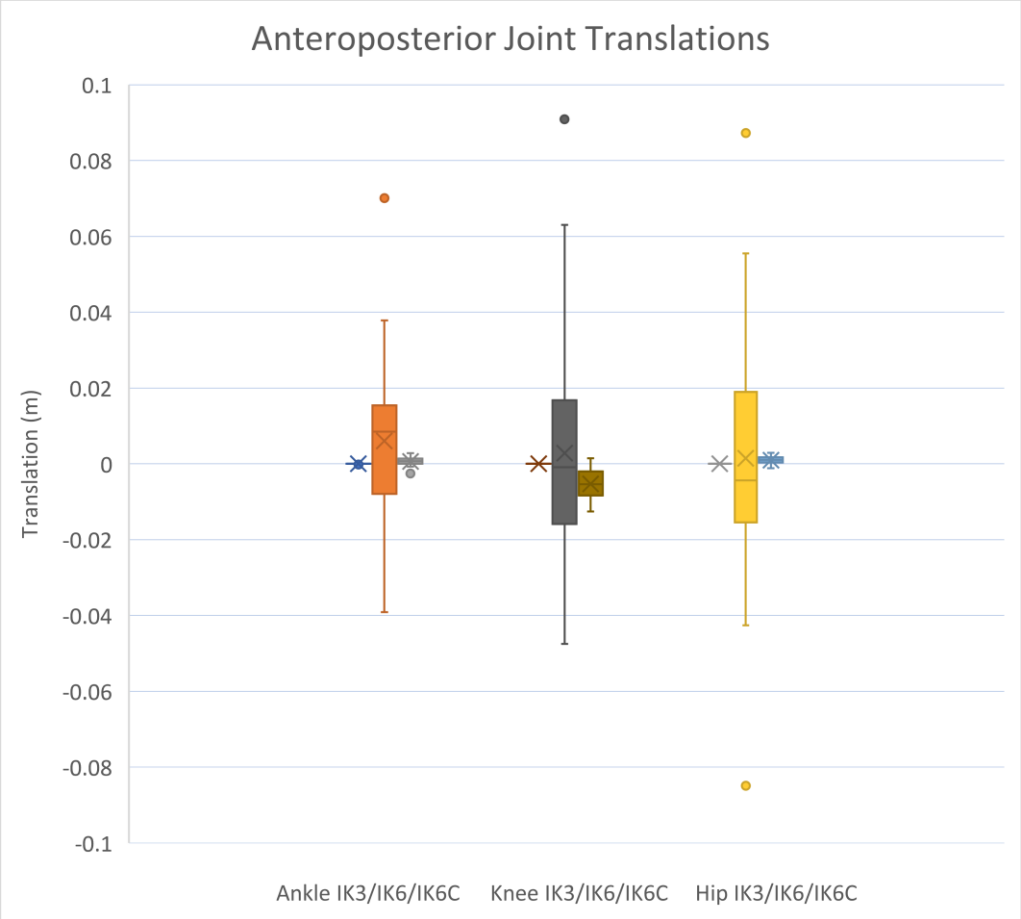


Figure 4.1: Anteroposterior joint translations using the IK3, IK6 and IK6C models: median, interquartile range and outliers

Results of the SPM analysis are shown in Figure 4.2, with time periods where significant within-subject differences occurred indicated as solid shading above the dotted line. Scalar power from IK3 and IK6C models were significantly different at all three joints, notably at push-off at the ankle and in swing at the hip. Results from a single subject are shown in Figure 4.3 as a typical example. Compared to the IK3 model, the IK6C model estimated increased power absorption at the ankle in early stance and reduced power generation in the second half of stance. At the knee, the IK6C model estimated increased power absorption followed by reduced and later power generation in mid-stance. Power absorption was reduced in early swing and increased in terminal swing. At the hip, the IK6C model estimated earlier and increased power generation in early stance, increased and later power absorption in terminal stance, later power generation in early swing and increased power generation in late swing (Table 4.2).

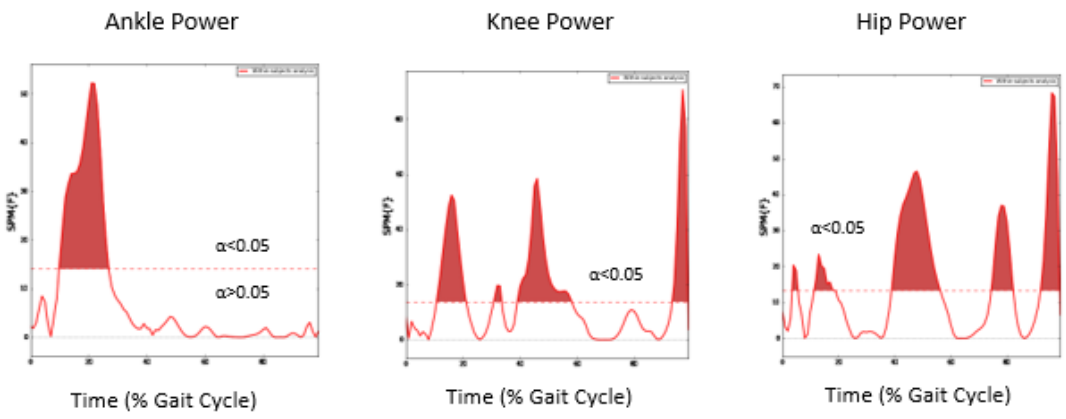


Figure 4.2: SPM results (one-way repeated measures ANOVA). The dotted line indicates the critical random field theory threshold

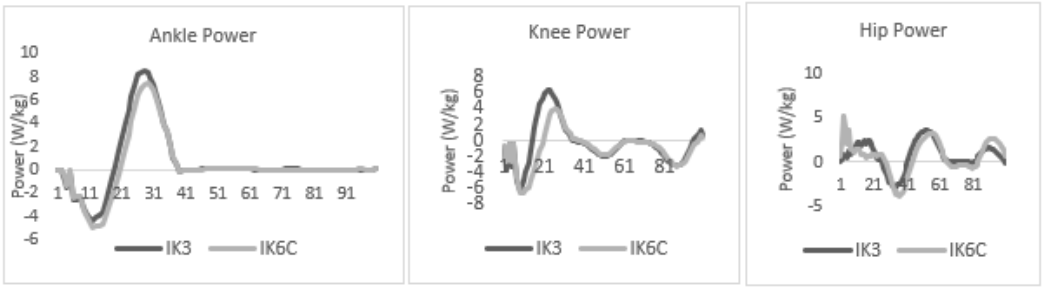


Figure 4.3: Comparison of ankle, knee and hip power from a single subject

Table 4.2: Joint powers in W/kg at specified points of the time-normalised gait cycle

Measure	IK3	IK6C
Mean Ankle Power 12%	-4.23 ± 1.84	-5.47 ± 2.67*
Mean Ankle Power 25%	5.7 ± 3.25	4.58 ± 3.14*
Mean Knee Power 15%	-0.73 ± 2.78	-3.29 ± 2.6*
Mean Knee Power 32%	0.44 ± 0.93	0.68 ± 0.98
Mean Knee Power 50%	-1.42 ± 1.01	-1.13 ± 0.82*
Mean Knee Power 95%	-1.29 ± 1.41	-1.66 ± 1.23*
Mean Hip Power 5%	1.03 ± 4.21	2.56 ± 4.97*
Mean Hip Power 15%	1.05 ± 1.57	0.26 ± 1.50*
Mean Hip Power 50%	2.01 ± 1.32	0.91 ± 0.77*
Mean Hip Power 95%	2.07 ± 1.75	2.83 ± 1.66*

W/kg = Watts per kilogram; * significantly different to IK3 model

4.2.3 Discussion

The main finding of this study was that specified joint constraint boundaries significantly affected the estimation of joint powers at the ankle, knee and hip across the jogging gait cycle. These differences were clinically significant for the population of children with CP and could result in different clinical interpretations. The IK6C model resulted in both a downward shift and a slight phase shift in the ankle and knee power curves compared to the IK3 model, with greater absorption and less generation in the stance phase. These results are in agreement with a previous study which found significantly less work at the ankle with a 6DOF model compared to a 3DOF model[267]. The magnitude of the shift was equal to differences which have been considered clinically significant in the literature[268-271].

The model applied (IK3 or IK6C) had a more complex effect on hip power than ankle or knee power. The IK6C model resulted in greater power generation in early stance and terminal swing and increased power absorption in late stance. There was a phase shift in early swing, with later power generation using the IK6C model compared to the IK3 model. In the first half of stance the hip extensors generate power to move the trunk over the supporting foot and in late swing extend the hip in preparation for initial contact [56]. From toe-off to peak hip flexion in mid-swing, the hip flexors use the stretch-shortening mechanism to generate power to flex the hip [56, 62]. It would therefore seem that the IK6C model gives comparatively greater estimates of the power generated by the hip extensors and

comparatively later estimates of power generated by the hip flexors when compared to the IK3 model. This is important for a comprehensive understanding of power generation patterns during running and the effect of pathology on power generation, including weaknesses and compensations [84, 267].

It has been reported that biomechanical work calculated during walking using a 3DOF model fails to account for over 30% of the positive energy change during push-off[255, 267]. Using a 6DOF model it was possible to fully account for positive work during walking and provide a more comprehensive estimate of how work is distributed[255]. The results of this study suggest that if the same relationship to power was assumed in running, that the "missing" power may largely be hip extensor power. The calculation of work over the running gait cycle is an avenue for further investigation, to determine whether the IK6C model would fully account for biomechanical work during running. If this proved to be the case, it may be that the hip extensors play a more important role in power generation during running than was previously thought.

It was concluded that the number of DoF used in a biomechanical model significantly affected lower limb joint power calculations, but this analysis could not identify the source of the difference, whether joint angular velocity or joint moments. Further analysis was therefore undertaken, which is presented in the following section.

4.3 The effect of joint translation constraint on within-participant variability of kinematics and kinetics during running in cerebral palsy

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The effect of joint translation constraint on within-participant variability of kinematics and kinetics during running in cerebral palsy

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ABSTRACT

Background: Biomechanical data in cerebral palsy are inherently variable but no optimal model of translational joint constraint has been identified. The primary aim of this study was to determine which model of translational joint constraint resulted in the lowest within-participant variability of lower limb joint angles and moments. The secondary aim was to determine which model best distinguished known functional groups in Cerebral Palsy.
Methods: Three models (three degrees of freedom, six degrees of freedom and six degrees of freedom with specified joint translation constraint) were applied to data from running trials of 40 children with cerebral palsy.
Findings: Joint angle standard deviations were largest using the six degrees of freedom model and smallest using the constrained six degrees of freedom model ($p < 0.050$). For all joints in all planes of motion, joint moment standard deviations were largest using the six degrees of freedom model and smallest using the constrained six degrees of freedom model; standard deviations using the constrained model were smaller than the three degrees of freedom model by 10–30% of moment magnitude (0.01–0.03 Nm/kg; $p < 0.001$). The six degrees of freedom models distinguished functional subgroups with larger effect size than the three degrees of freedom model only for hip power generation in swing.
Interpretation: A model with specified joint constraint minimized within-participant variability during running and was useful for detecting differences in functional capacity in cerebral palsy.

1. Introduction

Cerebral palsy is a term describing permanent but not unchanging disorders of movement and posture which result from an insult to the developing brain (Rosenbaum et al., 2007). Children with cerebral palsy are classified into five groups according to the Gross Motor Function Classification System (GMFCS) (Palisano et al., 2007). Children in GMFCS Level I are able to run, albeit with some limitations, while about half the children in GMFCS level II are able to run, with more difficulty than those children in GMFCS level I (Böhm et al., 2018). Three-dimensional gait analysis is often undertaken to describe the effect of neuromuscular impairments or intervention on the gait of people with cerebral palsy (Böhm et al., 2018).

Kinematic and kinetic measurements generated by three dimensional gait analysis are inherently variable (Chia and Sangeux, 2017). Intrinsic variability is biological and represents flexibility of the neuromotor system (Barrett et al., 2008). Intrinsic variability is higher in the cerebral palsy population compared to the typically developing population due to neuromotor impairments (Klejman et al., 2010; Steinwender et al., 2000). Extrinsic variability is introduced by the data collection and processing workflow and includes sources of error. The most significant sources of error are movement of skin markers with respect to the bone, termed soft tissue artefact, and the assumptions of the biomechanical model (Chia and Sangeux, 2017). Extrinsic variability in a data set may reduce statistical power which can result in a failure to detect true differences or changes (Sullivan and Feinn, 2012).

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Table 1
Marker set.

Segment	Abbreviation	Placement/full name	Anatomical	Tracking
Foot	RMT1, LMT1	Right/left 1st metatarsal head	✓	✓
	RMT5, LMT5	Right/left 5th metatarsal head	✓	✓
	RCAL, LCAL	Right/left calcaneus (middle of posterior aspect)	✓	✓
Shank/leg	RMMAL, LMMAL	Distal end of tibia, medial malleolus	✓	
	RLMAL, LLMAL	Distal end of fibula, lateral malleolus	✓	
	RTB1, LTB1	Right/left tibia: Long bar runs along tibia axis and the short bar wraps laterally.		✓
	RTB2, LTB2			
	RTB3, LTB3			
Thigh	RMFC, LMFC	Right/left medial femoral condyle	✓	
	RLFC, LLFC	Right/left lateral femoral condyle	✓	
	RTH1, LTH1	Right/left thigh: long bar runs along Iliotibial band and short bar wraps anteriorly		✓
	RTH2, LTH2			
	RTH3, LTH3			
Pelvis	RASI, LASI	Right/left anterior superior iliac spine	✓	✓
	RPSI, LPSI	Right/left posterior superior iliac spine	✓	✓

This is a challenge for research in cerebral palsy, where sample sizes are often small, and the population is heterogeneous.

Inverse kinematics (IK) optimization is a technique proposed to reduce extrinsic variability by utilising modelled joint constraints (Leardini et al., 2005; Lu and O'connor, 1999), in which the position and orientation of the segment is calculated by minimizing the sum of squared displacements between the measured coordinates of all markers in the model and their modelled coordinates (Leardini et al., 2017). This is done according to the degrees of freedom (DoF) specified at each joint, termed joint constraints (Robinson et al., 2014). Joint constraint boundaries can be specified to mimic physiological joint limitations and can be derived directly from in-vivo methods or indirectly using data from published literature (Leardini et al., 2017). In a research study with many participants and many time points, it may not be considered feasible to employ individual biomechanical models. A joint constraint model which can be applied to all participants at all time points may represent a significant efficiency, provided accuracy is maintained.

The magnitude of specified joint constraint impacts the variability of kinematic and kinetic gait measures (Kainz et al., 2016). For example, Potvin and colleagues (Potvin et al., 2017) reported a smaller standard deviation (SD) for knee rotation angle in the transverse plane during walking when knee joint constraints were informed from published in-vivo data, compared to a knee model with no constraints. A reduction in within-participant variation due to modelling implies a reduction in random error due to measurement, with the assumption that biological variability is maintained (Hopkins, 2000). The effect of specifying joint constraint boundaries at all modelled joints on the within-subject variability of kinematic and kinetic gait measures during running in people with cerebral palsy is unknown.

In intervention-based studies, such as in a Randomized Controlled Trial, less within-participant variability is desirable. For a given difference in mean score of a biomechanical variable, smaller within-participant variability will result in greater effect size (Sullivan and Feinn, 2012). Consequently, a reduction in within-participant variability should increase the statistical power of a study and minimize the chance of falsely accepting the null hypothesis (Hopkins, 2000; Sullivan and Feinn, 2012).

The primary aim of this study was to determine which model of translational joint constraint: Model IK3 (3DoF); Model IK6 (6DoF) or Model IK6Constrained (specified joint translation constraint) resulted in the lowest within-participant variability of lower limb joint angles and moments. A biomechanical model which reduces within participant SD will be important for research and clinical use, as it represents a greater opportunity to detect an established known difference between different patient categories. The secondary aim of this study was to undertake a known groups analysis (Davidson, 2014) to identify which model/s were most effective at distinguishing known functional groups GMFCS level I and level II (Verschuren et al., 2010). By calculating effect sizes for the different derived variables between children who are classified as GMFCS level I and those who are classified as GMFCS level II using each of the three models, we aim to determine whether any of the models separate the GMFCS levels, or 'known groups' with more statistical power than the others. If one of the models has larger effect sizes than the others, using this model in an intervention study may help to avoid a Type II error.

2. Methods

2.1. Participants

The present study represents the results of a sub-study from a larger project investigating the effects of a physical training program in children with cerebral palsy (Gibson et al., 2017). Data for the present study came from the three-dimensional running biomechanics collected at baseline prior to intervention. Forty-three children with cerebral palsy (aged 9–18 years) with GMFCS level I-II and who were able to run independently were recruited (Gibson et al., 2017). Three participants were excluded from the present study due to the absence of a flight phase during running. A sample size of 40 provided us with 87% power to detect an effect size of 0.5 at an alpha value of 0.05 (G*Power v 3.1.9.2) (Faul et al., 2009). The study was approved by the Ethics Committees of Princess Margaret Hospital for Children, Perth, Western Australia (201405SEP) and Curtin University, Perth, Western Australia (HR 219/2014).

2.2. Data collection

One experienced physiotherapist applied a modified Cleveland Clinic Foundation marker protocol (Sutherland, 2002) (Table 1). Participants wore their usual sport shoes and foot markers were placed on the shoes over the relevant landmarks. Participants were not allowed to use orthoses extending above the malleoli. Participants were asked to run at three speeds, 1) jog "like a warm-up", 2) run "faster but not your fastest", and 3) "sprint like you are in a race", along a 30 m runway. At least five trials at each speed were collected unless the participant was too fatigued to continue. A two-minute sitting break was permitted between speeds if required. Kinematic data were recorded by an 18-camera motion capture system at 250 Hz (Vicon T-series, Oxford Metrics, UK). Synchronized ground reaction forces were collected at 1000 Hz using three in-ground force platforms in series (AMTI, Watertown, MA, USA). Marker trajectories were labelled and filled using Vicon Nexus 2.5 (Vicon Motion Systems, Oxford, UK).

2.3. Data processing

All data were processed using Visual 3D™ version 6 (C-Motion, Inc.). Kinematic and force plate data were filtered at 18 Hz using a zero-lag 4th order low-pass Butterworth filter following residual analysis (Yu et al., 1999). Three IK models were created from the same conditioned data, the only difference between models being the magnitude of the joint constraints applied (Table 2). All trials were processed using each of the three models described in Table 2. Model IK3 allowed 3DoF at each joint; flexion/extension, abduction/adduction and internal/external rotation with no translation permitted. Model IK6 allowed 6DoF without joint constraints; flexion/extension, abduction/adduction, internal/external rotation, medial/lateral translation, anterior/posterior translation and inferior/superior translation. Model IK6Constrained also allowed 6DoF but with specified joint translation constraints derived from the literature (Section 2.3.1).

2.3.1. Joint constraint parameters

Femoral head translation in healthy adults has been reported to be < 4 mm (Gilles et al., 2009). Children and adolescents with cerebral palsy GMFCS level I or II who are able to run are at low risk of hip displacement (Kentish et al., 2011; Robin et al., 2009) but may have more hip joint translation than typically developing children (Kentish et al., 2011; Robin et al., 2009). Hence, 5 mm of hip joint translation was permitted in model IK6Constrained (Table 2). There is less agreement in the research literature with respect to knee joint translation amplitude, but knee joint translation is consistently reported as greater in the anterior-posterior direction than medial-lateral direction in healthy adults (Sheehan et al., 2008), which was reflected in Model IK6Constrained (Table 2). There was diversity in reporting of ankle joint translations in healthy adults, including both the activity (walking versus seated ankle plantarflexion/dorsifleixon) and the joints assessed (talocrural, talus relative to tibia or calcaneus relative to tibia) (de Asla et al., 2006; Imai et al., 2009; Sheehan et al., 2007). Ankle translations measured during walking (de Asla et al., 2006) were chosen for incorporation into Model IK6Constrained (Table 2) as data for running have not been reported.

2.3.2. Kinematics and kinetics

For each model, pelvic segment angle, joint angles for hip, knee and ankle and net internal joint moments for the hip, knee and ankle were calculated for each trial in three planes (sagittal, frontal and transverse) and time normalized to 101 points from initial contact to initial contact of the same limb. For ankle, knee and hip joint angles an X-Y-Z (flexion-extension, abduction-adduction, internal-external rotation) cardan sequence was used, while for the pelvic segment angle a Z-Y-X sequence was used (Baker, 2001; Cole et al., 1993). Inertial and geometric properties of the segments were based on previously published models (Dempster, 1955; Hanavan Jr, 1964). A regression equation was used to calculate hip joint centres (Bell et al., 1989). Knee and ankle (talocrural) joint centres were calculated using the proximal joint centre and the midpoint between the medial and lateral femoral condyles, and malleoli respectively. Joint centres were located on the proximal end of the distal segment. The inferior-superior axis of each segment coordinate system lay along a vector connecting the joint centres. Each segment coordinate system was created using the anatomical plane and defined joint centres (O'Connor and Bottum, 2009; Palmieri-Smith et al., 2009).

Joint moments were resolved in the coordinate system of the proximal segment (Williams et al., 2004). Force plate threshold was set at 20 N with foot on and foot off detected automatically. Gait events occurring off the force plates were automatically detected based on the axial and anteroposterior position of the proximal end of the foot for initial contact and the distal end of the foot for toe-off (Stanhope et al., 1990).

2.3.3. Within-participant, within-session variability

Data were grouped by speed (jog, run, sprint) and the between-trial standard deviations (SDs) calculated for each data point for each participant at each of the three running speeds for each of the three models (Kainz et al., 2017). The average SD across the gait cycle for each joint angle and moment was retained as a dependent variable (n = 42 for each model at each speed for each subject).

2.4. Statistical analysis

2.4.1. Within-participant variability

Natural log transformations were performed on joint angle SDs to correct right skewedness. Joint moment data were separated into stance and swing phases as these data were quite distinct. Two-step transformations to normality using the inverse distribution function were performed on stance and swing joint moment SDs to correct right skewedness. The transformed data were then analyzed in Statistical Analysis Software version 9.4 (SAS Institute Inc., Cary, NC, USA) using linear mixed models with fixed effects joint, model and gait phase (stance or swing) and random effects subject and speed. Interactions between fixed effects were excluded when not significant with alpha set at 0.05. Covariances were modelled as compound symmetry.

2.4.2. Known groups analysis

Known groups analysis was undertaken with participants classified as GMFCS level I or GMFCS level II. Derived variables considered pertinent to running were included; peak ankle, knee and hip power absorption in stance phase (A1, K1 and H2 respectively), peak ankle, knee and hip power generation in stance phase (A2, K2 and H1 respectively) and peak hip power generation in swing phase (H3). Spatiotemporal variables were not included as these were not expected to change depending on the IK model. Hedges g was calculated for all variables for the three models. Hedge's g is the most appropriate measure of effect size where sample size is < 20 and the two sample sizes are different (Lakens, 2013). Hedge's g was thus considered most appropriate for this analysis of GMFCS level I (n = 25) and GMFCS level II (n = 15).

3. Results

3.1. Participants

Forty-three participants were recruited to the study. Three were excluded from the analysis due to the absence of a flight phase. Participants were aged 12.69 years (SD = 2.7 years); 25 males and 15 females; 25 GMFCS level I and 15 GMFCS level II; 21 bilateral cerebral palsy and 19 unilateral cerebral palsy. Two participants completed jog trials only. Three participants completed jog and run trials only. Of those who completed sprint trials, 13 completed three sprint trials, 14 completed four sprint trials and 8 completed five or more sprint trials.

3.2. Model performance

The IK6 and IK6C joint translations are reported in Table 2. The unconstrained 6DoF model resulted in larger joint translations than the constrained 6DoF model, especially at the hip.

3.3. Joint angles

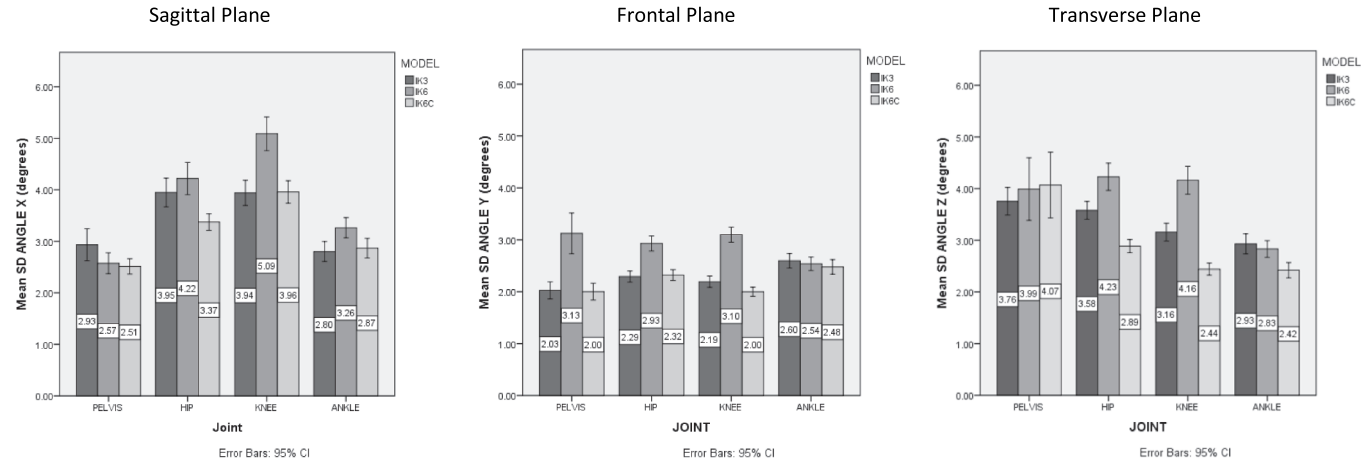
In general, the IK6 model resulted in the largest joint angle SDs and the IK6Constrained model resulted in the smallest joint angle SDs. The model applied only influenced pelvis segment angle in the frontal plane where both IK3 SDs and IK6Constrained SDs were 1.1° smaller than IK6 SDs ($p < 0.001$ and $p < 0.001$) (Figs. 1 and 2). At the hip, IK6Constrained SDs were 0.6° smaller than IK3 SDs in the sagittal plane ($p = 0.003$) and 0.7° smaller in the transverse plane ($p < 0.001$), but not different in the frontal plane ($p = 0.425$). IK3 SDs were smaller than

Table 2
IK models.

Model	POSE filter frequency	Optimizer	Segment	Weight factor	Translations	Translation constraints (proximal end of distal segment with respect to distal end of proximal segment)			Model performance (translation mean (SD))			
IK3	6 Hz	Levenberg Marquardt: default in Visual 3D™	Pelvis	4	X Y Z							
			RTH	2	0 0 0							
			RSK	3	0 0 0							
			RFT	4	0 0 0							
IK6	6 Hz	Levenberg Marquardt	Pelvis	4	X Y Z	None						
			RTH	2	X Y Z				Hip	X	−17 mm (17 mm)	20 mm (22 mm)
										Y	−24 mm (32 mm)	21 mm (15 mm)
										Z	−20 mm (21 mm)	17 mm (12 mm)
			RSK	3	X Y Z				Knee	X	−11 mm (17 mm)	9 mm (12 mm)
										Y	−7 mm (13 mm)	15 mm (21 mm)
										Z	−31 mm (15 mm)	1 mm (14 mm)
			RFT	4	X Y Z				Ankle	X	−7 mm (8 mm)	4 mm (8 mm)
										Y	−10 mm (9 mm)	4 mm (9 mm)
										Z	−11 mm (9 mm)	0 mm (5 mm)
IK6C	6 Hz	LBFGSB: uses an estimation to the inverse Hessian matrix for optimizing variables subject to simple bounds[a]. It is more time consuming than the Levenberg Marquardt optimizer.	Pelvis	4	X Y Z							
			RTH	2	X Y Z	Hip	X	−5 mm (lateral)	5 mm (medial)	Hip	X	−5 mm (1 mm)	5 mm (1 mm)
							Y	−5 mm (posterior)	5 mm (anterior)		Y	−5 mm (1 mm)	6 mm (1 mm)
							Z	−5 mm (inferior)	5 mm (superior)		Z	−5 mm (2 mm)	6 mm (2 mm)
			RSK	3	X Y Z	Knee	X	−15 mm (lateral)	10 mm (medial)	Knee	X	−10 mm (4 mm)	10 mm (4 mm)
							Y	−15 mm (posterior)	15 mm (anterior)		Y	−15 mm (2 mm)	12 mm (5 mm)
							Z	−20 mm (inferior)	0 mm (superior)		Z	−20 mm (7 mm)	−4 mm (5 mm)
			RFT	4	X Y Z	Ankle	X	−5 mm (lateral)	3 mm (medial)	Ankle	X	−4 mm (1 mm)	4 mm (1 mm)
							Y	−6 mm (anterior)	4 mm (anterior)		Y	−6 mm (1 mm)	4 mm (1 mm)
							Z	−3 mm (inferior)	5 mm (superior)		Z	−2 mm (6 mm)	2 mm (2 mm)

Hz = Hertz; IK3 = three degree of freedom inverse kinematic model; IK6 = six degree of freedom inverse kinematic model; IK6C = six degree of freedom inverse kinematic model with specified joint constraint boundaries; LBFGSB = limited-memory Broyden-Fletcher–Goldfarb–Shanno; mm = millimeters; POSE = position and orientation of a segment; RTH = right thigh; RSK = right shank; RFT = right foot; SD = standard deviation.

[a] Zhu C, Byrd RH, Lu P, Nocedal J. Algorithm 778: L-BFGS-B: Fortran subroutines for large-scale bound-constrained optimization. ACM Trans Math Softw. 1997;23:550–60.



SD=standard deviation, CI=confidence interval, IK6C=IK6Constrained model

Fig. 1. Mean joint angle SDs.

IK6 SDs in all planes (sagittal $p = 0.026$, frontal $p < 0.001$ and transverse $p < 0.001$) (Fig. 1). At the knee, IK6Constrained SDs were 0.2° smaller than IK3 SDs in the frontal ($p = 0.038$) transverse planes ($p < 0.001$), but not different in the sagittal plane ($p = 0.393$). IK3 SDs were smaller than IK6 SDs in all planes (sagittal $p < 0.001$, frontal $p < 0.001$ and transverse $p < 0.001$) (Fig. 1). At the ankle, IK6Constrained SDs were 0.1° smaller than IK3 SDs in the frontal plane ($p = 0.038$) and 0.5° smaller in the transverse plane ($p < 0.001$), but not different in the sagittal plane ($p = 0.216$). IK3 SDs were smaller than IK6 SDs in the sagittal plane ($p < 0.001$) but not different in the transverse plane ($p = 0.725$). In the frontal plane there was no difference between IK6Constrained and IK6 ($p = 0.135$) or IK6 and IK3 ($p = 0.558$) (Figs. 1 and 2).

3.4. Joint moments

For all joints in all planes of motion, IK6Constrained SDs were smaller than IK3 by 10–30% (0.01–0.03 Nm/kg ($p < 0.001$ for each joint in each plane)) and IK3 SDs smaller than IK6 by 22–33% (0.03–0.13 Nm/kg ($p < 0.001$ for each joint in each plane)) (Figs. 2 and 3).

3.5. Speed

Speed (jog/run/sprint) did not have a significant effect in the linear mixed models.

3.6. Hedges g effect sizes

Large effect sizes ($g > 0.8$) were found for A1, A2 and H2 for all IK models. Medium effect sizes ($0.5 > g < 0.8$) were found for K2 and H1 for all IK models. Small effect sizes ($0.2 > g < 0.5$) were found for K1 for all IK models. For H3, effect size was small for model IK3 (0.48) and medium for IK6 (0.58) and IK6Constrained (0.72) (Fig. 4).

4. Discussion

The main finding of this study was that specified joint constraint boundaries resulted in small but significant reductions in within-participant variability of estimated joint angles and moments at the ankle, knee and hip compared to the IK3 and IK6 models. The second main finding was that specified joint constraint boundaries resulted in a larger effect size when separating GMFCS levels I and II for hip power generation in swing, compared to the other two models.

4.1. Kinematic and kinetic variability

Extrinsic variability is introduced to a data set through the data collection and processing workflow and includes sources of error (Chia and Sangeux, 2017). Large amounts of extrinsic variability reduce statistical power and make it more difficult to identify true differences between groups, or between time points in a given population (Hopkins, 2000). In this study, only the joint constraint boundaries were manipulated, so any reduction in variability can be confidently attributed to a reduction in extrinsic variability, which is desirable. Reducing extrinsic variability increases the likelihood of identifying true differences between groups or changes over time due to natural progression or intervention.

Overall, the IK6 model resulted in the greatest within-participant variability in both kinematics and kinetics. In a six DoF joint model, soft tissue artefact can result in overestimation of joint translation including apparent joint dislocations (Kainz et al., 2016; Leardini et al., 2005; Ojeda et al., 2016), which may explain the greater extrinsic variability. Compared to the IK3 model, the IK6Constrained model resulted in smaller within-participant variability for all joint moments, for joint angles in the transverse plane and knee joint angle in the frontal plane. Constraining a joint to three DoF can potentially mitigate soft tissue artefact by removing joint centre translation (Leardini et al., 2017), however assuming adjacent segments are pinned together also introduces error (Schmitz et al., 2016). Utilising joint constraint boundaries to allow restricted joint centre translation may explain why the IK6Constrained model resulted in lower variability than both the IK3 model and the IK6 model. This explanation is consistent with the findings of Potvin and colleagues (Potvin et al., 2017), who reported a smaller range of knee rotation using five DoF with bone-pin informed constraint compared to using five DoF without informed joint constraint boundaries in walking in healthy adults. The finding is important because both running gait and cerebral palsy are associated with high intrinsic variability (Estep et al., 2016;Klejman et al., 2010; Steinwender et al., 2000) and it has traditionally been difficult to recruit large numbers to studies in this population due to the heterogeneity of their clinical presentation. Minimizing extrinsic variability is therefore important to improve statistical power in research studies in this population.

At the ankle, our IK3 joint angle SDs were similar or smaller in the sagittal and transverse planes to those reported using a three DoF model in healthy adult walking (Charlton et al., 2004; Schmitz et al., 2016), but larger in the frontal plane than those reported in healthy adults walking (Charlton et al., 2004) or running (Hamacher et al., 2016) and larger than those reported in chronically unstable ankles while running

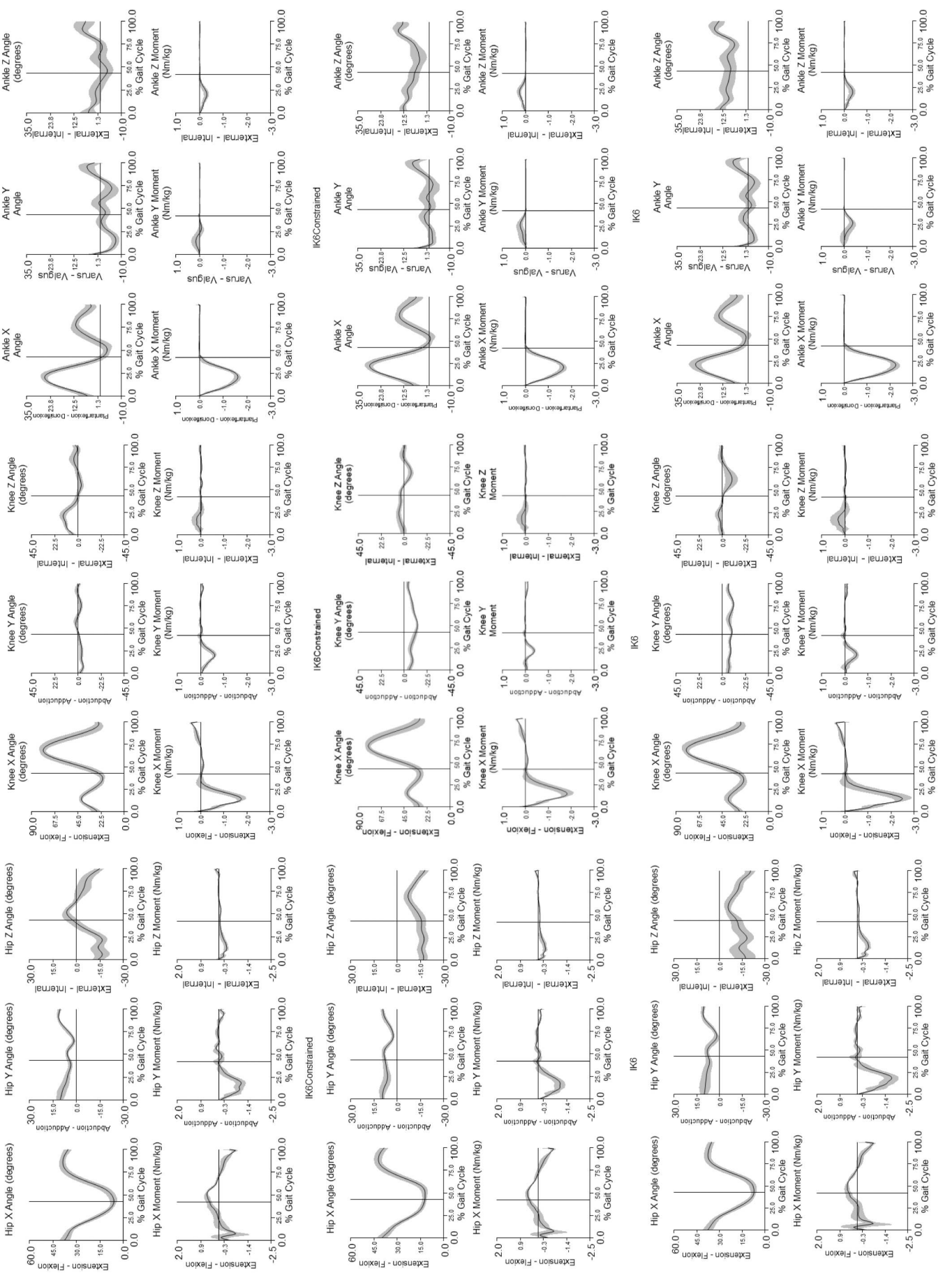
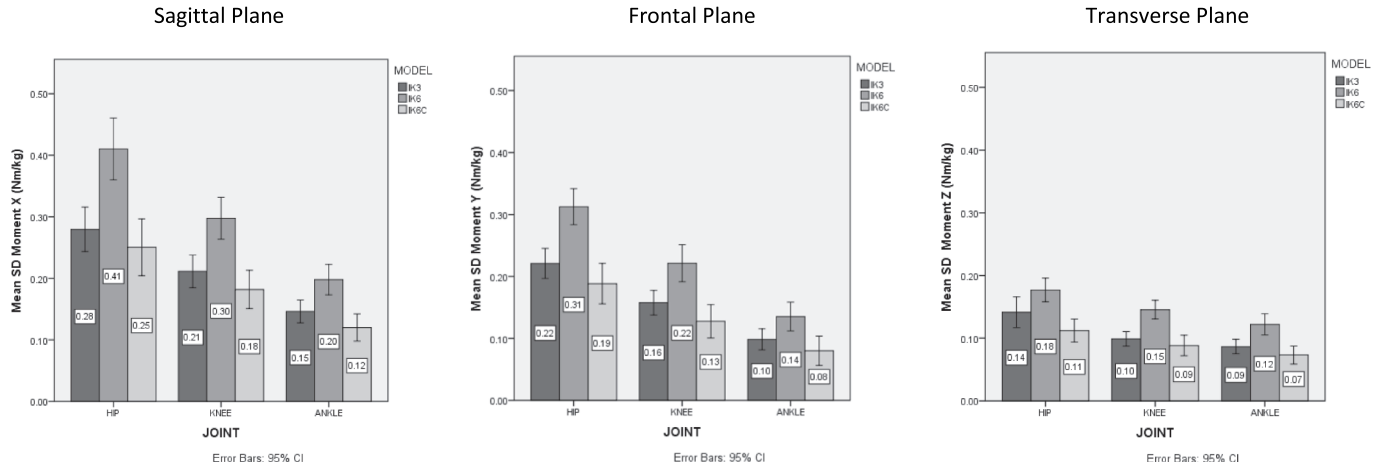


Fig. 2. Comparison of hip, knee and ankle angles and moments from three models within a subject; mean (solid line) and standard deviation (shaded).



SD=Standard deviation, Nm/kg=Newton metres per kilogram, CI=confidence interval, IK6C=IK6Constrained model

Fig. 3. Mean joint moment SDs.

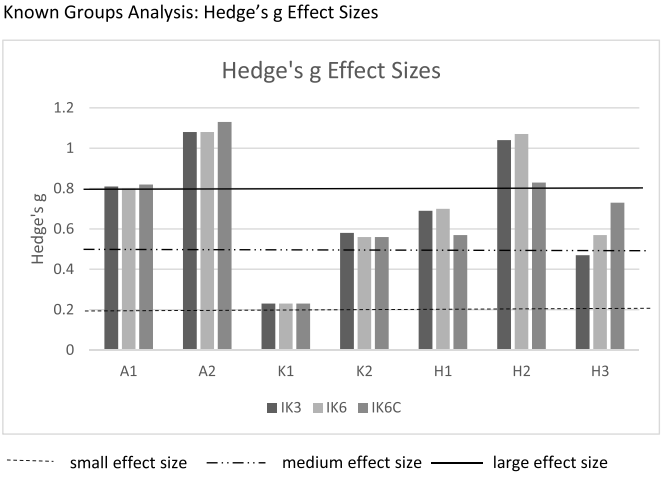


Fig. 4. Known groups analysis: Hedge's g effect sizes.

(Hamacher et al., 2016). As the ankle joint complex consists of both the talo-crural joint and the subtalar joint, modelling it as a single joint is less realistic and therefore increases error in estimation of the position and orientation of the segment (Leardini et al., 2017). This may be magnified in children with cerebral palsy in whom ankle malalignment, instability and spasticity may all increase multi-planar movements at the ankle complex (Davids, 2010). Our medio-lateral ankle joint translation boundaries were based on healthy adult data as this was the most relevant data available. These boundaries may have been too restrictive, resulting in increased error.

Our IK6Constrained model resulted in joint moment SDs 0.02–0.03 Nm/kg smaller than the IK3 model, which equates to a change of 10–30% of moment magnitude. Allowing a constrained amount of joint translation may permit the joint centre to be more accurately located during kinetic calculations, thereby reducing extrinsic variability. Joint moment SDs were larger than previously published studies of walking in healthy participants using three DoF joint models (Sangeux et al., 2016; Wong et al., 2010) but this is not surprising given joint moments are larger and more variable in running compared to walking (Estep et al., 2016; Novacheck, 1998), and more variable in children with cerebral palsy than typically developing children (Klejman et al., 2010; Steinwender et al., 2000). Potvin and colleagues (Potvin et al., 2017) reported a smaller effect of joint model DoF on joint moments compared to joint angles in walking, whereas the present study has found a significant reduction in all joint moment SDs using the IK6Constrained model. This may be because the IK6Constrained model incorporates joint constraint boundaries at all three lower limb joints, whereas Potvin and colleagues only specified joint constraint boundaries at the knee (Potvin et al., 2017). The effect of reducing error by using the IK6Constrained model may also have a magnified effect on force calculations due to the greater forces associated with running compared with walking (Smale et al., 2017).

From a clinical perspective, the effect of specified joint translation constraint on kinetics is perhaps more meaningful than the effect on kinematics (McGinley et al., 2009). For example, ankle push-off plantarflexor moment in this cohort ranges from 1 Nm/Kg during jogging to 3.5 Nm/Kg during sprinting. A reduction in SD of 0.1 Nm/kg in ankle push-off therefore equates to up to 10% reduction during a critical phase for forward propulsion. As the magnitude of clinically significant difference is yet to be defined, it is important to select the most sensitive methods available in both the research and clinical settings (Allison and Fukushima, 2003).

4.2. Effect sizes

In the known groups analysis all three models had medium-to-large effect sizes when discriminating between GMFCS level I and II for peak power absorption and generation in stance phase, except for power absorption at the knee, which had a small effect size for all three models. The notable exception was for hip power generation in swing phase, where the IK3 model had a small effect size and both IK6 and IK6Constrained models had a medium effect size. This may mean that for research questions relating to the swing phase of running, six DoF models provide a larger effect size in a group-by-time interaction. Therefore, the number of participants required to be recruited will be less using a six DoF model. During swing phase of running the hip is moved rapidly through a large arc of movement (Novacheck, 1998) and thus is subject to high levels of soft tissue artefact (Peters et al., 2010). The application of joint constraints is one potential method to reduce the effect of soft tissue artefact but is dependent on the joint constraints specified (Leardini et al., 2017; Potvin et al., 2017). Our findings suggest that applying joint translation constraints appropriate for children with cerebral palsy can reduce the effect of measurement error on skeletal movement. The ability to generalise these findings across different diagnostic groups mediated by the phase of running may warrant further research.

4.3. Limitations

The main limitation of this study is that we did not have a gold standard with which to evaluate the accuracy of the three models, thus the mechanism by which joint translation constraint reduces within-participant variability cannot be determined. However, using bone-pins in this population is invasive and difficult to justify. Calculation of marker residuals and knee joint kinematic cross-talk would add to our understanding of the accuracy of a 6DoF model with specified joint constraints but was beyond the scope of this paper. This field remains an avenue for further research.

The joint constraint boundaries used in the present study were extracted from the research literature, rather than subject-specific in-vivo measures. The boundaries were applied to all subjects, as this may be preferable in a study with many participants and many time points. In clinical gait analysis when the individual is the focus, joint constraint boundaries which consider individual variations in bony geometry, ligamentous laxity and muscle length may be preferred (Lenaerts et al., 2008; Smale et al., 2017). However, there will be situations where time or resources exclude the possibility of individual modelling. In these circumstances, a 6DoF model with prescribed joint boundaries may be considered.

5. Conclusion

The application of specified joint constraint boundaries to an IK model reduces within-participant variability of kinetic and kinematic data during running in children and adolescents with cerebral palsy and results in greater sensitivity in the detection of between-group differences, particularly in the swing phase of running.

Conflict of interest statement

The authors declare no conflict of interest.

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Chapter 5 Propulsion strategy in running in children and adolescents with cerebral palsy

5.1 Preface

This chapter describes the pattern of power generation in children and adolescents with CP during jogging, running and sprinting, prior to intervention. The description of power generation for forward propulsion in children and adolescents with CP is important because it reflects running strategy. The power generation strategy employed by a runner has consequences for mechanical efficiency[68, 240]. There is relatively little information in the research literature regarding power generation during running in children and adolescents with CP, as was identified in the systematic review presented in Chapter 2. Therefore, a cross-sectional study was conducted to describe patterns of power generation in children with CP and in typically developing children, with a focus on ankle power generation at push-off (A2) and hip power generation during swing (H3). This chapter reports the outcomes of that analysis.

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Propulsion strategy in running in children and adolescents with cerebral palsy

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ABSTRACT

Background: Running is a fundamental movement skill important for participation in physical activity. Children with cerebral palsy (CP) who are classified at Gross Motor Function Classification Scale (GMFCS) level I and II are able to run but may be limited by neuromuscular impairments.
Research question: To describe the propulsion strategy (PS) during running of children and adolescents with CP.
Methods: This cross-sectional study used kinematic and kinetic data collected during running from 40 children and adolescents with unilateral or bilateral CP and 21 typically developing (TD) children. Maximum speed, peak ankle power generation (A2), peak hip flexor power generation in swing (H3) and PS (PS = A2/(A2 + H3)) were calculated. Linear mixed models were developed to analyze differences between groups.
Results: Maximum speed, A2 and PS were significantly less in children with CP GMFCS level I than in TD children and significantly less in children in GMFCS level II than level I. For children with CP, A2 and PS were significantly smaller in affected legs than non-affected legs. In affected legs, H3 was significantly larger in children in GMFCS level II than GMFCS level I but not different between TD children and children in GFMCS level II.
Significance: The contribution of ankle plantarflexor power to forward propulsion in running is reduced in young people with CP and is related to GMFCS level. This deficit appears to be compensated in part by increased hip flexor power generation but limits maximum sprinting speed.

1. Introduction

Running is a fundamental skill developed in childhood which is necessary for participation in many recreational and sporting activities. The ability to run requires each leg to alternately propel the body into a flight phase [1]. People with CP, GMFCS level I or II, usually develop the ability to run, although they may lack speed or coordination [2]. Power for forward progression in running is provided during ground contact, primarily by the ankle plantarflexors at push-off, referred to as A2 [3]. The hip extensors also generate power in early stance, primarily to maintain a stable trunk but also to move the body over the supporting leg [1].

The ankle plantarflexors have short, pennate muscle fibres with a long, compliant tendon [4,5] and are elongated in mid-stance before shortening at push-off [4]. These features make them ideally suited to the efficient storage and recycling of elastic energy, which reduces the work of the muscle fibres and therefore expenditure of metabolic energy [4–6]. Hence the plantarflexors provide a large percentage of propulsive power at slower jogging speeds [7–9]. At higher running speeds ground contact time is brief and the ability of the plantarflexors to generate more power in less time becomes limited [10]. Further increases in velocity are achieved by increasing cadence [10], primarily by a faster pull-through of the femur by the hip flexors in swing, referred to as H3 [7,8].

During running, the centre of mass is accelerated forwards only during the last 40% of stance phase, which constitutes the propulsive phase of stance [11]. For this reason, investigations of strategies for forward propulsion have focussed on power generated by the plantarflexors at the end of stance. The relative contribution of ankle power to forward velocity has been termed propulsion strategy (PS) and is calculated using the formula A2/(A2 + H3) [8,9]. A higher PS indicates a relatively greater contribution of the plantarflexors to forward velocity and therefore higher metabolic efficiency due to use of the stretch shortening cycle [4–6]. In typically developing (TD) individuals, PS

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decreases with increasing velocity as the hip flexors generate power to increase cadence after plantarflexor power generation has reached a maximum [9]. In TD children, PS of 0.75–0.81 has been reported in jogging and 0.69–0.75 in fast running [8,9].

It has been demonstrated that reduced ankle power generation at push-off results in increased hip flexor power generation in swing during impaired walking [12] and during running in children with developmental coordination disorder (DCD) [8]. The strategy normally used to achieve high velocities is utilized at lower velocities to compensate for reduced ankle power generation. It is unknown whether children with CP use the same compensation strategy during running. In a descriptive study of running in children with diplegic CP, reduced power generation at the ankle, yet similar power generation at the hip, was reported in children with diplegic CP compared to children who were TD [2]. This suggests that ankle power is the limiting factor in running in people with CP, rather than simply a generalized reduction in limb strength and coordination. Determining whether ankle power generation is the limiting factor in running in people with CP would be useful as it would provide direction for intervention.

The three aims of the present study were therefore to (1) describe the PS and components of the PS (A2 and H3) of affected and non-affected legs of children and adolescents with CP across a range of running speeds from jogging to sprinting; (2) determine whether the PS and components of the PS (A2 and H3) are affected by the level of gross motor function as delineated by the GMFCS; and (3) determine whether PS, A2, H3 and maximum speed were different in children with CP compared to children who were TD. It was hypothesized that:

(1) Maximum speed of running would be fastest in TD participants followed by participants classified at GMFCS level I then GMFCS level II.
(2) In participants with CP the affected limb would demonstrate a lower PS, lower A2 and higher H3 than that of the unaffected limb.
(3) In participants with CP both the affected and non-affected limbs would demonstrate a lower PS, lower A2 and higher H3 than that of the TD participants, and that this difference would be largest in the GMFCS II group.

2. Methods

2.1. Participants

Baseline data of participants with CP who were recruited from a community service provider for a larger study conducted in 2015 investigating the effect of a training programme on running in children aged 9–18 years was utilized. The study was approved by the Ethics Committees of Princess Margaret Hospital for Children, Perth, Western Australia (201405SEP) and Curtin University, Perth, Western Australia (HR 219/2014). The trial was prospectively registered with the Australian New Zealand Clinical Trials Notification ACTRN12614000467639. Informed consent was given by the parent/guardian and where applicable, the participant.

The comparative TD data was obtained from a sample of convenience, from participants who were recruited from a cohort of active children aged between 10 and 12 years of age, as part of a different study. Informed consent was given by the parent/guardian and where applicable, the participant.

2.2. Gait data collection

Reflective markers 10 mm in diameter were placed on the skin of participants by an experienced physiotherapist using a modified Cleveland Clinic Foundation marker protocol [13]. Participants in the CP study wore their usual sport shoes; calcaneus and metatarsal markers were placed on the shoes over the relevant landmark. Orthotics extending above the malleoli were not permitted. TD participants performed their trials barefoot. A regression equation was used to calculate the hip joint centre [14]. Knee joint centres were calculated as the midpoint between the medial and lateral femoral condyles [15]. Ankle joint centres were calculated as the midpoint between the medial and lateral malleoli [15]. Inertial and geometric properties of the segments were based on previously published models [16,17].

After a warm up, participants were asked to run at three speeds along a straight walkway in the Curtin University Motion Analysis Laboratory. The speeds were: (1) jog “like a warm-up or like a jog around the oval at school”; (2) run “faster than jogging, but not your fastest” and (3) sprint “like you are in a race”. Ten metres were available before and after the force plates to allow for acceleration and deceleration. At least five trials at each speed were collected unless the participant was too fatigued to continue. A two-minute sitting break was permitted between speeds if required. Kinematic data were recorded by an 18-camera motion capture system at 250 Hz (Vicon T-series, Oxford Metrics, UK). Synchronized ground reaction forces (GRF) were collected at 1000 Hz using three in-ground force platforms in series (AMTI, Watertown, MA). Marker trajectories were labelled and filled using Vicon Nexus 2.5 (Vicon Motion Systems, Oxford, UK).

2.3. Data processing

All data were processed using Visual 3D™ version 6 (C-Motion, Inc.). Kinematic and force plate data were filtered at 18 Hz using a zero-lag 4th order Butterworth filter. An inverse kinematic model with 6-degrees of freedom and specified joint translation boundaries was used [39]. Normalized speed (defined as velocity of the pelvis divided by height), A2, H3 and PS were calculated for each stride. The location of the centre of pressure along the *Y* axis of the foot was identified at initial contact and foot-strike classified as rear-foot (posterior third of the foot), mid-foot (middle third) or fore-foot (anterior third).

2.4. Statistical analysis

For variables A2, H3 and PS all trials were used for analysis. For maximum speed, the fastest trial speed achieved by each participant was used. A2 and H3 were transformed using a Box-Cox transformation to correct right skewedness. A linear mixed model was developed in Statistical Analysis Software (SAS) for each variable. Model validity and optimization were confirmed by meeting convergence criteria and by the Akaike Information Criterion and Bayesian Information Criterion values. Interactions between fixed effects were assessed and excluded if not significant at $p < 0.05$. For all models, random effects were Subject and Subject*Side.

Firstly, the affected and non-affected legs of the participants with CP were compared. Non-affected legs were the non-affected leg of participants with hemiplegic CP, while affected legs were both legs of those participants with bilateral CP and the affected leg of participants with hemiplegic CP. For PS and transformed A2 the fixed effects were, limb status (affected/non-affected), GMFCS level and normalized speed. In the transformed H3 model there was a significant three-way interaction, hence affected and non-affected legs were analyzed separately. For affected legs, fixed effects were GMFCS level and speed, and for non-affected legs fixed effects were GMFCS level, normalized speed and GMFCS level*normalized speed.

Secondly, the affected legs of the participants with CP were compared to the legs of the TD participants. Fixed effects were GMFCS level, normalized speed and GMFCS level*normalized speed for PS, transformed A2 and transformed H3.

Thirdly, the non-affected legs of the participants with CP were compared to the legs of the TD participants. For transformed A2 and PS fixed effects were GMFCS level and normalized speed. For transformed H3 fixed effects were GMFCS level, normalized speed and GMFCS level*normalized speed.

Table 1
Participant demographics.

	CP cohort	TD cohort
Age, mean (SD)	12 years, 11 months (2 years, 9 months)	10 years, 2 months (6 months)
Gender, *n* (%)		
Male	25 (62%)	15 (68%)
Female	15 (38%)	7 (32%)
GMFCS level, *n* (%)		
Level I	25 (62%)	
Level II	15 (38%)	
CP distribution, *n* (%)		
Unilateral		
GMFCS I	11	
GMFCS II	8	
Total	19 (47%)	
Bilateral		
GMFCS I	14	
GMFCS II	7	
Total	21 (53%)	

	CP cohort		TD cohort
	GMFCS I	GMFCS II	
Footstrike pattern			
Jog			
Rearfoot	50%	55%	38%
Midfoot	23%	19%	26%
Forefoot	15%	23%	29%
Inconsistent	13%	3%	7%
Run			
Rearfoot	45%	50%	24%
Midfoot	26%	39%	40%
Forefoot	26%	11%	33%
Inconsistent	4%	0%	2%
Sprint			
Rearfoot	24%	48%	2%
Midfoot	37%	28%	41%
Forefoot	28%	20%	50%
Inconsistent	11%	4%	7%

SD = standard deviation; *n* = number; GMFCS = Gross Motor Function Classification System; % = percent.

3. Results

Participant characteristics are summarized in Table 1. Baseline data of 43 participants in the intervention study were available, three participants were excluded from the current study due to absence of a flight phase.

3.1. Maximum speed

Participants' maximum running speed was significantly slower in GMFCS I than in TD ($t = -5.65$; $p < 0.001$), and significantly slower in GMFCS II than GMFCS I ($t = -3.64$; $p < 0.001$) (Table 2).

3.2. A2

A2 was smaller in affected legs than non-affected legs ($F = 19.91$; $p < 0.001$) and smaller in GMFCS II than GMFCS I ($F = 9.61$; $p = 0.002$). In affected legs, A2 was smaller in GMFCS I than TD ($t = -10.75$ $p < 0.001$) and smaller in GMFCS II than GMFCS I ($t = -9.53$; $p < 0.001$). In non-affected legs, A2 was smaller in GMFCS I than TD ($t = -2.01$; $p = 0.045$) and smaller in GMFCS II than GMFCS I ($t = -3.01$; $p = 0.002$). Speed had a significant effect on A2, which increased exponentially with increasing speed in all limbs ($p < 0.001$) (Fig. 1). As normalized speed was considered as a fixed effect in the linear models, significant differences in A2 found between groups were independent of speed.

3.3. H3

In affected legs, H3 was significantly larger in GMFCS II than GMFCS I ($t = 2.36$; $p = 0.018$) but not significantly different between TD and GFMCS II ($t = -0.79$; $p = 0.427$). In non-affected legs, H3 was not significantly different between GMFCS I and II ($F = 2.84$; $p = 0.093$) or between TD and CP legs ($F = 2.77$; $p = 0.063$). Speed had a significant effect on H3, which increased exponentially with increasing speed in all limbs ($p < 0.001$) (Fig. 1).

3.4. Propulsion strategy

PS was smaller in affected legs than non-affected legs ($t = -3.06$; $p = 0.002$) and smaller in GMFCS II than GMFCS I ($t = -4.11$; $p < 0.001$). In affected legs, PS was smaller in GMFCS I than TD ($t = -2.54$; $p = 0.011$) and smaller in GMFCS II than GMFCS I ($t = -7.16$; $p < 0.001$). In non-affected legs, PS was not significantly different between TD and GMFCS I ($t = -0.74$; $p = 0.458$) but smaller in GMFCS II than GMFCS I ($t = -2.23$; $p = 0.026$). Speed had a significant effect on PS, which decreased with increasing speed in all limbs ($p < 0.001$). PS decreased more quickly in TD than GMFCS I affected legs ($t = 2.71$; $p = 0.007$) and decreased more quickly in GMFCS I affected legs than GMFCS II affected legs ($t = 3.93$; $p < 0.001$) (Figs. 1 and 2).

4. Discussion

The main finding of the present study is that the PS for running in children and adolescents with CP is smaller than the PS of TD children and adolescents. The PS is smaller in affected legs than non-affected legs and smaller in GMFCS II than GMFCS I (Fig. 2). A smaller PS indicates a relatively smaller contribution of the plantarflexors compared with the hip flexors to forward progression of the body. At slower running speeds, in affected legs, children with CP utilize the hip flexors

Table 2
Power and propulsion strategy by GMFCS level – mean of raw data across all speeds.

Group	Affected	Maximum normalized speed Mean (SD)	A2 Mean (SD)	H3 Mean (SD)	PS Mean (SD)
TD	TD	3.57 (0.21)	15.56 W kg^{-1} (4.95 W kg^{-1})	7.87 W kg^{-1} (5.71 W kg^{-1})	0.69 (0.12)
GMFCS I	Affected	2.89 (0.52)[a]	8.64 W kg^{-1} (3.91 W kg^{-1})[a]	3.33 W kg^{-1} (0.67 W kg^{-1})[a]	0.67 (0.11)[a]
	Non-affected		11.10 W kg^{-1} (3.83 W kg^{-1})[a]	3.37 W kg^{-1} (0.73 W kg^{-1})	0.73 (0.09)
GMFCS II	Affected	2.36 (0.44)[ab]	6.00 W kg^{-1} (2.93 W kg^{-1})[ab]	2.76 W kg^{-1} (0.59 W kg^{-1})[b]	0.59 (0.14)[ab]
	Non-affected		7.89 W kg^{-1} (3.93 W kg^{-1})[ab]	2.06 W kg^{-1} (0.64 W kg^{-1})	0.64 (0.19)[ab]

A2 = peak ankle plantarflexor power generation in stance; H3 = peak hip flexion power generation in swing; normalized speed = velocity/height; m = metres; s = second; SD = standard deviation; PS = propulsion strategy A2/(A2 + H3); TD = typically developing; GMFCS = Gross Motor Function Classification Scale; W = Watts; kg = kilogram; [a]significantly different to TD ($p < 0.05$); [b]significantly different to GMFCS I ($p < 0.05$).

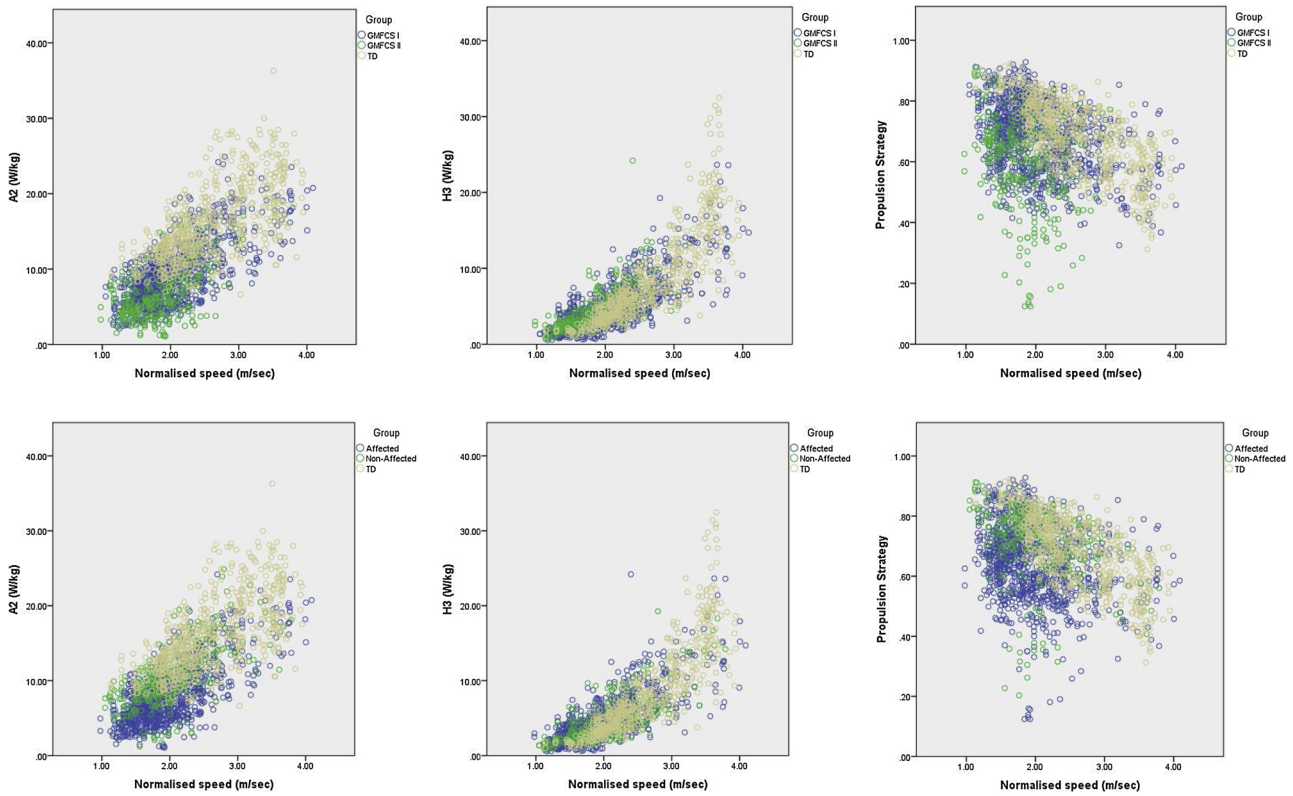

Fig. 1. Peak ankle plantarflexor power generation in stance (A2), Peak hip flexion power generation in swing (H3) and Propulsion Strategy A2/(A2 + H3) (PS), by normalized speed (velocity/height). W = Watts; kg = kilogram; normalized speed = speed/height; m = metre; s = second; GMFCS = Gross Motor Function Classification Scale; TD = Typically developing.

(a proximal strategy) to compensate for reduced plantarflexor power (Fig. 1), which has been reported previously in walking in children with CP [12] and in running in children with DCD [8]. In the present study, A2 was smaller in GMFCS I than TD and smaller in GMFCS II than GMFCS I. Conversely, there was no difference in H3 between groups in non-affected legs, and in fact H3 was larger in GMFCS II than GMFCS I in affected legs at the same speed. This suggests that in affected legs of children with CP, GMFCS level II, H3 is increased to compensate for a smaller A2. This is a strategy that is normally used at higher speeds by people who are TD [10]. In other words, children with CP utilize a normal proximal strategy at slower speeds to compensate for a distal power deficit.

The second main finding of this study is that reduced ankle plantarflexor power in children with CP appears to be the limiting factor for sprinting velocity. In the present study, maximum running velocity was slower in GMFCS I than TD and slower in GMFCS II than GMFCS I. This

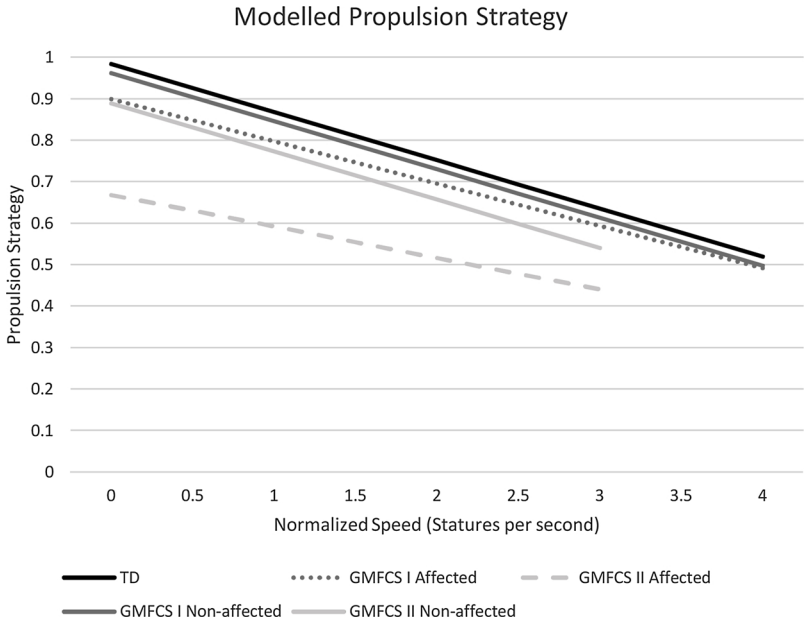

Fig. 2. Modelled Propulsion Strategy.

pattern matches the pattern of A2 in both affected and non-affected legs, which was also smaller in GMFCS I than TD and smaller in GMFCS II than GMFCS I. All groups increased H3 with increasing speed, however this occurred earlier in GMFCS II affected legs compared to GMFCS I affected legs, as demonstrated by both the increased magnitude of H3 in GMFCS II affected legs compared to GMFCS I affected legs at the same speed, and by the slower decrease of PS in GMFCS II affected legs than GMFCS I affected legs (Fig. 2). As the proximal strategy was already employed at slower speeds in GMFCS level II affected legs to compensate for reduced A2, there was less capacity to increase H3 to increase sprinting velocity. Ankle plantarflexor power generation has previously been identified as a limiting factor for running speed in TD sprinters [18] and in children with DCD [8].

A deficit in plantarflexor power in children with CP may be explained by muscle changes which limit the ability to utilize the storage and recycling of elastic energy, termed the stretch-shortening cycle (SSC). This is an important mechanism in running for metabolic efficiency [6]. The SSC is suited to long muscle fascicles and a stiff tendon [19]. During sprinting the plantarflexor muscle fibres act almost isometrically to enhance tendon stretch and recoil [5]. In young people with spastic CP the achilles tendon is longer and thinner than in the TD population which is likely to reduce the efficacy of the SSC [20]. The gastrocnemius muscle fascicles are shorter [20,21] and have a reduced ability to elongate which restricts dorsiflexion range [22], while spasticity may also restrict dorsiflexion range [23]. Hypothetically, the combination of hyperreflexia and reduced compliance of the musculotendinous unit in people with CP could be favourable for the storage and recycling of elastic energy, and therefore the activity of running. Children with CP, GMFCS level II are more likely to be able to run if they have gastrocnemius spasticity [24]. In the present study, children in GMFCS level II had greater maximum A2 with higher levels of spasticity compared to lower levels (Fig. 3). This finding suggests that gastrocnemius spasticity assists running in this group, although their maximum speed was slower than children in GMFCS level I. The largest maximum A2 in children in GMFCS level I occurred with moderate levels of spasticity. Those with either mild or strong spasticity had reduced maximum A2 compared to moderate spasticity (Fig. 3). These findings have implications for interventions which reduce spasticity, such as botulinum toxin injection and tone-reducing medication.

Spastic muscles have been reported to have reduced muscle belly volume, cross sectional area, thickness and length [21] compared to healthy muscles which is likely to result in a reduced capacity for force generation. Muscle weakness in CP may also result from poor muscle recruitment and activation [25]. In running, the ability to generate force rapidly is important [10,26] as ground contact time is very short. Rate of force development has been reported to be reduced in children with CP compared to TD children [26]. Increasing the power (force x velocity) of the plantarflexors in children with CP has been reported to improve walking velocity, cadence and step length [27]. Power training has been reported to be more effective than traditional strength training in increasing muscle fascicle length, muscle belly cross sectional area and velocity of movement in youth with CP [28]. The present study has demonstrated a plantarflexor power deficit in children and adolescents with CP during running which agrees with previous findings and suggests that increasing power generation of the plantarflexors would improve running performance in this population. It is yet to be determined whether power training of the plantarflexors can improve A2, increase maximum speed or reduce proximal compensation during running. This finding should also be considered in context when making decisions regarding interventions with potential to reduce plantarflexor power, such as botulinum toxin injection, calf lengthening surgery and the use of ankle–foot orthoses.

Measures of H3 in the present study were similar to values previously reported in the literature in children with CP [2,29] and children who were TD [2,30]. Measures of A2 in the present study were higher than those previously reported in the literature in children with CP [2,29] and TD children [2,8,30]. For example, A2 of 3–5 W kg^{-1} has previously been reported in children with CP running at 2.3–2.6 ms^{-1} [2,29], while the present study reports a mean A2 of 5.4 W kg^{-1} in GMFCS II and 7.5 W kg^{-1} in GMFCS I during jogging at 2.5–2.7 ms^{-1}. A2 of up to 14.4 W kg^{-1} has been reported in TD children sprinting at 4.2 ms^{-1} [8], while the present study reports A2 of 19.9 W kg^{-1} during sprinting at 5.1 ms^{-1}. We surmise that the higher values of A2 are due primarily to the faster self-selected speeds by participants in our study, compared to previous studies. The higher values of A2 compared to previously published results and the similarity of H3 to previously published results mean we can be confident that any significant lowering of PS is a true reflection of a deficit in plantarflexor power.

4.1. Limitations

A limitation of this study is that although data for both groups were collected in the same laboratory utilizing the same protocol, children with CP wore shoes while TD children were barefoot. Shoe-mounted markers tend to inflate ankle range of motion compared to skin-mounted markers in running [31]. Shod running tends to increase stride length [32] and increase the incidence of a rearfoot striking pattern [32,33] compared to barefoot running. Foot-strike patterns (Table 1) confirm that the CP group had a higher incidence of rearfoot strike than the TD group. Ankle plantarflexion moment has been reported to be 0.28 Nm/kg greater with forefoot strike pattern compared to rearfoot strike pattern [34] and ankle power generation has been reported to be the same [35] or slightly greater [32] in the barefoot condition compared to shod. Both these effects could exaggerate the differences between CP and TD groups. However, participants with CP in this study were on average two years older than the TD group and we did not control for age in our statistical analysis, although we did normalize speed by height. Both maximal speed and horizonal power increase with age in children (approximately 2 W kg^{-1} from pre- to mid-peak height velocity) [36,37] which in this study could reduce the differences between the CP and TD groups. Age and shod condition/striking pattern have opposing influences on the data and the magnitude of each is small compared to the differences found between groups in this study. This paper only considered intra-limb compensation. It is possible that there is an inter-limb compensation pattern and this would be worth investigating further.

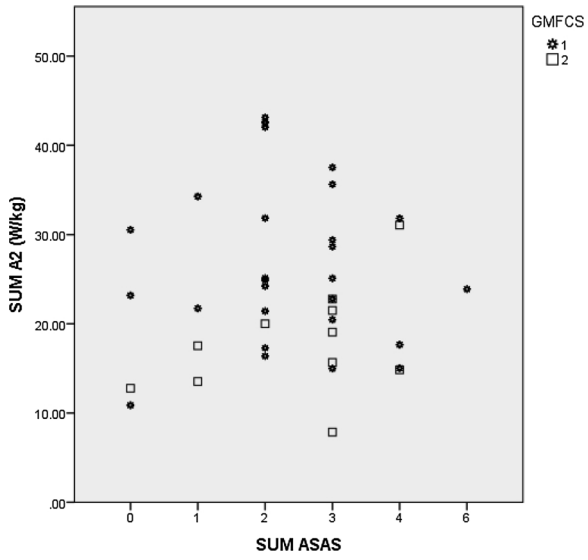

Fig. 3. Ankle Power Generation and Gastrocnemius Spasticity. A2 = Ankle power generation at push-off; ASAS = Australian Spasticity Assessment Scale [38]; GMFCS = Gross Motor Function Classification System; SUM = Left plus Right.

4.2. Conclusion

Children and adolescents with CP have reduced plantarflexor power generation at push off when running, which is compensated for by increasing hip flexor power generation in swing. It appears that reduced A2 limits maximum sprinting speed. Both A2 and maximum running speed are related to GFMCS level, with greater deficits in GMFCS II than GMFCS I.

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Chapter 6 Functional leg stiffness during running in children and adolescents with cerebral palsy

6.1 Preface

This chapter describes the function of the eccentric phase of the stretch-shortening cycle (SSC) during running in children and adolescents with CP, through an examination of leg stiffness, prior to intervention. The study presented in Chapter 5 reported a deficit in ankle power generation for forward propulsion during running. Ankle power generation during running is enhanced by the action of the SSC[272]. It was not clear whether the neuromuscular features of CP would enhance or diminish the efficiency of the SSC and its contribution to power generation during running. Therefore, a cross-sectional study was conducted to describe leg stiffness during jogging, running and sprinting in children with CP and in typically developing children. This chapter reports the outcomes of that study, which was published as "Chappell A, Allison GT, Gibson N, Williams G, Morris S. A comparison of leg stiffness in running between typically developing children and children with cerebral palsy. Clinical Biomechanics. 2021 Mar 23:105337."
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Lecture

A comparison of leg stiffness in running between typically developing children and children with cerebral palsy

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ABSTRACT

Background: Leg stiffness is important during running to increase velocity and maximise efficiency by facilitating use of the stretch-shortening cycle. Children with cerebral palsy who have neuromuscular impairments may have altered leg stiffness. The aim of this study was to describe leg stiffness during running in typically developing children and those with cerebral palsy in Gross Motor Function Classification Scale levels I and II at a range of speeds.

Methods: This cross-sectional study examined kinematic data collected from typically developing children ($n = 21$) and children with cerebral palsy (Gross Motor Function Classification Scale level I $n = 25$, Gross Motor Function Classification Scale level II $n = 13$) during jogging, running and sprinting. Derived variables were resultant ground reaction force, change in leg length and three-dimensional leg stiffness. Linear mixed models were developed for statistical analysis.

Findings: Children with cerebral palsy had reduced stiffness when jogging (Gross Motor Function Classification Scale level I affected $t = 3.81$ $p < 0.01$; non-affected $t = 2.19$ $p = 0.03$; Gross Motor Function Classification Scale level II affected $t = 2.04$ $p = 0.04$) and running (Gross Motor Function Classification Scale level I affected $t = 3.23$ $p < 0.01$) compared to typically developing children. Affected legs were less stiff than non-affected legs only in Gross Motor Function Classification Scale level I during running ($t = 2.26$ $p = 0.03$) and sprinting ($t = 2.95$ $p < 0.01$).

Interpretation: Children with cerebral palsy have atypical leg stiffness profiles which differ according to functional classification.

1. Introduction

Running is a fundamental movement skill which is important in play and physical activity. The human leg has spring-like characteristics during running (Sawicki et al., 2009), as the muscles of the leg store and release elastic energy (Butler et al., 2003). The extent to which it does so is related to its stiffness, defined as the amount of force required to shorten the limb a given distance (Butler et al., 2003). Stiffness reflects the properties and actions of ligaments, tendons, nerves, muscles, bone and cartilage of the entire limb (Pruyn et al., 2012).

Stiffness can be measured in different ways, most commonly as either vertical stiffness or leg stiffness. Both vertical and leg stiffness are measured in the weight-acceptance phase of running. Vertical stiffness, defined as the vertical displacement of the center of mass (CoM) divided by the maximum vertical ground reaction force (GRF), increases with increasing speed. Vertical stiffness is reliable and easy to calculate, but does not measure lower limb deformation nor account for horizontal motion, which is important in running (Maloney and Fletcher, 2018). During walking or running gait, it may be preferable to calculate leg stiffness (K_{leg}), defined as the change in length of the lower limb vector divided by GRF in the line of the lower limb vector, as it takes into account the arc of movement described by the lower limb while the foot is planted (Brughelli and Cronin, 2008; McMahon and Cheng, 1990). However, the calculation of K_{leg} requires more instrumentation than the calculation of vertical stiffness. Vertical stiffness and K_{leg} have different relationships with running speed because of the way they are calculated (Arampatzis et al., 1999).

K_{leg} is important as it reflects the ability to rapidly store elastic

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energy, enabling shorter stance times which are necessary for high velocity sprinting (Butler et al., 2003). K_{leg} is greater with increased joint stiffness, particularly at the knee, increased muscle activation immediately before and after initial contact, increased cadence and decreased stride length (McMahon et al., 2012). In neurotypical adults, K_{leg} remains relatively constant in jogging and running, before increasing at speeds over 5 ms^{-1} (Arampatzis et al., 1999; Brughelli and Cronin, 2008; Heise and Bachman, 2000; McMahon and Cheng, 1990). A high degree of K_{leg} has been associated with improved running performance, including increased running velocity and decreased energy cost of running (Butler et al., 2003).

Optimal mechanical stiffness of the lower limb reflects both the compliance of the anatomical structures and the modulation of active muscular stiffness by neurological activity (Debenham et al., 2016). In the lower limb, the latency of the stretch reflex needs to be coordinated with the K_{leg} and the neurological drive (Debenham et al., 2017). Too much K_{leg} invokes the stretch reflex earlier which requires a high cadence to coordinate the system, while too little K_{leg} requires the muscles to be voluntarily activated in a feed forward response as the stretch reflex is delayed (Debenham et al., 2017). Therefore, in people with cerebral palsy (CP) who have altered muscle tone, muscle weakness and/or contracture, the modulation of K_{leg} becomes a complex motor control problem. It is not easy to predict the impact of a combination of impairments on K_{leg} during running.

Children with spastic diplegia have been reported to have reduced K_{leg} during walking compared to healthy controls (Wang et al., 2015). K_{leg} is yet to be reported during running in this population and may be an important factor to consider as running at different speeds is important for play, sport and recreation. The aim of this study was to describe K_{leg} during running in children and adolescents with CP. We hypothesized that in running 1) K_{leg} would be reduced in children with CP compared to TD children; 2) K_{leg} would be reduced in children with CP with more impaired running function (i.e. Gross Motor Function Classification System (GMFCS) level II compared to level I); 3) K_{leg} would be reduced in the affected legs of children with CP compared to non-affected legs.

2. Methods

2.1. Participants

Data was extracted from baseline data of a larger study investigating the effects of a running training programme in children and adolescents with CP (Gibson et al., 2017). Participants were aged 9–18 years with diagnosis of CP and able to walk 10 m unaided. Participants were excluded if they had undergone surgery in the previous six months or had cognitive or behavioural impairments preventing completion of assessments. Ethical approval for the study was provided by both the Child and Adolescent Health Ethics Committee, Perth, Western Australia (201405SEP) and Curtin University, Perth, Western Australia (HR 219/2014). Comparative TD data was obtained from a sample of convenience of children aged 10–12 years enrolled in recreational sport, who were part of a separate study in the same location using the same protocol. This study had ethical approval from Curtin University (RDHS-48-15). For both studies informed consent was given by the parent/guardian and assent given by the participant.

2.2. Gait data collection

The data collection and processing protocol utilising 18 cameras at 250 Hz (Vicon T-series, Oxford Metrics, UK), three in-ground force platforms in series (AMTI, Watertown, MA), Vicon Nexus 2.5 (Vicon Motion Systems, Oxford, UK) and Visual 3D™ version 6 (C-Motion, Inc.) has been described previously (Chappell et al., 2019a; Chappell et al., 2019b). All participants were barefoot. Reflective markers 10 mm in diameter were placed according to a modified Cleveland Clinic Foundation marker protocol (Sutherland, 2002).

Participants warmed up and were then asked to run at three speeds along a straight walkway, in this order: 1) jog "like a warm-up or like a jog around the oval at school"; 2) run "faster than jogging, but not your fastest" and 3) sprint "like you are in a race". No equipment was used, and speed was not dictated, to avoid altering participants' natural running pattern. Ten meters were available before and after the force plates to allow for acceleration and deceleration. At least five trials at each speed were collected unless the participant was too fatigued to continue. A two-minute sitting break was permitted between speeds if required.

2.3. Data processing

All steps with a clean force plate strike were included for analysis. The hip joint centre was calculated using a regression equation (Bell et al., 1989), the knee joint centre was calculated as the midpoint between the medial and lateral femoral condyles (Pohl et al., 2010), and the ankle joint centre was calculated as the midpoint between the medial and lateral malleoli (Pohl et al., 2010). Inertial and geometric properties of the segments were based on previously published models (Dempster, 1955; Hanavan Jr, 1964). All data were processed using Visual 3D™ version 6 (C-Motion, Inc.). Kinematic and force plate data were filtered at 18 Hz using a zero-lag 4th order Butterworth filter. An inverse

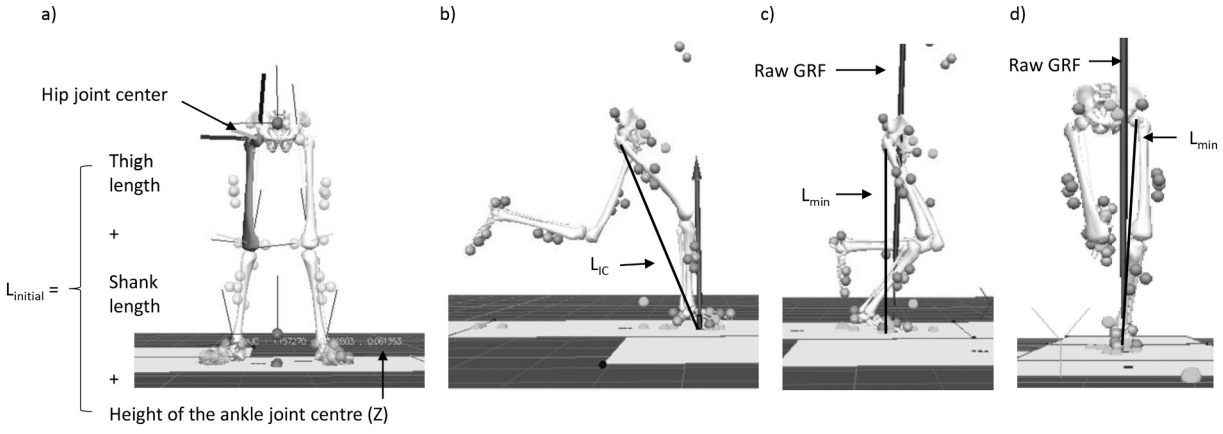

Fig. 1. Calculation of $L_{initial}$, L_{IC} and L_{min} a) definition of leg length is the distance between the center of pressure and hip joint center, b) leg length at initial contact (L_{IC}), c) minimum leg length (L_{min}), d) raw GRF is displayed here from which the portion of GRF in line with the leg vector at L_{min} is calculated.

Table 1
Participant characteristics.

	CP Cohort	TD Cohort
Age Mean(SD)	12 yrs., 7 m (2 yrs., 9 m)	10 yrs., 2 m (6 m)
Gender n (%)	Male = 24 (63%)	Male = 14 (67%)
	Female = 14 (37%)	Female = 7 (33%)
GMFCS Level n (%)	Level I = 25 (66%)	
	Level II = 13 (34%)	
CP Distribution n (%)	Unilateral	
	GMFCS I = 11	
	GMFCS II = 7	
	TOTAL = 18 (47%)	
	Bilateral	
	GMFCS I = 14	
	GMFCS II = 6	
	TOTAL = 20 (53%)	

SD=Standard Deviation; n = number; yrs. = years; m = months; GMFCS = Gross Motor Function Classification System; % = percent.

kinematic model with 6-degrees of freedom and specified joint translation boundaries was used (Chappell et al., 2019b). Normalized speed was defined as velocity of the pelvis divided by height and reported as statures per second (SPS). Step length and flight time were calculated in Visual 3D™, step length was normalized to initial leg length ($L_{initial}$), where $L_{initial}$ was the sum of the length of the thigh segment, length of the shank segment and vertical height of the ankle joint centre in quiet stance (Fig. 1). Foot flat posture was ensured during quiet stance.

Leg length during running was defined as the length of the leg vector, the leg vector being the straight-line distance between the hip joint center and the center of pressure of the stance foot. L_{IC} was the leg length at initial contact and L_{min} the minimum leg length in stance (Liew et al., 2017) (Fig. 1). Resultant GRF (rGRF) was defined as the component of GRF in line with the leg vector at L_{min}, normalized to body weight (body mass*9.81). K_{leg} was calculated as dimensionless units by using the normalized GRF vector and dividing the ΔL by $L_{initial}$:

$$K_{leg} = \frac{\frac{GRF_{vector(Lmin)}}{mg}}{\frac{L_{IC}-L_{min}}{L_{initial}}}$$

Direct kinematic-kinetic modelling of leg stiffness is considered the "Gold-standard" for quantifying leg stiffness during gait, and utilising a three-dimensional method has been reported to represent biological leg function better than a uni-dimensional method (Liew et al., 2017).

2.4. Analysis

Data from both legs of all participants were included for analysis. The non-affected leg of participants with hemiplegic CP was categorized as "non-affected" and all other legs were categorized as "affected". Data analysis was undertaken in Statistical Analysis Software (SAS 9.4, SAS Institute Inc., Cary, NC, USA). Boxcox transformation was used for K_{leg} and rGRF to correct right skewedness. Mixed models were developed for K_{leg}, rGRF, ΔL, step length and flight time with fixed effects Descriptor (GMFCS level (TD, GMFCS I, GMFCS II) and affected/non-affected) and SPS. Random effects were subject and subject*side. Least square means of the fixed effects of K_{leg}, rGRF, ΔL, step length and flight time were extracted from the models at SPS = 1, 2 and 3. These speeds were chosen to reflect jogging, running and sprinting speeds, based upon the recorded speeds of participants.

3. Results

3.1. Participants

Baseline data was collected from 43 participants with CP and 21 TD participants. For this analysis four participants with CP were excluded for lack of a flight phase (flight phase being at least one frame with neither foot in contact with the floor) and one participant with CP was excluded due to incomplete data (excessive marker occlusion). Characteristics of included participants are shown in Table 1. Data from both legs of all included participants were analysed. An average of seven jog trials, five run trials and four sprint trials per leg were included.

3.2. Running speed

In the TD cohort speed ranged from 1.14 to 3.74 SPS. In GFMCS level I speed ranged from 1.01 to 3.92 SPS and in GFMCS level II speed ranged from 0.99 to 3.12 SPS. In children with CP, K_{leg} increased more rapidly with increasing speed compared to TD children (GMFCS I affected $p < 0.01$; non-affected $p < 0.01$; GMFCS II affected $p = 0.04$), except for in

Table 2
Group averages for each running related variable by group (GMFCS level I, II or TD).

Variable	Group	JOG Mean (SD)	RUN Mean (SD)	SPRINT Mean (SD)
Velocity (ms^{-1})	TD	3.04 (0.38)	4.00 (0.41)	4.94 (0.31)
	GMFCS I	2.40 (0.34)	3.52 (0.55)	4.40 (0.73)
	GMFCS II	2.19 (0.47)	3.11 (0.66)	3.74 (0.70)
Leg Stiffness	TD	46.34 (27.89)	50.69 (28.37)	51.58 (25.84)
	GMFCS I Not affected	38.37 (17.67)	56.33 (32.60)	52.42 (23.48)
	GMFCS I Affected	29.91 (13.97)	32.93 (15.94)	38.84 (20.76)
	GMFCS II Non-affected	29.25 (6.31)	34.57 (12.24)	35.86 (11.99)
	GMFCS II Affected	31.37 (11.15)	38.55 (16.46)	46.99 (31.89)
Resultant GRF	TD	2.79 (1.12)	2.83 (1.15)	2.66 (1.06)
	GMFCS I Not affected	2.30 (0.71)	2.19 (0.42)	2.51 (0.46)
	GMFCS I Affected	2.06 (0.30)	2.60 (0.74)	2.20 (0.42)
	GMFCS II Non-affected	1.76 (0.13)	1.86 (0.16)	1.95 (0.15)
	GMFCS II Affected	1.84 (0.25)	1.95 (0.34)	1.95 (0.27)
% change in leg length	TD	7.14 (2.94)	6.59 (2.77)	5.82 (2.20)
	GMFCS I Not affected	6.81 (2.48)	5.53 (2.28)	5.58 (2.74)
	GMFCS I Affected	8.04 (3.06)	7.66 (2.84)	6.74 (2.68)
	GMFCS II Non-affected	6.28 (1.29)	5.99 (2.04)	5.93 (1.69)
	GMFCS II Affected	6.46 (1.93)	5.96 (2.41)	5.56 (2.62)
Step Length/leg length	TD	1.25 (0.17)	1.36 (0.15)	1.41 (0.17)
	GMFCS I Not affected	0.93 (0.13)	1.24 (0.16)	1.34 (0.13)
	GMFCS I Affected	0.98 (0.13)	1.19 (0.18)	1.37 (0.23)
	GMFCS II Non-affected	0.91 (0.13)	1.01 (0.15)	1.13 (0.13)
	GMFCS II Affected	0.94 (0.12)	1.10 (0.20)	1.22 (0.17)

GRF = Ground reaction force; % = percentage; ° = degrees; SD = standard deviation; GMFCS = Gross Motor Function Classification Scale.

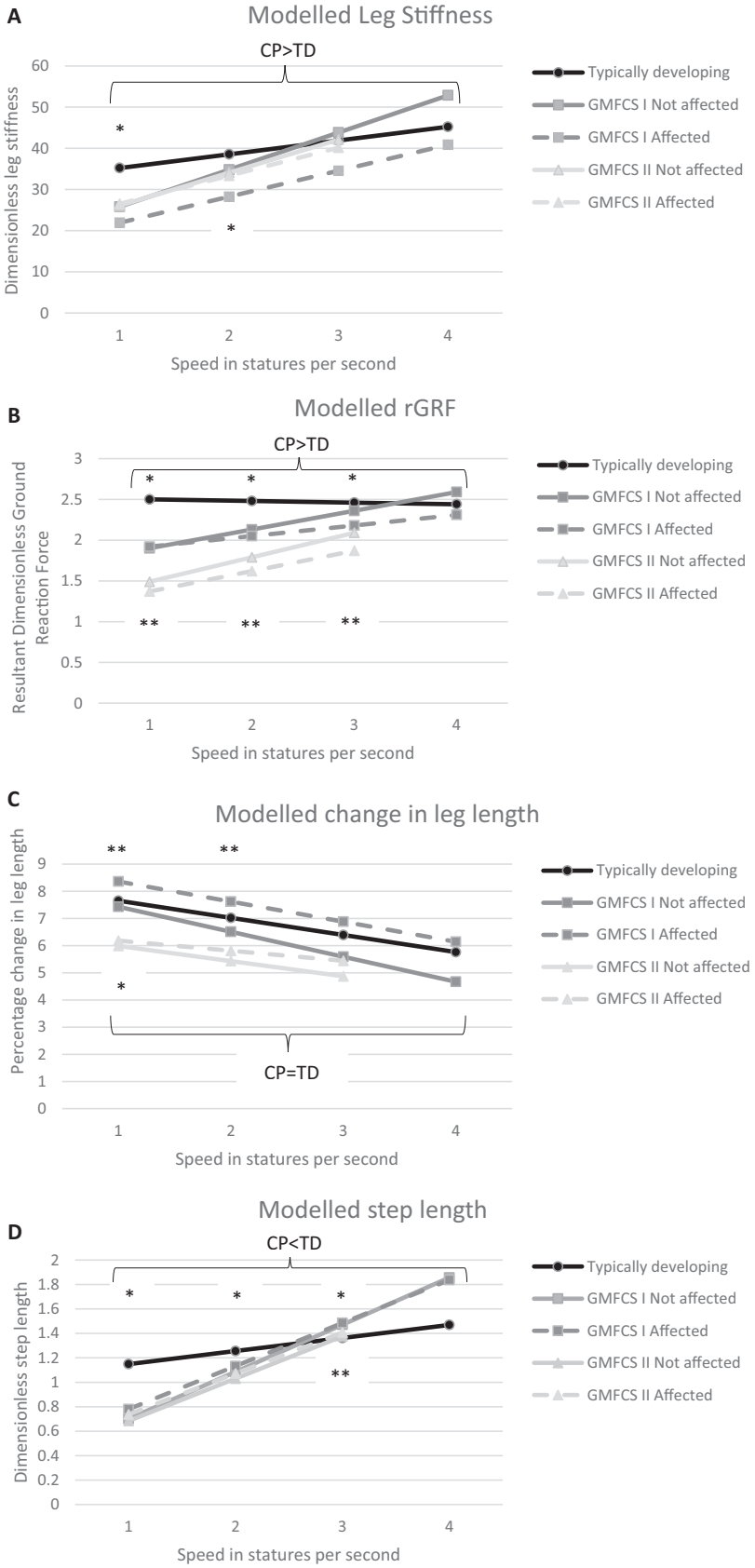

Fig. 2. Modelled a) leg stiffness, b) rGRF, c) change in leg length and d) step length by GMFCS level (TD, I or II) and affected/non-affected. *Difference between TD and CP $p < 0.05$; **Difference between GMFCS I and II $p < 0.05$; CP = cerebral palsy; TD = typically developing.

GMFCS level II non-affected legs ($p = 0.07$) (Table 2, Fig. 2a). With increasing speed in TD children rGRF remained constant ($p = 0.22$) but increased in children with CP with increasing speed (all pairwise comparisons $p < 0.01$) (Table 2, Fig. 2b). In all groups, ΔL decreased with increasing speed ($p < 0.01$), there was no difference in slope between groups (Table 2, Fig. 2c). Step length increased with increasing speed in all groups ($p < 0.01$) and increased more rapidly in children with CP compared to TD children ($p < 0.01$; Table 2, Fig. 2d).

3.3. Jogging (SPS = 1)

TD participants had greater K_{leg} than those in GMFCS level I (affected $t = 3.81$ $p < 0.01$; non-affected $t = 2.19$ $p = 0.03$) and affected legs of those in GMFCS level II ($t = 2.04$ $p = 0.04$). All other pairwise comparisons were non-significant (Table 2, Fig. 2a). TD participants had greater rGRF than those in GMFCS level I (affected $t = 6.13$ $p < 0.01$; non-affected $t = 5.45$ $p < 0.01$) or GMFCS level II (affected $t = 9.80$ $p < 0.01$; non-affected $t = 7.70$ $p < 0.01$). Further, participants in GMFCS level I had greater rGRF than those in GMFCS level II (all pairwise comparisons $p < 0.01$). There was no difference in rGRF between affected and non-affected legs in GMFCS level I ($t = 0.08$ $p = 0.93$) or GMFCS level II ($t = 1.31$ $p = 0.19$) (Table 2, Fig. 2b). ΔL was less in affected legs of those in GMFCS level II than TD participants ($t = 2.02$ $p = 0.05$). ΔL was greater in affected legs of those in GMFCS level I than those in GMFCS level II (affected $t = 3.10$ $p < 0.01$; non-affected $t = 2.38$ $p = 0.02$). All other pairwise comparisons were non-significant (Table 2, Fig. 2c). TD participants had longer step length than those in GMFCS level I (affected $t = 11.39$ $p < 0.01$; non-affected $t = 11.74$ $p < 0.01$) and GMFCS level II (affected $t = 10.72$ $p < 0.01$; non-affected $t = 10.24$ $p < 0.01$). Affected legs had longer step length than non-affected legs (GMFCS level I $t = 2.93$ $p < 0.01$; GMFCS II $t = 2.25$ $p = 0.03$). All other pairwise comparisons were non-significant (Table 2, Fig. 2d).

3.4. Running (SPS = 2)

In GMFCS level I affected legs were less stiff than non-affected legs ($t = 2.26$ $p = 0.03$) and less stiff than those of TD participants ($t = 3.23$ $p < 0.01$). All other pairwise comparisons were non-significant (Table 2, Fig. 2a). TD participants had greater rGRF than those in GMFCS level I (affected $t = 4.71$ $p < 0.01$; non-affected $t = 3.25$ $p < 0.01$) or GMFCS level II (affected $t = 7.73$ $p < 0.01$; non-affected $t = 5.54$ $p < 0.01$). Participants in GMFCS level I had greater rGRF than those in GMFCS level II (all pairwise comparisons $p < 0.05$). There was no difference in rGRF between affected and non-affected legs in GMFCS level I ($t = 1.57$ $p = 0.12$) but rGRF was greater in non-affected legs than affected legs in GMFCS level II ($t = 2.22$ $p = 0.03$) (Table 2, Fig. 2b). ΔL was greater in GMFCS level I affected legs than non-affected legs ($t = 2.22$ $p = 0.03$) and greater than in GMFCS level II (affected $t = 2.82$ $p < 0.01$; non-affected $t = 2.50$ $p = 0.01$). All other pairwise comparisons were non-significant (Table 2, Fig. 2c). TD participants had longer step length than those in GMFCS level I (affected $t = 4.55$ $p < 0.01$; non-affected $t = 5.51$ $p < 0.01$) and GMFCS level II (affected $t = 5.63$ $p < 0.01$; non-affected $t = 6.19$ $p < 0.01$). GMFCS level I affected legs had longer step length than non-affected legs (GMFCS level I $t = 2.37$ $p = 0.02$; GMFCS II $t = 2.89$ $p < 0.01$). All other pairwise comparisons were non-significant (Table 2, Fig. 2d).

3.5. Sprinting (SPS = 3)

In GMFCS level I affected legs were less stiff than non-affected legs ($t = 2.95$ $p < 0.01$). All other pairwise comparisons were non-significant (Table 2, Fig. 2a). TD participants had greater rGRF than those in GMFCS level II (affected $t = 5.11$ $p < 0.01$; non-affected $t = 2.71$ $p < 0.01$) and affected legs of those in GMFCS level I ($t = 3.06$ $p < 0.01$). There was no difference between TD legs and non-affected legs of GMFCS level I ($t = 0.78$ $p = 0.44$). In GMFCS level I non-affected legs had greater rGRF than affected legs ($t = 3.07$ $p < 0.01$). Non-affected legs of participants in GMFCS level I had greater rGRF than those in GMFCS level II (affected $t = 4.11$ $p < 0.01$; non-affected $t = 1.98$ $p = 0.049$). rGRF was greater in affected legs of participants in GMFCS level I than GMFCS level II ($t = 2.68$ $p = 0.01$) but were not different from non-affected legs in GMFCS level II ($t = 0.56$ $p = 0.58$) (Table 2, Fig. 2b). In GMFCS level I ΔL was greater in affected legs than non-affected legs ($t = 2.37$ $p = 0.02$). All other pairwise comparisons were non-significant (Table 2, Fig. 2c). Participants in GMFCS level I had longer step length than TD children (affected $t = 4.12$ $p < 0.01$; non-affected $t = 2.95$ $p < 0.01$) and GMFCS level I affected legs had longer step length than GMFCS level II non-affected legs ($t = 2.26$ $p = 0.02$). All other pairwise comparisons were non-significant (Table 2, Fig. 2d).

4. Discussion

The main finding of this study is that children with CP had reduced leg stiffness when jogging and running compared to TD children, although leg stiffness increased more rapidly with increasing speed in the CP group. Affected legs were less stiff than non-affected legs only in GFMCS level I during running and sprinting, which seemed to be due to an increased ΔL in the affected leg. Therefore, our first hypothesis, that K_{leg} would be reduced in children with CP compared with TD children was accepted only for jogging and running, while our second hypothesis, that K_{leg} would be reduced in GMFCS level II compared to level I, was rejected. Our third hypothesis, that K_{leg} would be reduced in affected legs compared to non-affected legs, was only accepted for GMFCS level I.

Mathematically, K_{leg} is directly proportional to rGRF (McMahon and Cheng, 1990). At higher speeds, greater ankle power generation at push-off is associated with more vertical displacement of the COM and therefore increased vertical GRF in the subsequent loading phase (Nilsson and Thorstensson, 1989). Our results suggest that rGRF reflects functional capacity, as it was largest in TD, and larger in GMFCS level I than level II. Children with CP have a deficit in ankle power generation during running which is more marked in GMFCS level II than level I (Chappell et al., 2019a), resulting in a smaller rGRF compared to TD children. This finding is a likely explanation for the reduction in K_{leg} observed in children with CP, however, it does not sufficiently explain why there was no difference in K_{leg} between GMFCS levels I and II, or between affected and non-affected legs within functional groups.

In neurotypical adult runners K_{leg} remains reasonably constant until cadence increases at a threshold of about 5 ms^{-1} (Brughelli and Cronin, 2008). This study has found that in all children K_{leg} increased over a range of speeds and that in children with CP, K_{leg} increases more rapidly than in TD children. In children with CP, rGRF increased with increasing speed rather than remaining constant as it did in TD children. Vertical GRF increases as speed increases (Arampatzis et al., 1999). Therefore, at midstance in TD children, the position of the hip joint center must move progressively away from the vertical GRF with increasing speed, to maintain rGRF at a constant level. Conversely, children with CP appear to maintain the hip joint center over the center of pressure in mid-stance as speed increases, causing rGRF to increase with increasing speed. This suggests that when children with CP run they have a shorter step length, rapidly increasing cadence, reduced flight height and reduced trunk inclination (termed a 'gliding' pattern) compared to the greater vertical displacement and longer strides utilised by TD children (or a 'bounding' pattern (Kenyon, 2013)) (Bohm and Doderlein, 2012; Davids et al., 1998). In a gliding pattern the foot contacts the ground further in front of the CoM, with higher hip extensor power generation in early stance to move the pelvis over the stance foot (Kenyon, 2013). The upright trunk maintains the line of the raw GRF through the hip joint such that rGRF increases with increasing speed, and thereby K_{leg}.

Mathematically, K_{leg} is inversely proportional to ΔL (McMahon and Cheng, 1990). In running and sprinting in GMFCS level I, affected legs had greater ΔL than non-affected legs, which is the most likely explanation for the reduced stiffness in affected legs compared to non-affected

legs in GMFCS level I. In jogging and running, ΔL was smallest in GMFCS level II which may be because they have the lowest rGRF (Fig. 2b), i.e. less force through the leg producing less ΔL. As rGRF was smaller in GMFCS II than I, less eccentric plantarflexor strength was required in this group to resist dorsiflexion in the loading phase. GMFCS level I affected legs have a deficit in plantarflexor strength compared to TD children (Chappell et al., 2019a) and a greater eccentric strength requirement than GMFCS level II, which may explain why ΔL was greatest in this group. If this is indeed so, it suggests that improving the control of both the lengthening and shortening phases of the stretch-shortening cycle in cyclical loading activities is important in children with CP.

In neurotypical adults high running velocities are achieved by increasing stride frequency, which necessitates a reduction in ground contact time and therefore a reduced excursion at the knee during contact i.e. decreased ΔL (Arampatzis et al., 1999; Cavagna et al., 1988). The need to reduce ΔL with increasing speed is reflected in our finding that there was no significant difference in the slope of ΔL between groups. Our findings support previous reports that children with CP have reduced stride length compared to TD children (Fig. 2d) (Davids et al., 1998) and rely on a higher cadence to achieve the same velocity (Bohm and Doderlein, 2012). This may be a factor limiting maximum velocity in this group, as the leg reaches a minimum shortening of about 5% at slower velocities.

Heise and colleagues reported unchanging K_{leg} with speed in TD children (Heise and Bachman, 2000), although they had only one data point for speeds over 5 ms^{-1}. In contrast, we had many data points over 5 ms^{-1}, with a maximum velocity 5.69 ms^{-1}, which may have allowed us to identify the increase in K_{leg} at high velocities. The values of K_{leg} obtained in this study at $\geq$3 SPS were similar to those previously reported in TD adults running at 5 ms^{-1} using the same multi-planar method. Sprinting is a distinctive form of running, with minimal leg shortening and maximal GRF (Miyashiro et al., 2019). Not all participants with CP were able to achieve speeds over 3 SPS, particularly participants in GMFCS level II, so the study may have failed to find more differences between groups during sprinting due to being underpowered at this speed. It is also possible that those participants with CP who were able to sprint at higher speeds were those whose sprinting characteristics were not that dissimilar to those of TD children.

In summary, the results of this study support previous findings that a lack of plantarflexor power at push-off is a fundamental deficit during running in children with CP, and that this is more pronounced in GMFCS level II than level I (Chappell et al., 2019a). Therefore, running training programs for this population should incorporate calf power training. Furthermore, the amount of leg shortening during loading was excessive for the amount of force, which suggests that running training programs should also incorporate cyclical loading activities (progressively weighted jumping, hopping or jogging (Gibson et al., 2017)) to facilitate learned control of the weight-acceptance phase. Both components require training for children with CP to achieve typical levels of leg stiffness and improve running function.

4.1. Limitations

K_{leg} has been reported to increase with maturation during maximum sprint running (Rumpf et al., 2013), which is a confounding factor in this study of youth aged 9–18 years. However, the TD cohort was on average younger than the CP cohort which, if anything, would cause us to expect them to have lower K_{leg} than the CP group, while we have found the opposite. This analysis of K_{leg} focuses on running function and while the investigation of the correlations between impairments and K_{leg} is interesting, such analysis is beyond the scope of this paper. This analysis has utilised a three-dimensional calculation of K_{leg} which is recommended in the literature (Maloney and Fletcher, 2018) but makes comparison with previous reports of two-dimensional K_{leg} difficult. All calculations of K_{leg} from modelled data require accurate localisation and tracking of the hip joint center. Future improvements in modelling accuracy may further refine estimates of K_{leg}.

Declaration of Competing Interest

The authors declare no conflict of interest.

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Chapter 7 The effect of a running training intervention on ankle power generation in children and adolescents with cerebral palsy: a randomised controlled trial.

7.1 Preface

Chapter 5 presented an analysis of baseline data that supported previous reports that ankle power generation during running was reduced in children and adolescents with CP[131]. The systematic review reported in Chapter 2 of this thesis identified a lack of studies investigating the effect of running training on the biomechanics of running. A small number of studies have used the Muscle Power Sprint Test to evaluate the effect of training interventions on anaerobic muscle power in children and adolescents with CP, with mixed results[141, 147, 181, 182, 190]. In these studies, the mechanism of improvement was not reported. Therefore, it remains unknown whether increases in running speed in people with CP are achieved by remediating ankle power generation or by increasing proximal compensation.

This chapter describes the effect of a task-specific, low-load plyometric running training program on power generation in children and adolescents with CP. The study in this chapter was published as "Chappell, A., Allison, G. T., Williams, G., Gibson, N., & Morris, S. (2020). The effect of a running training intervention on ankle power generation in children and adolescents with cerebral palsy: A randomized controlled trial. Clinical Biomechanics, 76." https://doi.org/10.1016/j.clinbiomech.2020.105024.

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The effect of a running training intervention on ankle power generation in children and adolescents with cerebral palsy: A randomized controlled trial

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ABSTRACT

Background: Children and adolescents with cerebral palsy who are classified as Gross Motor Function Classification Scale level I or II are usually able to run but lack ankle power generation for push-off. The aim of this study was to analyze the efficacy of a running training program in improving ankle power generation in children and adolescents with cerebral palsy.

Methods: This randomized controlled trial compared kinematic and spatiotemporal data collected during running from 38 children and adolescents with unilateral or bilateral cerebral palsy before and after a 12-week running program. Normalized speed, stride length, cadence, foot strike pattern, peak ankle power generation, peak hip flexor power generation in swing and propulsion strategy were calculated. Linear mixed models were developed to analyze differences between groups.

Findings: At follow-up the intervention group had increased normalized speed of running ($t = -3.68$ $p < .01$) while the control group got slower ($t = 3.17$ $p < .01$). In running, children in Gross Motor Function Classification Scale level II in the intervention group increased ankle power ($t = 2.49$ $p = .01$) while the control group did not change ($t = 0.38$ $p = .71$). In sprinting, children in Gross Motor Function Classification Scale levels I and II in the intervention group maintained ankle power (level I $t = 0.32$ $p = .75$; level II $t = 1.56$ $p = .12$) while those in the control group decreased ankle power (level I $t = 4.69$ $p < .01$; level II $t = 2.52$ $p = .01$). Most within-group differences did not result in significant between-group differences at follow-up.

Interpretation: Power generation for running may be responsive to targeted intervention in children with cerebral palsy.

1. Introduction

Cerebral palsy (CP) is the most common physical disability of childhood, affecting about two in every 1000 live-born infants (Galea et al., 2019). Children and adolescents with CP who are classified as Gross Motor Function Classification System (GMFCS) level I and approximately half those classified as GMFCS level II are able to run (Böhm et al., 2018). Running is a fundamental movement skill which is important for participation in both recreational and school activities.

The propulsive phase of running, when the center of mass is accelerated forwards, occurs in the last 40% of stance phase, corresponding to the activity of the plantarflexors (Hamner et al., 2010). In typically developing (TD) individuals, power for forward propulsion during running is provided primarily by the ankle plantarflexors at push-off (Diamond et al., 2014; Lye et al., 2016; Schache et al., 2015), termed A2 (Winter, 1983). Other contributors to forward velocity include the hip extensors which generate power in early stance (termed H1 (Winter, 1983)) both to advance the pelvis over the support foot and to maintain a stable trunk (Bezodis et al., 2008), and the hip flexors which generate power in swing (termed H3 (Winter, 1983)) to pull the femur through (Diamond et al., 2014; Schache et al., 2015). In sprinting, H3 has an increasingly important contribution to forward velocity compared to running or jogging. This is because at high velocities ground contact time is brief and the ability of the ankle plantarflexors to generate larger amounts of power rapidly plateaus hence further increases in velocity are dependent on increasing cadence

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(Schache et al., 2014).

The relative contributions of A2 and H3 to forward propulsion has been termed 'propulsion strategy' (PS) and is calculated as PS = A2/(H3 + A2) (Diamond et al., 2014). In TD children, PS ranges from 0.81 in jogging to 0.75 in sprinting (Diamond et al., 2014). Children and adolescents with CP run more slowly than TD peers and have a lower PS as they use a greater proportion of H3 at slow speeds to compensate for reduced A2 (Chappell et al., 2019; Davids et al., 1998). This premature shift to a proximal power generation pattern in youth with CP may have a higher energy cost compared to typical running (Ries and Schwartz, 2017).

As running is important in childhood and adolescence for play and participation (Logan et al., 2015) a number of studies have evaluated interventions to improve running. Both strength and plyometric training have been reported to increase muscle power and improve running performance in TD children (Behringer et al., 2011; Johnson et al., 2011). A small number of studies have used the Muscle Power Sprint Test to evaluate the effect of training interventions on anaerobic muscle power in children and adolescents with CP, with mixed results (Fisher-Pipher et al., 2017; Gibson et al., 2017; Hedgecock et al., 2015; Kenyon et al., 2010; Verschuren et al., 2007). In these studies, the mechanism of improvement was not reported. Therefore, it remains unknown whether increases in running speed in people with CP are achieved via a typical propulsion strategy or by increasing proximal compensation. Normalization of lower-limb power generation through increasing A2 would result in a higher PS while a greater reliance on a compensatory strategy would result in a lower PS. Increasing running speed is desirable for participation in play and sport but it is also important that, if possible, higher speeds are achieved using a strategy that is economical. Clinicians who train running in children with CP should know whether they can focus on a 'normalization' strategy or need to choose a 'compensatory' strategy.

Running training for adults with acquired brain injury (Williams and Schache, 2010) has been reported to improve ankle power generation, normalizing the running strategy (Williams and Schache, 2019). The program focuses primarily on increasing A2, utilizing ballistic exercises targeting firstly the ankle plantarflexors and secondly the hip flexors. We previously reported that running training for children and adolescents with CP was effective for achieving running related goals and increasing participation in physical activity at school (Gibson et al., 2017). To extend this work we undertook this analysis with the aim of determining whether the training program was effective in increasing ankle power generation and thereby normalizing the distribution of power generation for running in this cohort.

1.1. Hypotheses

We hypothesized that the training program would:

1. Increase peak ankle power generation (A2) in jogging, running and sprinting
2. Increase peak hip power generation in sprinting (H3)
3. Increase the contribution of ankle power generation to propulsion in jogging and running (PS)

2. Method

2.1. Participants

Participants included in this analysis were aged between 9 and 18 years; had a diagnosis of CP, GMFCS level I-II, able to walk 10 m unaided; had a goal to either a) run or b) improve their running; were willing and able to attend two training sessions weekly after school; and were willing to complete two additional exercise sessions at home per week. Participants were excluded if they had undergone surgery in the previous six months; had a medical condition which contraindicated strenuous exercise; were unable to complete assessments due to a cognitive impairment; or had cognitive or behavioral impairments which precluded intervention in a group setting. A flight phase was required for inclusion in the current analysis. A stratified random allocation to control or intervention group was conducted by an independent team member (Gibson et al., 2017). Allocation was stratified by age and High-Level Mobility Assessment Tool (HiMAT) score to keep groups equivalent. The study was approved by the Child and Adolescent Health Service Ethics Committee, Perth, Western Australia (201405SEP) and the Human Research Ethics Committee of Curtin University, Perth, Western Australia (HR 219/2014). The trial was prospectively registered with the Australian New Zealand Clinical Trials Notification ACTRN12614000467639. Informed consent was given by the parent/guardian and assent by the participant where applicable.

2.2. Gait data collection

All participants attended for data collection at the Curtin University Motion Analysis Laboratory before and after a 12-week running intervention program. One experienced physiotherapist placed reflective markers 10 mm in diameter on the skin of participants according to a modified Cleveland Clinic Foundation marker protocol (Sutherland, 2002). Participants were barefoot and instructed to: 1) jog "like a warm-up or like a jog around the oval at school"; 2) run "faster than jogging, but not your fastest" and 3) sprint "like you are in a race" along a straight runway. A 30-m running track with force plates positioned mid-way was used to ensure participants reached a constant velocity before traversing the plates. At least five trials at each speed were collected unless limited by the capacity of the participant to continue. A two-minute sitting break was permitted between speed groups (jog, run, sprint) if required. Kinematic data were recorded by an 18-camera motion capture system at 250 Hz (Vicon T-series, Oxford Metrics, UK). Synchronized ground reaction forces were collected at 1000 Hz using three in-ground force platforms in series (AMTI, Watertown, MA).

2.3. Data processing

Marker trajectories were labelled and filled using Vicon Nexus 2.5 (Vicon Motion Systems, Oxford, UK). Spline filling was used as a first option, followed by pattern filling. Data were then processed in Visual 3D using an inverse kinematic model with 6-degrees of freedom and specified joint translation boundaries (Chappell et al., 2019). Kinematic and force plate data were filtered at 18 Hz using a zero-lag 4th order Butterworth filter. Presence or absence of a flight phase was determined by an algorithm in Visual 3D™ version 6 (C-Motion, Inc.). Peak A2, peak H3 and normalized speed (statures per second (SPS)) were calculated for each stride. A2 and H3 were normalized by body weight (W/kg). The location of the center of pressure along the Y axis of the foot was identified at the fourth frame of initial contact by a Visual 3D™ algorithm and classified as rear-foot (posterior third of the foot), mid-foot (middle third) or fore-foot (anterior third).

2.4. Intervention

A 12-week individualized intervention program conducted twice-weekly in a group setting was delivered to the intervention group by physiotherapists employed by a community service provider. The intervention, which targeted ankle plantarflexor power and hip flexor power, has been described in more detail in a previously published paper (Gibson et al., 2017). Briefly, at its core, the intervention consisted of:

- Calf power exercises progressing from a de-weighted position to over-ground
- Hip flexor power exercises progressing from a de-weighted position to over-ground

- Motor learning exercises for running technique
- Transference of learned patterns to over-ground running

The control group received usual care over the intervention period. Usual care was not controlled but typically consisted of provision of a home exercise program and consultation on request.

2.5. Statistical analysis

Box Cox transformations were used to correct right skewedness in A2 and H3, and left skewedness in PS to facilitate model fitting. Linear mixed models were developed in Statistical Analysis Software (SAS) 9.4 (SAS Institute, Cary NC) for each GMFCS level for variables A2, H3, PS, stride length and cadence. Fixed effects were group, time and normalized speed, random effects were subject and subject(side) (except for stride length and cadence). Model validity and optimization were confirmed by meeting convergence criteria and by the Akaike Information Criterion and Bayesian Information Criterion values. Three-way interactions between fixed effects were excluded if not significant at $p < .05$. Least squares means were compared at SPS = 1, 2 and 3 to reflect jogging, running and sprinting, respectively.

A linear mixed model was developed in SAS 9.4 (SAS Institute, Cary NC) for normalized speed according to instructions “jog”, “run” and “sprint”. Fixed effects were group and time, random effects were subject and subject*side. Model validity was confirmed and interactions between fixed effects were excluded if not significant at $p < .05$.

All trials were used for analysis of foot-strike. The mean for each speed group (jog, run, sprint) was used for analysis. Univariate linear modelling was undertaken for each speed group (jog, run, sprint; IBM® *SPSS*® version 24) for foot-strike using baseline foot-strike as a covariate and group as a fixed factor.

3. Results

3.1. Participants

Forty-three participants were recruited to the study. Four participants (GMFCS level II) were excluded from this analysis due to absence of a flight phase. One participant from the control group did not present for follow up gait data collection and two participants had incomplete data sets and were excluded. Baseline characteristics of the 36 participants included in this analysis are summarized in Table 1. All participants completed at least five jog trials, two participants completed only four run trials while all others did five or more. Three participants had difficulty following the sprint command, number of sprint trials ranged from two (one participant) to seven with a mode of three.

Table 1
Participant Characteristics at baseline.

	Control group	Intervention group
Age Mean(SD)	12y 10 m (2y 8 m)	12y 8 m (2y 8 m)
Gender n (%)	Male = 10	Male = 13
	Female = 8	Female = 5
Body Mass Index	21.6 (5.8)	19.8 (3.8)
GMFCS Level n	Level *I* = 10	Level *I* = 11
	Level II = 8	Level II = 7
CP Distribution n	Unilateral = 9	Unilateral = 8
	Bilateral = 9	Bilateral = 10
Gastrocnemius spasticity (n)	ASAS 0 = 8	ASAS 0 = 8
	ASAS 1 = 17	ASAS 1 = 11
	ASAS 2 = 11	ASAS 2 = 14
	ASAS 3 = 0	ASAS 3 = 3

SD = Standard Deviation; n = number; GMFCS = Gross Motor Function Classification System; y = years m = months; ASAS = Australian Spasticity Assessment Scale.

3.2. Normalized speed and foot-strike

From baseline to follow-up, both groups increased normalized speed of jogging (control $t = -3.6$ $p < .01$; intervention $t = -2.02$ $p = .04$), the intervention group increased normalized speed of running ($t = -3.68$ $p < .01$) while the control group got slower ($t = 3.17$ $p < .01$) and there was no change in normalized speed of sprinting in either group (control $p = .18$, intervention $p = .35$). At follow-up there was no difference in foot-strike pattern between groups, except in sprinting in GMFCS level II, in which the intervention group had shifted the foot-strike backward ($F = 4.70$ $p = .04$) (Fig. 1).

3.3. Jogging (SPS = 1)

At baseline there were no significant between-group differences.

3.3.1. GMFCS level I

From baseline to follow-up the control group increased A2 ($t = 4.80$ $p < .01$) and PS ($t = 3.60$ p $< .01$) while there was no change in the intervention group (A2 $t = 0.89$ $p = .37$; PS $t = 0.30$ $p = .77$). The intervention group decreased stride length ($t = 3.18$ $p < .01$) while the control group did not change ($t = 1.00$ $p = .32$). There was no change in H3 (control $t = 0.21$ $p = .83$; intervention $t = 0.39$ $p = .70$) or cadence (control $t = 0.56$ $p = .57$; intervention $t = 0.67$ $p = .50$) in either group. None of these within-group changes resulted in a significant between-group difference at follow-up (Fig. 2, Table 2).

3.3.2. GMFCS level II

From baseline to follow-up both groups increased A2 (control $t = 3.71$ $p < .01$; intervention $t = 3.96$ $p < .01$) and PS (control $t = 2.59$ $p = .01$; intervention $t = 4.46$ $p < .01$). The intervention group increased stride length ($t = 3.46$ $p < .01$) and decreased cadence ($t = 2.34$ $p = .02$) while the control group did not change (stride length $t = 0.07$ $p = .94$; cadence $t = 0.71$ $p = .48$; foot-strike $t = 0.44$ $p = .66$). None of these within-group changes resulted in a significant between-group difference at follow-up.

Neither group had statistically significant changes in H3 (control $t = 0.06$ $p = .95$; intervention $t = 1.63$ $p = .10$), however the small changes that did occur resulted in the control group having statistically greater H3 at follow-up ($t = 2.69$ $p = .02$) (Fig. 2, Table 2).

3.4. Running (SPS = 2)

At baseline there were no significant between-group differences.

3.4.1. GMFCS level I

From baseline to follow-up both groups had no change in A2 (control $t = 0.56$ $p = .58$; intervention $t = 1.16$ $p = .25$), increased H3 (control $t = 5.07$ $p < .01$; intervention $t = 5.81$ $p < .01$) and decreased PS (control $t = 2.84$ $p < .01$; intervention $t = 4.39$ $p < .01$). The intervention group decreased stride length ($t = 3.18$ $p < .01$) and increased cadence ($t = 5.98$ $p < .01$) while the control group did not change (stride length $t = 1.00$ $p = .32$; cadence $t = 1.44$ $p = .15$). None of these within-group changes resulted in a significant between-group difference at follow-up (Fig. 2, Table 2).

3.4.2. GMFCS level II

From baseline to follow up the intervention group increased A2 ($t = 2.49$ $p = .01$) while the control group did not change ($t = 0.38$ $p = .71$). The control group increased H3 from baseline to follow-up ($t = 4.84$ $p < .01$) while the intervention group did not change ($t = 1.66$ $p = .10$) which resulted in a significant between-group difference at follow-up ($t = 2.51$ $p = .03$). From baseline to follow-up the control group decreased PS ($t = 3.59$ $p < .01$) while the intervention group did not change ($t = 0.68$ $p = .50$). The intervention group increased stride length ($t = 3.46$ $p < .01$) while the control group did

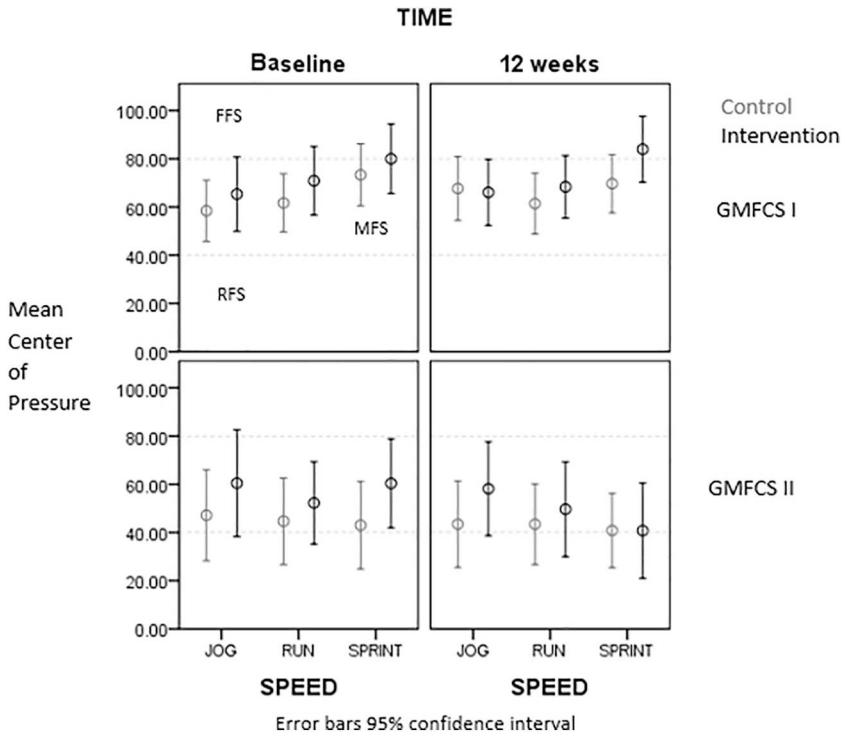


Fig. 1. Foot-strike by GMFCS level (I or II), Time (baseline or follow-up) and Group (Control or intervention).
GMFCS = Gross Motor Function Classification Scale; RFS = rear-foot strike; MFS = mid-foot strike; FFS = fore-foot strike.

not change (t = 0.08 p = .94). Within-group differences in A2, PS and stride length did not result in statistically significant between-group differences at follow-up. From baseline to follow-up the control group increased cadence (t = 3.44 p < .01) while the intervention group decreased cadence (t = 2.20 p = .03) which resulted in a significant between-group difference at follow-up (t = 3.00 p = .01) (Fig. 2, Table 2).

3.5. Sprinting (SPS = 3)

At baseline there were no significant between-group differences, except for GMFCS level II, in which the control group had greater cadence than the intervention group (t = 3.51 p < .01).

3.5.1. GMFCS level I

From baseline to follow-up the control group decreased A2 (t = 4.69 p < .01) while the intervention group did not change (t = 0.32 p = .75). Both groups increased H3 (control t = 5.50 p < .01; intervention t = 6.80 p < .01) and decreased PS (control t = 7.23 p < .01; intervention t = 5.14 p < .01). The intervention group decreased stride length (t = 3.18 p < .01) and increased cadence (t = 4.75 p < .01) while the control group did not change (stride length t = 1.00 p = .32; cadence t = 1.72 p = .09). None of these within-group changes resulted in a significant between-group difference at follow-up (Fig. 2, Table 2).

3.5.2. GMFCS level II

From baseline to follow-up the control group decreased A2 (t = 2.52 p = .01) while the intervention group did not change (t = 1.56 p = .12). Both groups increased H3 (control t = 3.75 p < .01; intervention t = 3.03 p < .01) and decreased PS (control t = 4.71 p < .01; intervention t = 3.70 p < .01). The intervention group increased stride length (t = 3.46 p < .01) while the control group did not change (stride length t = 0.07 p = .94). The control group increased cadence (t = 2.14 p = .03) while the intervention group did not change (t = 0.12 p = .91), which maintained the between-group difference at follow-up (t = 4.53 p < .01). None of the other within-group changes resulted in a significant between-group difference at follow-up (Fig. 2, Table 2).

4. Discussion

This randomized controlled trial found few systematic differences in A2, H3 or PS between groups following a 12-week intervention program targeting running technique in children and adolescents with CP. However, significant within-group differences were found which are interesting and worthy of discussion. Between-group differences at follow-up largely failed to reach statistical significance, which is probably the effect of the relatively small sample size and the heterogeneity of the CP population.

Both groups increased normalized speed in jogging, which may have been due to familiarization with the motion analysis laboratory. Only the intervention group increased normalized speed in running, which was related to increased A2 and increased stride length in GMFCS level II, but increased cadence in GMFCS level I. This supports previous reports that speed of running is increased firstly by increasing ankle power generation, and secondly by increasing hip power generation (Chappell et al., 2019). Furthermore, it highlights the different of needs of children in GMFCS levels I and II. Children in GMFCS level II have a greater deficit in ankle power generation than children in GMFCS level I and are unable to fully compensate using a proximal strategy (Bohm and Doderlein, 2012; Chappell et al., 2019; Davids et al., 1998). After intervention, children in GMFCS level II had reduced the distal deficit in A2, while children in GMFCS level I primarily strengthened their proximal compensation, shown by increased cadence. This leads us to recommend that in future interventions plantarflexor power exercises should form a larger portion of the program than hip flexor power exercises.

At follow-up the control group had increased distal deficit in sprinting, while the intervention group maintained A2, which suggests that the usual course of running in CP is one of increasing distal deficit and increasing proximal compensation, which the intervention was able

to mitigate to some extent, but not significantly so in the 12-week timeframe. This finding may be clinically important because it suggests that ankle power generation could be responsive to extended, targeted intervention. Improvement in running may therefore be possible via a mechanism of normalization of lower limb power generation, rather than strengthening a proximal compensatory pattern (Ishihara et al., 2015).

Power training has been reported to increase muscle fascicle length, muscle belly cross sectional area and velocity of movement in youth with CP (Moreau et al., 2013). Increased force production through muscle fiber hypertrophy or increased rate of force development and therefore increased ankle joint angular velocity at push off could

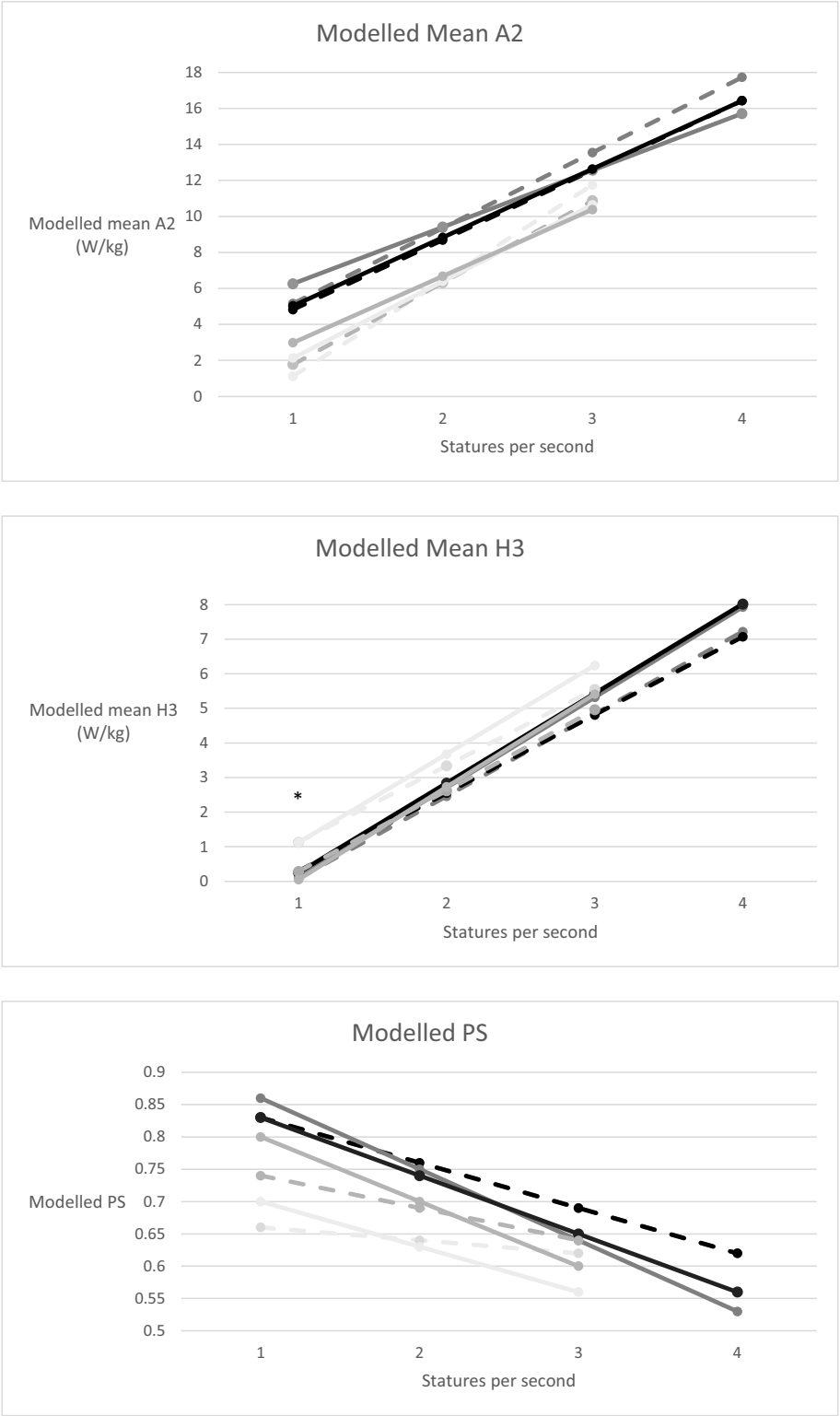
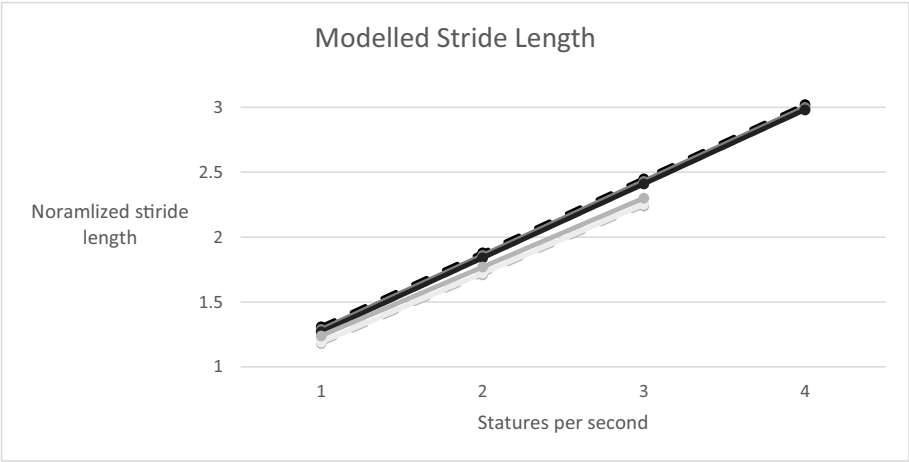


Fig. 2. Modelled mean a) ankle power generation (A2), b) hip power generation (H3) c) propulsion strategy (PS) d) normalized stride length and e) cadence (steps/s) by GMFCS level and normalized speed.
GMFCS = Gross Motor Function Classification Scale; * significant between-group difference at follow-up in GMFCS level II.



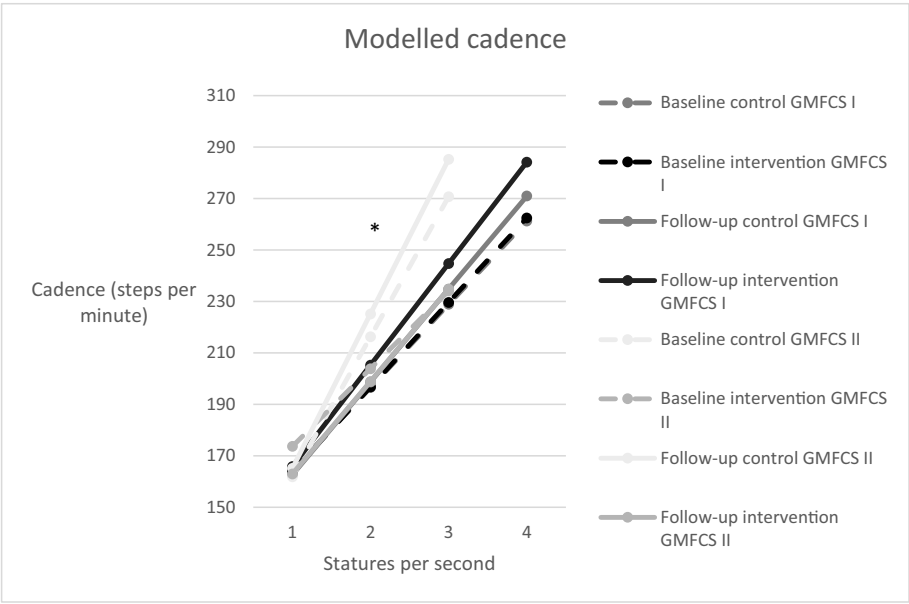


Fig. 2. (*continued*)

explain the increase in A2 in the intervention group. Alternatively, A2 may increase due to a larger ankle joint moment associated with a shift towards a forefoot strike pattern or if forward trunk lean is increased. Our results do not support a change in foot-strike being the mechanism for change in the intervention group. We did not collect trunk data, so cannot comment on the impact of vertical trunk alignment on ankle joint moment.

Propulsion strategy decreased in both groups during running and sprinting from baseline to follow-up, except for GMFCS level II in the intervention group, who had increased A2 at follow-up. For everyone

Table 2
Summary of significant within-group changes.

		Jogging		Running		Sprinting	
		Control	Intervention	Control	Intervention	Control	Intervention
Speed		↑	↑	↓	↑		
A2	GMFCS I	↑				↓	
	GMFCS II	↑	↑		↑	↓	
H3	GMFCS I			↑	↑	↑	↑
	GMFCS II		*	↑		↑	↑
PS	GMFCS I	↑		↓	↓	↓	↓
	GMFCS II	↑	↑	↓		↓	↓
Stride length	GMFCS I		↓		↓		↓
	GMFCS II		↑		↑		↑
Cadence	GMFCS I				↑		↑
	GMFCS II		↓	↑	↓ *	↑	

GMFCS = Gross Motor Function Classification Scale; A2 = ankle power generation; H3 = hip power generation; PS = propulsion strategy; ↑ = significant increase; ↓ = significant decrease; * = significant between-group difference.

else, the hip flexors were doing a greater proportion of work at follow-up compared to baseline, which supports our hypothesis that distal deficit (A2) is compensated by greater proximal power generation (H3). In GMFCS level I, in the intervention group, this was associated with increased cadence and decreased stride length, which is encouraged in the intervention to encourage a forefoot strike pattern, which we saw in sprinting but not running.

Sprinting, or maximal effort, is a different construct to jogging and running, in which the sprinter uses everything available run as fast as possible. While the intervention group maintained A2 in sprinting, H3 in sprinting was increased compared to baseline, so the PS was lower. It is possible that when asked for maximal effort the participants were able to recruit their improved hip power in swing but failed to recruit more ankle power at push-off, due to the high rate of force development demanded. As the intervention delivered in this study focused on running strategy and good form, sprinting repetitions were not a standard feature of the intervention and maximum speed avoided if it corresponded to a loss of good form. A longer program may have allowed the participants to progress to training sprinting thereby training the plantarflexors to generate maximal power more rapidly as is required for sprinting.

5. Limitations

This study aimed to report the effect of an intervention on running kinetics. This study did not address the physiological mechanisms of change due to the intervention, nor did it assess metabolic energy expenditure. These are avenues for further investigation. As there were low numbers of participants in this study, it was not possible to separate participants with bilateral and unilateral CP for analysis. Running mechanics and response to intervention could be different between these two groups. We acknowledge this as a limitation of the study. We have chosen to use statures to normalize velocity as bipedal gait is not a sequence of independent unilateral systems working alternatively, the system stores and transfers energy which impacts on the synergy of the bipedal system. Normalizing to statures allows the preservation of one speed for each steady-state run. Anthropometric scaling of velocity in bipedal running gait with asymmetries is an area for future research.

6. Conclusion

A 12-week running training program for children and adolescents with CP resulted in within-group differences in power generation at follow-up, which failed to reach statistical significance for between-group comparison. Within-group changes suggest that ankle power generation may be responsive to targeted intervention, especially in GMFCS level II, and that it may be possible to normalize the strategy for running propulsion in this population. Future intervention programs should consider a) including a larger proportion of ankle power generation exercises, b) including new or different exercises for ankle power generation, and c) delivering the intervention for a longer duration.

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Declaration of Competing Interest

The authors declare no conflicts of interest.

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Chapter 8 The effect of a running training intervention on leg stiffness in children and adolescents with cerebral palsy: a randomised controlled trial.

8.1 Preface

An examination of power generation for forward progression following the intervention program, which was presented in Chapter 7, showed that while running, participants in GMFCS Level II increased ankle power generation, but participants in GMFCS Level I had a different response to intervention. An analysis of baseline leg stiffness while running in this cohort, presented in Chapter 6, showed that leg stiffness profiles are different in children in GMFCS Level I compared to Level II. It was not clear whether this was relevant to the response of ankle power generation to intervention. This chapter describes the effect of a task-specific, low-load plyometric running training program on the function of the SSC through an examination of leg stiffness.

8.2 The effect of a running training intervention on leg stiffness in children and adolescents with cerebral palsy: a randomised controlled trial.

Abstract

Aim: To determine whether a running intervention utilising plyometric activities improved leg stiffness in youth with cerebral palsy (CP).

Method: This stratified randomised controlled trial examined the lower limb kinetics and kinematics of a sample of youths with CP during sub-maximal hopping and running, prior to and immediately following a 12-week running intervention that incorporated low-load plyometric training. Participants were divided into a control group (n=13; mean age 13 years 2 months [SD 2 years 7 months]; six males; nine GMFCS Level I; six unilateral) and an intervention group (n=18; mean age 12 years 9 months [SD 2 years 10 months]; 13 males; 11 GMFCS Level I; nine unilateral). Derived variables included three-dimensional leg stiffness as well as resultant ground reaction force and change in leg length. Generalised

linear mixed models were developed for statistical analysis.

Results: At follow-up, the intervention group had greater leg stiffness compared to the control group during submaximal hopping (p<0.01). At follow-up, participants in the intervention group in GMFCS Level I had greater leg stiffness than the control group during jogging (p=0.04).

Interpretation: A running training intervention that includes plyometric activities can improve leg stiffness in young people with CP, especially those in GMFCS Level I. Increased leg stiffness following intervention may be due to improved use of the plantarflexor stretch-shortening cycle.

Introduction

During running, the lower limb, acting in a spring-like fashion, has the capacity to shorten under loading and then lengthen to return energy[273]. The stiffness of the lower limb during the loading phase of running is modulated by both passive (biomechanical structures) and active elements (activated muscles). The interaction of passive and active subsystems is also mediated by sophisticated neurological control and motor learning. Since lower limb stiffness has been associated with performance and injury, the ability to optimise stiffness during running is an area of interest for many clinicians managing athletes and/or individuals with functional impairments[273, 274].

Stiffness can be calculated in different ways depending on the question to be answered and the underlying assumptions[273]. *Vertical stiffness* (K_{vert})is an appropriate measure for linear movements such as hopping or jumping [273] and is calculated by dividing the peak vertical ground reaction force (GRF) by the vertical displacement of the centre of mass (CoM) during loading[275]. *Leg stiffness* (K_{leg}) is commonly calculated during running and is the ratio between the GRF and the change in leg length (ΔL) during ground contact time[273]. During hopping, in which the direction of movement is only vertical, leg stiffness (K_{leg}) and vertical stiffness will have the same value[276]. Calculation of K_{leg} during running assumes the same spring-mass model of the lower limb as vertical stiffness but involves a greater number of variables due to the multiplanar nature of the movement[277].

In people with cerebral palsy (CP), altered muscle tone, muscle weakness or contracture may impact on K_{leg}. We previously reported that youth with CP in Gross Motor Function Classification Scale (GMFCS)[12] Levels I and II had K_{leg} profiles different to those of typically developing (TD) youth when running[278]. Youth with CP had lower K_{leg} than TD youth during jogging which we hypothesised was due to reduced ankle power generation at push-off.

Youth with CP achieved the same K_{leg} as TD youth at higher speeds by reducing knee flexion in loading phase (ΔL); however, this strategy limited maximum velocity. Knee flexion in loading phase is minimal at maximum running speed[58], which relates to the capacity of the lower limb to store and return energy to the system through use of the plantarflexor stretch-shortening cycle (SSC) to both hold the body up against gravity and propel the body forward.

In TD adults and adolescents, endurance running, power training and plyometric training have all been reported to increase K_{leg} during sub-maximal hopping, however these studies did not report whether K_{leg} was increased during running[117, 279, 280]. Running is a skill which requires motor control that is different to sub-maximal hopping, so it cannot be assumed that an improvement in one will result in an improvement in the other. Resisted sled-towing has been reported to decrease K_{leg} during sprinting in TD adolescents, due to increased ΔL[281]. Low-load plyometric training i.e. repetitive tasks at loads less than body weight, may therefore be more effective at optimising K_{leg} than loaded plyometric training in populations with low baseline K_{leg}[281, 282]. Changes in stiffness due to plyometric exercise are postulated to be due to training adaptations in both connective and muscular tissues[279, 283] and more effective use of the SSC[117]. It is unknown whether K_{leg} can be changed by intervention in people with CP, although musculotendinous architecture, which affects K_{leg}, has been reported to change in this population following strength training[284].

The aims of this study were to: 1) determine whether an intervention that includes low-load plyometric training in youth with CP would increase K_{leg} during sub-maximal hopping more than usual care; and 2) determine whether any increase in K_{leg} during sub-maximal hopping would transfer to the functional tasks of jogging, running or sprinting. We hypothesised that: 1) at follow-up, K_{leg} would be greater in the intervention group than the control group during sub-maximal hopping; 2) at follow-up, K_{leg} would be greater in the intervention group than the control group during jogging but no different in running and sprinting, as we have previously reported this to be the same as in the TD population[278].

Methods

Participants

Participants were aged 9-18 years, had CP, were able to walk 10m unaided and had a goal of improving their running. Participants were excluded if they had undergone musculoskeletal surgery in the previous six months, had contraindications to rigorous exercise, had a cognitive impairment preventing completion of assessments or had a

behavioural impairment precluding intervention in a group setting. Participants were stratified by age and mobility function and randomly allocated to control or intervention group by coin toss. The intervention group received 12 weeks of twice-weekly individualised intervention in a group setting (as previously described[190] but summarised below) while the control group received usual care. The study was approved by of the Child and Adolescent Health Ethics Committee, Perth, Western Australia (201405SEP) and Curtin University, Perth, Western Australia (HR 219/2014). The trial was prospectively registered with the Australian New Zealand Clinical Trials Notification ACTRN12614000467639. Informed consent was given by the parent/guardian.

Testing Procedures

Submaximal hopping data collection

K_{leg} was assessed during a sub-maximal hopping task performed on a custom-built leg sled (Figure 8.1) to facilitate the task by unloading the limb and reducing the balance demand[192]. Sub-maximal hopping on a leg sled has been reported to be a valid and reliable measure of plantarflexor SSC function[285]. An AMTI© force plate was installed at 90° to the base of the sled and connected to a laptop with Labview© program which recorded flight time and contact time. A counterweight was available to nullify the weight of the sled if this was required for the participant to successfully complete the task. If participants used the counterweight at baseline, they also used it at follow-up. Participants performed 20 consecutive one-legged hops on each leg at a self-selected frequency. Data were collected from both right and left legs.

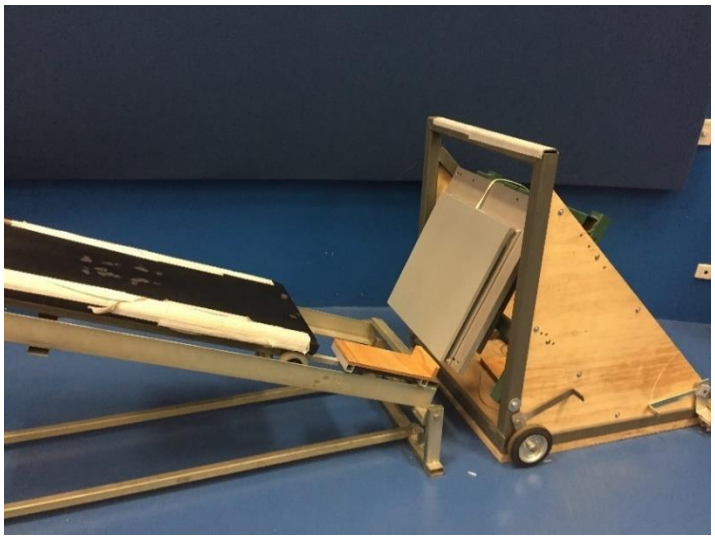


Figure 8.1 : Custom-built leg sled with force plate at 90° to the sled

Jogging, running and sprinting data collection

Reflective markers 10mm in diameter were placed on the skin using hypoallergenic double sided tape according to a modified Cleveland Clinic Foundation marker protocol[232]. A regression equation was used to calculate the hip joint centre[235]. Knee and ankle joint centres were calculated as the midpoint between the medial and lateral femoral condyles, and medial and lateral malleoli, respectively[236]. Inertial and geometric properties of the segments were based on previously published models[237, 238]. Kinematic data were recorded by an 18-camera motion capture system at 250Hz (Vicon T-series, Oxford Metrics, UK). Synchronised GRFs were collected at 1000Hz using three in-ground force platforms in series (AMTI, Watertown, MA).

All participants performed their trials barefoot. After a warm up, participants were asked to perform at least five trials at these speeds: 1) jog "like a warm up or like a jog around the oval at school"; 2) run "faster than jogging, but not your fastest" and 3) sprint "like you are in a race". Ten metres were available before and after the force plates to allow for acceleration and deceleration. A two-minute sitting break was permitted between speeds if required. Marker trajectories were labelled and filled using Vicon Nexus 2.5 (Vicon Motion Systems, Oxford, UK) and processed in Visual 3D™ version 6 (C-Motion, Inc.) using a previously published protocol[286].

Intervention

The individualised intervention was delivered by physiotherapists in a group setting at a community service provider and participants also received a home exercise program. The intervention has been described in more detail in a previously published paper[190], but in brief consisted of:

- Plyometric calf exercises (jumping, hopping, running) initially performed on a leg sled which was progressively raised to increase the proportion of bodyweight before transitioning to a small trampoline and then to overground
- Rapid hip flexion exercises progressed as above
- Task-specific exercises for running skill training

Participants attended twice weekly, for one hour each session, for 12 weeks (a total of 24 hours) and were given a supplementary home exercise program to complete twice per week.

Usual care

The control group received usual care over the intervention period. Usual care was not controlled but typically consisted of provision of a home exercise program and consultation on request.

Data processing

Submaximal hopping

Both legs of each participant were included for analysis. Ten consecutive hops in the middle of the 20 were chosen for analysis. K_{leg} was calculated as:

$$K_{leg} = \frac{m\pi(t_f + t_c)}{t_c^2(\frac{t_f + t_c}{\pi} - \frac{t_c}{4})}$$

where m=body mass, t_f=flight time and t_c=contact time[193]. Using this equation K_{leg} is expressed as Newtons per metre (Nm^{-1}).

Jogging, running and sprinting

All steps with a clean force plate strike, from both legs of all participants, were included for analysis. Normalised speed was defined as velocity of the pelvis, divided by height, and reported as statures per second (SPS). Resultant GRF (rGRF) was defined as the portion of GRF in line with the leg vector at minimum leg length (L_{min}), normalised to body weight (body mass*9.81). K_{leg} was calculated in dimensionless units by using rGRF and dividing change in leg length (ΔL) by initial leg length ($L_{initial}$):

$$K_{leg} = \frac{\frac{GRF_{Lmin}(in\ leg\ vector)}{mg}}{\frac{L_{IC} - L_{min}}{L_{initial}}}$$

where L_{IC} and L_{min} were the distance between the hip joint centre and the centre of pressure of the stance foot at initial contact and at minimum leg length, respectively [277] and $L_{initial}$ was the sum of the thigh segment length, shank segment length and the vertical distance between the ankle joint centre and the floor in quiet stance with foot flat (Figure 8.2)

Analysis

Statistical significance was set at $p<0.05$. A Bonferroni adjustment was considered as multiple variables were analysed. However, it has been recommended that analysis of kinematic and kinetic variables during running be undertaken with a minimum of 25

neurotypical adult participants and a minimum of 25 steps per participant at a self-self selected speed[246], while we had 21 participants with CP in each group. The chance of a type I error was deemed less likely than the chance of a type II error and for this reason no adjustment was applied. After Box-Cox transformation to correct right skewedness, a linear mixed model was developed in Statistical Analysis Software (SAS 9.4, SAS Institute Inc., Cary, NC, USA) for K_{leg} in submaximal hopping with fixed effect group*time and random effects subject and subject*side.

A Box-Cox transformation was used on running K_{leg} to correct right skewedness. Linear mixed models were developed in SAS for K_{leg} rGRF and ΔL with fixed effects group*time*SPS, and random effects subject and subject*side. The analysis of jogging, running and sprinting was done separately for each GMFCS level because of our previous findings that the two groups had different K_{leg} profiles [278]. Least squares means were compared at SPS=1, 2 and 3 to reflect jogging, running and sprinting, respectively.

Results

Participants

Forty-three participants were recruited to the study. Data included in the present study formed part of a larger body of work examining the effect of a running training intervention on the kinematics and kinetics of children and adolescents with CP. For the present study, data from 12 participants were excluded, seven participants had incomplete data sets (either hopping or running at either time point; five caused by a failure of force plate data to record to LabView and two by excessive marker occlusion), four participants did not have a flight phase (not able to run) and one (control group) did not attend follow-up. Included participants were 13 in the control group (mean age 13 years 2 months [SD 2 years 7 months]; six males; nine GMFCS Level I; six unilateral) and 18 in the intervention group (mean age 12 years 9 months [SD 2 years 10 months]; 13 males; 11 GMFCS Level I; nine unilateral). All included participants completed at least six jog trials. Five participants completed less than five run trials (minimum of three). The number of sprint trials ranged from two to seven with a mode of three.

Sub-maximal hopping

There was no between-group difference in K_{leg} at baseline (control median=1122Nm^{-1} LQ 874Nm^{-1} UQ 1821Nm^{-1}; intervention median=1491Nm^{-1} LQ 414 Nm^{-1} UQ 2883Nm^{-1}; t=0.10 p=0.92) (Figure 8.3). Four participants required a counterweight to successfully complete the task, three in the intervention group and one in the control group. At follow-up the

intervention group (median=3278Nm^{-1} LQ 1753Nm^{-1} UQ 4433Nm^{-1}) had twice the median K_{leg} compared to the control group (median=1556Nm^{-1} LQ 1127Nm^{-1} UQ 2077Nm^{-1}) during submaximal hopping (t=2.59; *p*=0.01) (Figure 8.3).

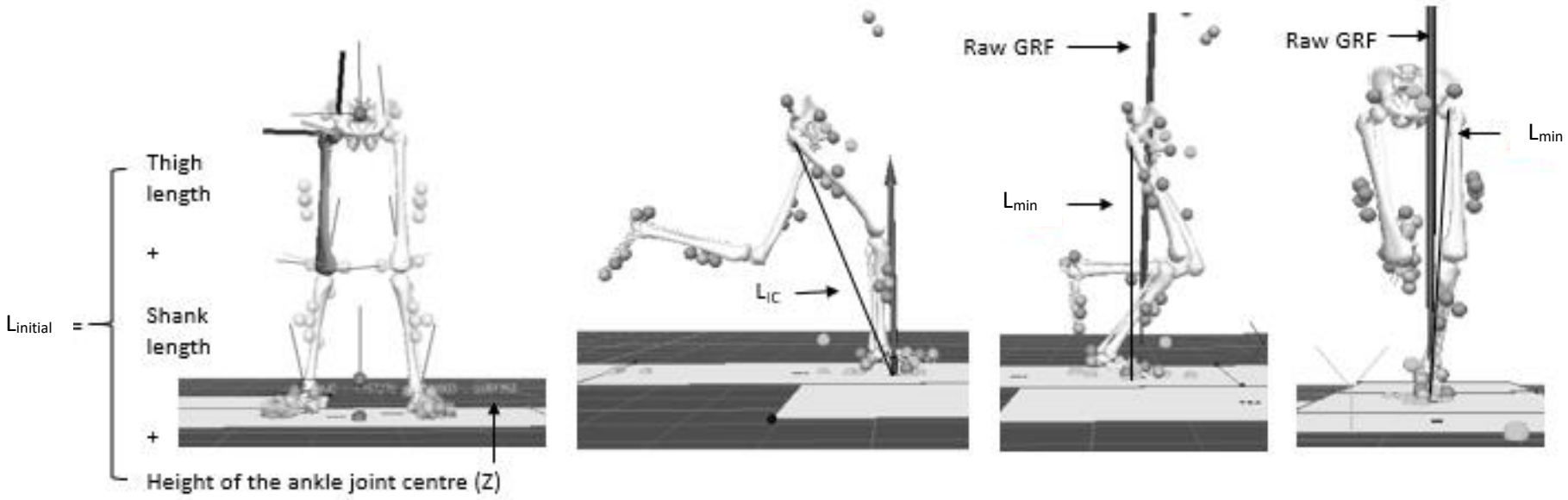


Figure 8.2: Calculation of $L_{initial}$, L_{IC} and L_{min}

a) definition of leg length is the distance between the centre of pressure and hip joint centre, b) leg length at initial contact (L_{IC}), c) minimum leg length (L_{min}) in the frontal and sagittal planes. Raw GRF is displayed here from which the portion of GRF in line with the leg vector at L_{min} is calculated.

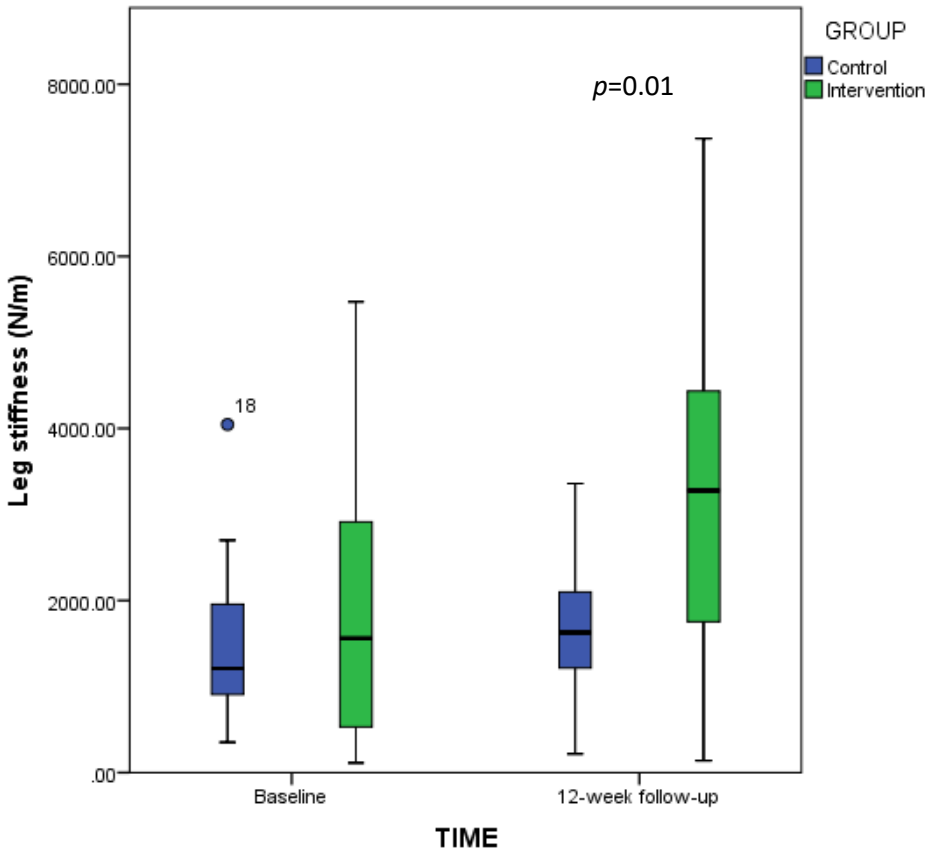


Figure 8.3: Leg stiffness in sub-maximal hopping: median and interquartile range with outliers

Jogging (SPS=1)

GMFCS Level I

There were no between-group differences at baseline for any of the derived variables. At follow-up, the intervention group had increased K_{leg} compared to the control group (*t*=2.61 *p*=0.01 *d*=0.5) (Table 8.1). There was no difference between groups in rGRF (t=0.21 *p*=0.84 *d*=0.2). ΔL was smaller in the intervention group than the control group (*t*=2.62 *p*=0.01 *d*=0.5) at follow-up (Table 8.1).

GMFCS Level II

From baseline to follow-up the control group increased K_{leg} (*p*=0.04 *t*=2.05), both groups increased rGRF (control *p*<0.01 *t*=5.08; intervention *p*=0.01 *t*=2.68) and the intervention group increased ΔL (*p*=0.01 *t*=2.58). Between-group differences were not significant at follow-up (K_{leg} *d*=0.6; rGRF *d*=0.2; ΔL *d*=0.6) (Figure 8.4).

Running (SPS=2)

GMFCS Level I

From baseline to follow-up the intervention group increased K_{leg} (p<0.01 t=4.72), the control group decreased rGRF (p=0.01 t=2.60) and both groups decreased ΔL (control p=0.01 t=2.56; intervention p<0.01 t=5.27). Between-group differences were not significant at follow-up (K_{leg} d=0.3; rGRF d=0.2; ΔL d=0.3)(Figure 8.4).

GMFCS Level II

From baseline to follow-up the control group increased Kleg (p<0.01 t=4.01), increased rGRF (p<0.01 t=2.97) and decreased ΔL (p=0.01 t=2.69). Between-group differences were not significant at follow-up (K_{leg} d=0.5; rGRF d=0.5; ΔL d=0.5)(Figure 8.4).

Sprinting (SPS=3)

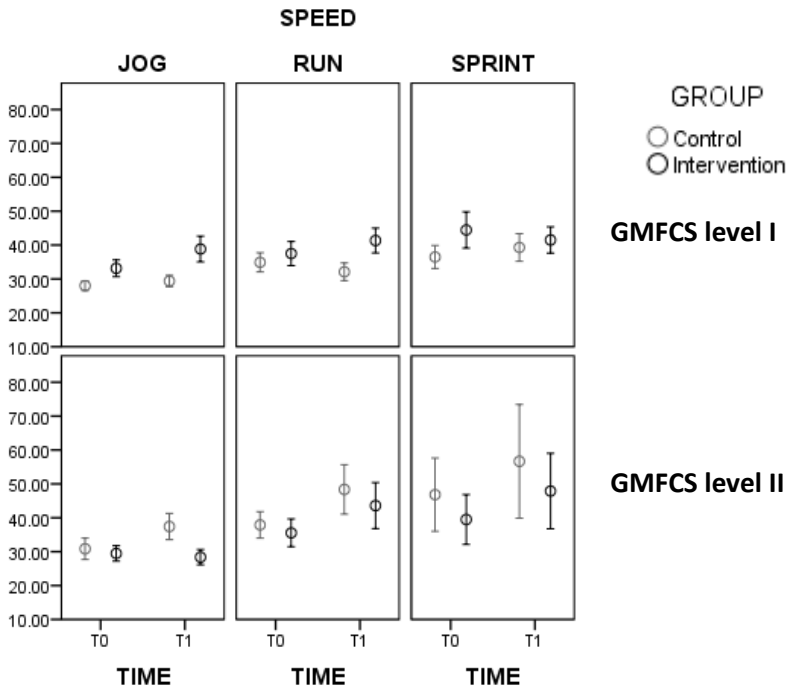
GMFCS Level I

From baseline to follow-up the control group decreased rGRF (p<0.01 t=5.12) and ΔL (p=0.01 t=2.50). Between-group differences were not significant at follow-up (K_{leg} d=0.2; rGRF d=0.0; ΔL d=0.1)(Figure 8.4).

GMFCS Level II

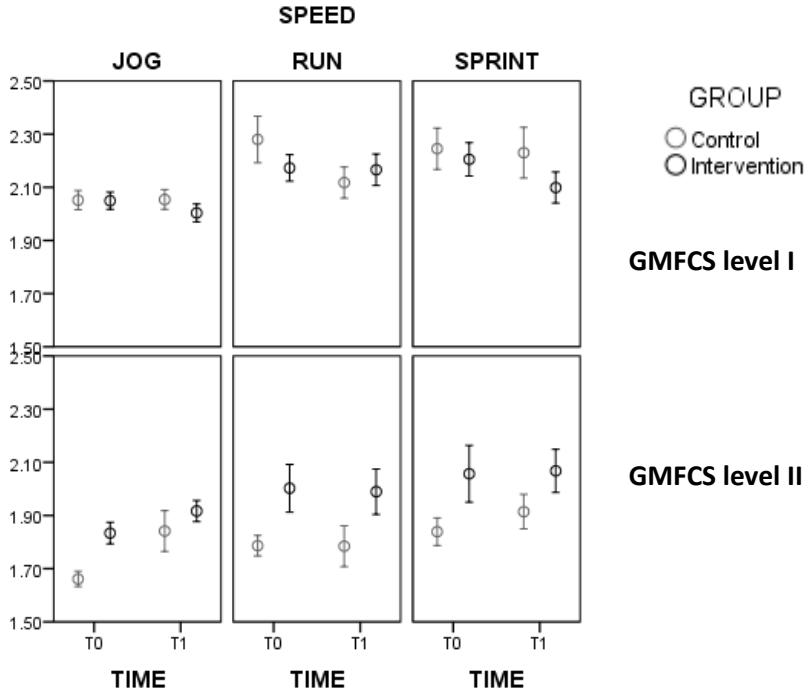
From baseline to follow-up there were no significant changes in either group. Between-group differences were not significant at follow-up (K_{leg} d=0.3; rGRF d=0.6; ΔL d=0.3)(Figure 8.4).

a) **Mean dimensionless leg stiffness**



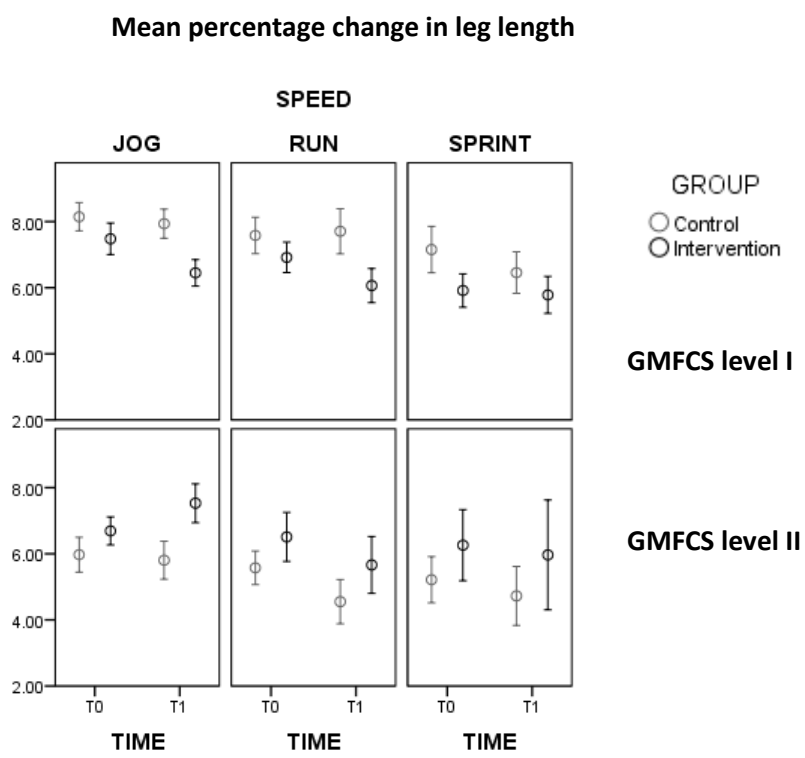
Error bars: 95% confidence interval

b) **Mean dimensionless rGRF**



Error bars: 95% confidence interval

c)



Error bars: 95% confidence interval

Figure 8.4: Means and 95% confidence intervals for a) leg stiffness (K_{leg}), b) resultant ground reaction force (rGRF) and c) percentage leg length change (ΔL) at baseline and follow-up by group (intervention or control)

Table 8.1: Group averages for each running-related variable by GMFCS Level (I, II or TD)

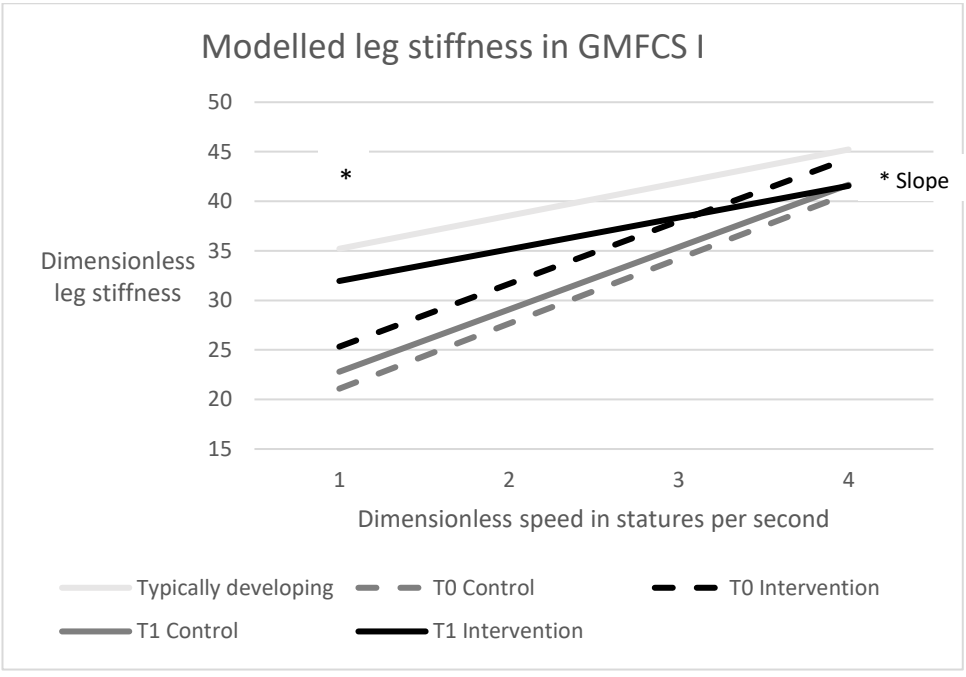
Variable	GMFCS	Group	JOG Mean (SD)		RUN Mean (SD)		SPRINT Mean (SD)	
Time			Baseline	12-wks	Baseline	12-wks	Baseline	12-wks
Speed (SPS)	GMFCS I	Control	1.57 (0.25)	1.64 (0.23)	2.35 (0.41)	2.19 (0.43)	2.89 (0.51)	2.79 (0.44)
		Intervention	1.59 (0.25)	1.57 (0.28)	2.31 (0.31)	2.29 (0.39)	2.96 (0.49)	2.77 (0.46)
	GMFCS II	Control	1.35 (0.19)	1.40 (0.19)	2.01 (0.29)	1.89 (0.25)	2.33 (0.30)	2.35 (0.24)
		Intervention	1.45 (0.20)	1.62 (0.24)	2.06 (0.36)	2.12 (0.46)	2.65 (0.30)	2.79 (0.20)
	Total	Control	1.52 (0.25)	1.57 (0.24)	2.22 (0.41)	2.10 (0.40)	2.69 (0.52)	2.66 (0.44)
		Intervention	1.55 (0.24)	1.59 (0.27)	2.24 (0.34)	2.23 (0.42)	2.88 (0.47)	2.78 (0.40)
Leg Stiffness	GMFCS I	Control	28.02 (9.52)	29.38 (11.11)	34.91 (15.82)	32.11 (12.87)	36.47 (14.77)	39.31 (15.70)
		Intervention	33.14 (16.00)	38.84 (25.55) *	37.51 (20.04)	41.34 (17.19)	44.45 (23.60)	41.51 (15.58)
	GMFCS II	Control	30.86 (11.12)	37.41 (16.32)	37.89 (16.89)	48.36 (23.70)	46.83 (33.82)	56.66 (42.40)
		Intervention	29.47 (9.38)	28.39 (9.86)	35.55 (14.09)	43.60 (22.10)	39.50 (18.25)	47.89 (25.09)
	Total	Control	28.67 (9.96)	31.79 (13.37)	36.04 (16.25)	37.14 (18.49)	40.08 (23.61)	44.69 (27.89)
		Intervention	32.09 (14.49)	35.68 (22.52)	36.96 (18.56)	42.08 (18.89)	43.20 (22.39)	43.14 (18.53)
Resultant GRF	GMFCS I	Control	2.05 (0.24)	2.05 (0.24)	2.28 (0.49)	2.12 (0.29)	2.25 (0.34)	2.23 (0.37)
		Intervention	2.05 (0.21)	2.00 (0.23)	2.17 (0.28)	2.17 (0.28)	2.21 (0.28)	2.10 (0.23)
	GMFCS II	Control	1.66 (0.10)	1.84 (0.32)	1.79 (0.17)	1.78 (0.25)	1.84 (0.16)	1.91 (0.16)
		Intervention	1.83 (0.17)	1.92 (0.18)	2.00 (0.31)	1.99 (0.28)	2.06 (0.26)	2.07 (0.18)
	Total Total	Control	1.96 (0.27)	1.99 (0.29)	2.09 (0.47)	2.01 (0.32)	2.10 (0.35)	2.13 (0.35)
		Intervention	1.99 (0.22)	1.98 (0.22)	2.13 (0.30)	2.11 (0.29)	2.17 (0.28)	2.09 (0.22)
% change in leg length	GMFCS I	Control	8.15 (2.83)	7.94 (2.88)	7.58 (3.07)	7.71 (3.36)	7.15 (3.05)	6.46 (2.43)
		Intervention	7.48	6.45 (2.71)	6.92	6.07	5.92	5.78

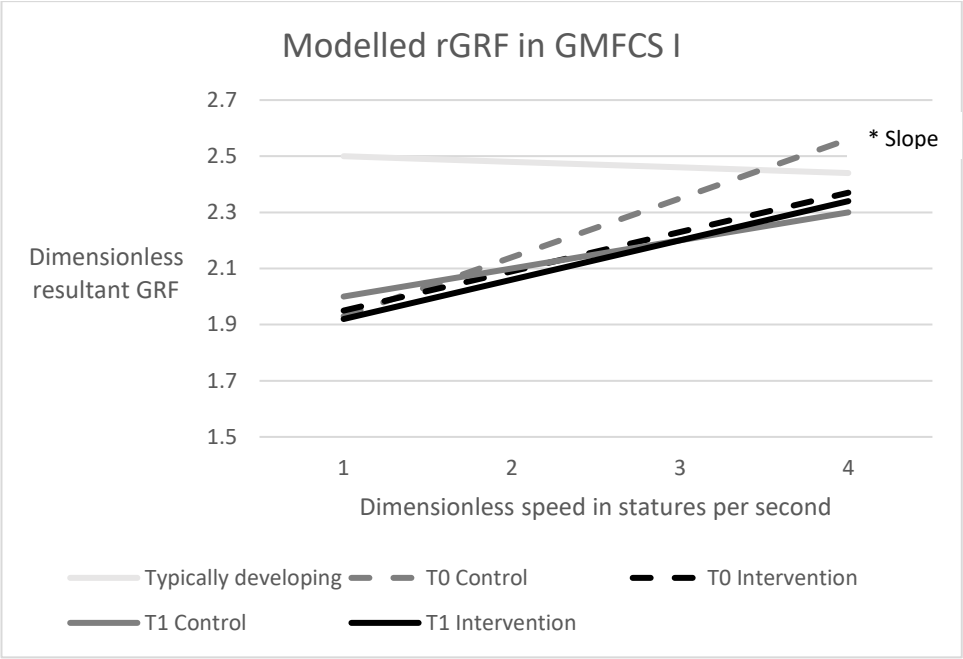
			(3.12)	*	(2.59)	(2.43)	(2.22)	(2.23)
	GMFCS II	Control	5.97 (1.87)	5.80 (2.43)	5.58 (2.18)	4.55 (2.17)	5.21 (2.19)	4.72 (2.25)
		Intervention	6.69 (1.73)	7.53 (2.58)	6.51 (2.55)	5.66 (2.78)	6.26 (2.66)	5.96 (3.74)
	Total Total	Control	7.65 (2.79)	7.30 (2.92)	6.82 (2.93)	6.73 (3.37)	6.48 (2.92)	5.92 (2.50)
		Intervention	7.25 (2.81)	6.77 (2.71)	6.80 (2.58)	5.93 (2.55)	6.00 (2.33)	5.83 (2.67)

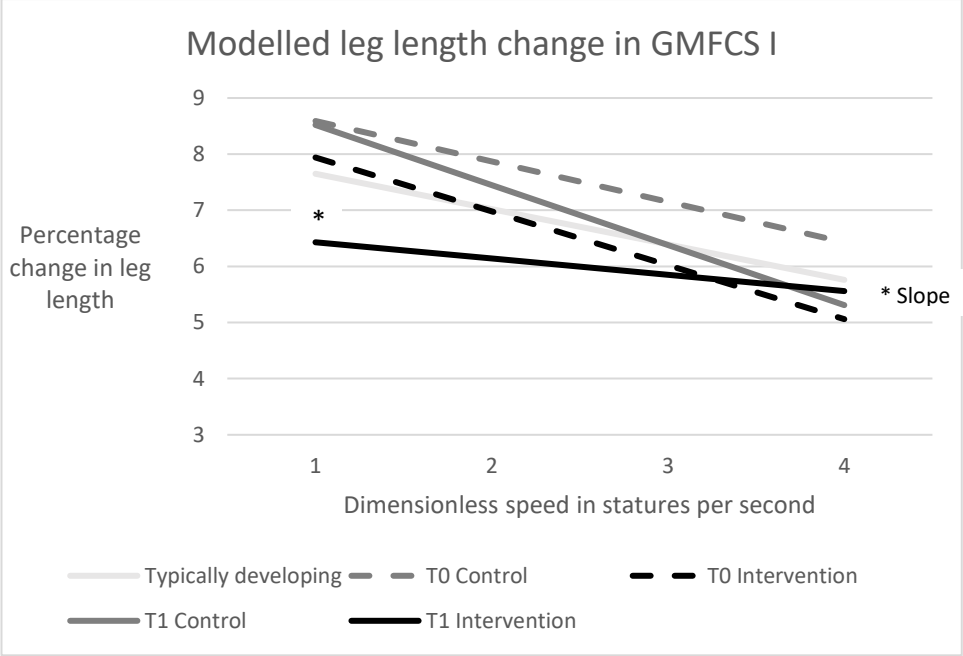
GRF=Ground reaction force; %=percentage; °=degrees; SD=standard deviation; GMFCS=Gross Motor Function Classification Scale; *=significant between-group difference at $p<0.05$

Relationship of Kleg, rGRF and ΔL to speed

At baseline in GMFCS Level I, the control group increased rGRF more rapidly with increasing speed compared to the intervention group and compared to follow-up ($p<0.01$). At follow-up in GMFCS Level I, the intervention group increased K_{leg} more slowly ($p\leq0.03$) and decreased ΔL more slowly with increasing speed compared to the control group and compared to baseline ($p\leq0.01$; Figure 8.5). At baseline in GMFCS Level II, the intervention group decreased ΔL more slowly with increasing speed compared to the control group and compared to follow-up ($p=0.02$) (Figure 8.6).

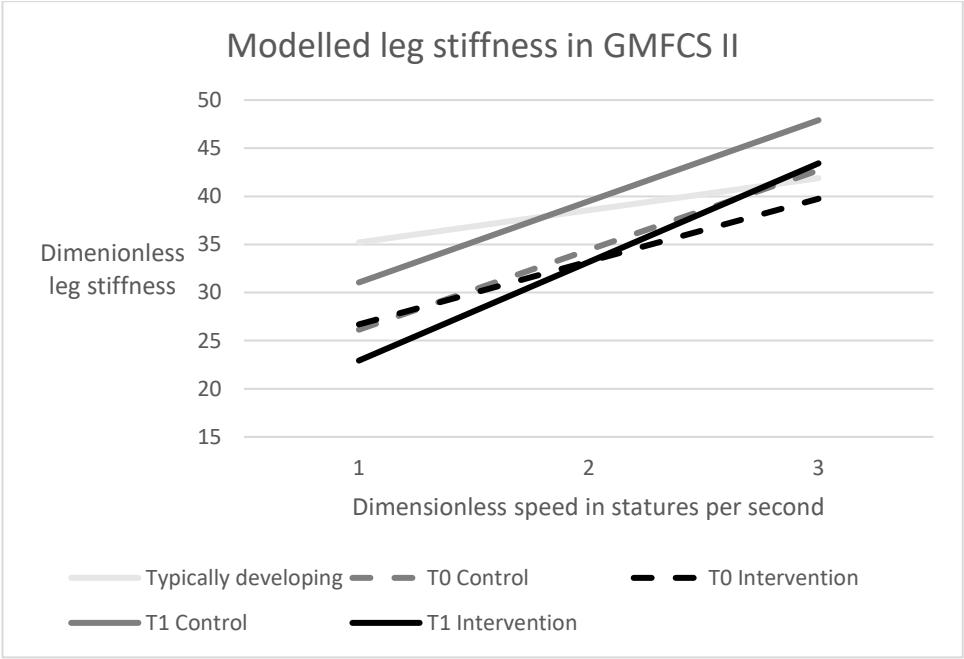


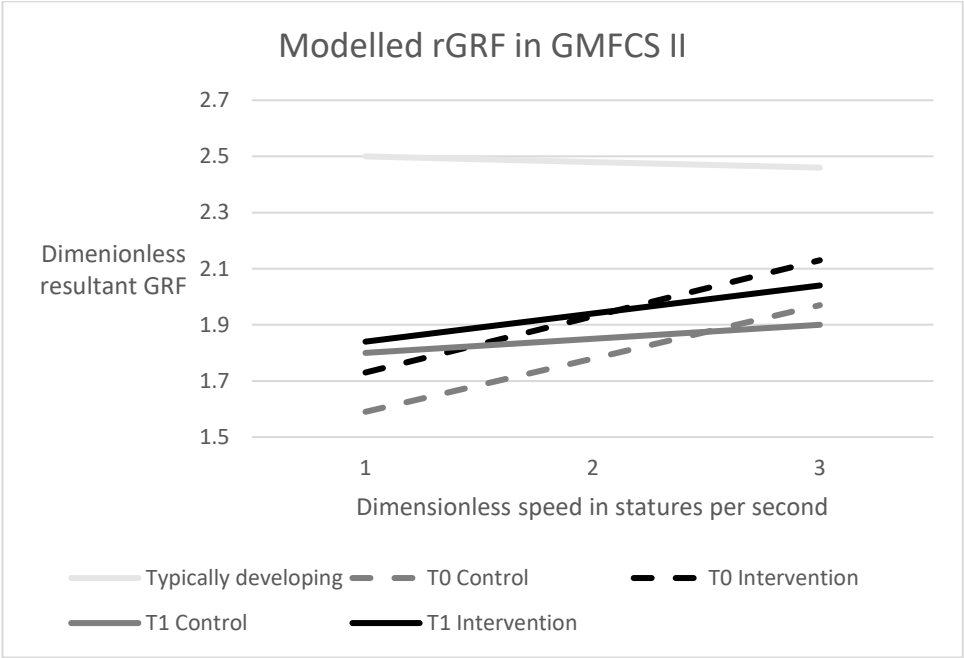


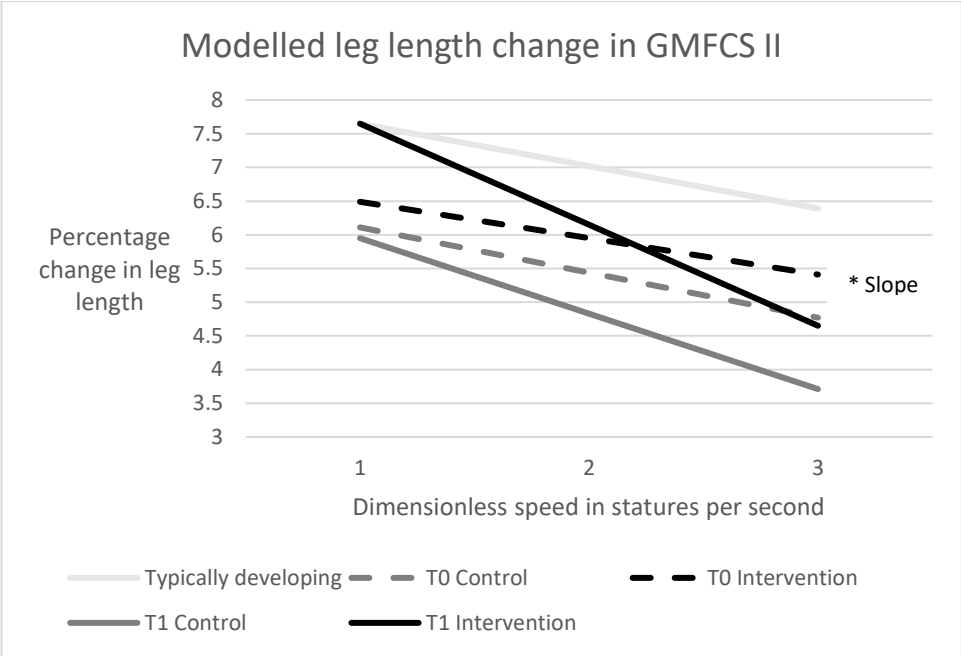


* = significant between-group difference at follow-up $p<0.05$; GRF=ground reaction force; GMFCS=Gross Motor Function Classification Scale NB: Typically developing values have been previously reported and are given here for reference[278].

Figure 8.5: Leg stiffness (k_{leg}), resultant ground reaction force (rGRF) and percentage leg length change (ΔL) - baseline and follow-up for CP children at GMFCS Level I with typically developing values for reference


Modelled leg stiffness in GMFCS II
Dimenionless leg stiffness
50
45
40
35
30
25
20
15
1
2
3
Dimensionless speed in statures per second
Typically developing
T0 Control
T0 Intervention
T1 Control
T1 Intervention


Modelled rGRF in GMFCS II
Dimenionless resultant GRF
2.7
2.5
2.3
2.1
1.9
1.7
1.5
1
2
3
Dimensionless speed in statures per second
Typically developing
T0 Control
T0 Intervention
T1 Control
T1 Intervention



* = significant between-group difference at follow-up p<0.05; GRF=ground reaction force; GMFCS=Gross Motor Function Classification Scale NB: Typically developing values have been previously reported and are given here for reference[278].

Figure 8.6: Leg stiffness (k_{leg}), resultant ground reaction force (rGRF) and percentage leg length change (ΔL) - baseline and follow-up for children with CP GMFCS Level II with typically developing values for reference

Discussion

The main finding of this study was that a low-load plyometric intervention to improve running increased K_{leg} during sub-maximal hopping in youth with CP, GMFCS Levels I and II. Modulation of K_{leg} is complex, involving supra-spinal, longer-latency responses, short-latency stretch reflex potentiation, and feed-forward muscle activity[287, 288]. Effective modulation of K_{leg} is a marker of neurological optimisation matching the mechanical properties of the lower limb[287, 288], therefore specific training of loading response in a cyclic loading model (bouncing with pre-planned eccentric actions) is fundamental to an intervention to rehabilitate neuromodulation. Our intervention included repetitive bouncing activities (alternating feet, hopping) which were progressed by increasing the percentage of body weight load (inclined leg sled to trampoline to overground)[190]. These exercises appear to have facilitated improved regulation of K_{leg} on landing to use the SSC more effectively.

We were also interested if the ability to modulate stiffness learned from the plyometric exercises transferred into different running speeds. The second main finding of this study was that following the intervention, youth classified as GMFCS Level I had increased K_{leg}

during jogging. This was associated with decreased ΔL to optimally match low rGRF. We previously reported that youth with CP had low rGRF which was associated with a 'terrestrial' running pattern[105, 278]. This pattern is associated with a deficit in ankle power generation at push-off[289], resulting in a short flight phase with reduced upward displacement of the centre of mass, and therefore a low rGRF[278]. At baseline, youth in GMFCS Level I had a relatively large ΔL considering rGRF was low. Following intervention, they appeared to have learned to use a pattern with reduced knee flexion in stance (i.e. reduced ΔL) to facilitate improved function of the eccentric phase of the SSC.

Youth in GMFCS Level II also have a low rGRF with an even greater deficit in ankle power generation compared to youth in GMFCS Level I[289]. However, they have a small ΔL compared to both TD children and youth in GMFCS Level I, which is probably because they have a shorter stride length and relatively high cadence[125]. Further, they rely on increasing cadence to increase running velocity rather than increasing stride length like their TD peers[125]. Higher cadence requires the runner to get on and off the stance foot quickly, which is facilitated by limiting knee flexion in stance (i.e. ΔL)[58]. Following the intervention, youth in GMFCS Level II had increased ΔL in jogging. Although the between-group difference was not significant, the effect size was moderate. Increased ΔL did not result in a change in K_{leg} because rGRF also increased, as it did also in the control group. These findings are encouraging, as they show a trend towards improved control of knee flexion in stance, which is the primary modulator of K_{leg} during running[290]. Youth in GMFCS Level II are more likely than those in Level I to have impairments at the knee which impact running gait[15] and impact the function of the plantarflexor SSC during running. It is possible that youth in GMFCS Level II need longer than 12 weeks of intervention to improve modulation of K_{leg} in running.

In a simple spring model where the ankle and knee are considered as two springs working in series, overall K_{leg} is greatest when both joints have optimal stiffness. In the same way, power output is greatest when power is transferred efficiently along the kinetic chain. When stiffness is too low this can cause a phase lag that disrupts the timing of the stretch reflexes and the GRF acting on the CoM[287, 288]. When stiffness is too high, reflex activity is triggered early and the timing of the GRF acting on the CoM is again disrupted[287, 288]. Therefore, an intervention should aim to stiffen joints that are too compliant but also enhance controlled movement in joints that are too stiff. If an individual can learn through training to synchronise the neurological drive with muscular activity to optimise K_{leg} then athletic performance will be optimised[273]. Our findings suggest that a low-load plyometric

intervention for running for youth in GMFCS Level II needs to focus specifically on increasing compliance at the knee as well as increasing ankle stiffness, while youth in GMFCS Level I should focus on increasing both knee and ankle stiffness. This requires further investigation in a larger sample.

A primary limitation of this study is the absence of EMG data, meaning we are not able to determine whether co-contraction, muscle activation prior to landing, or coordination of reflex activity was the reason for improved K_{leg} in the intervention group. This is an avenue for further investigation. Our findings are only applicable to children with CP who can run barefoot, and the sample size is small given that we divided the group into GMFCS levels I and II. This may have resulted in a type II error, meaning that we were under-powered to detect a true between-group difference at follow-up. Repeating the study with larger numbers of participants will allow more definite conclusions to be reached. Future improvements in musculoskeletal modelling may allow for more accurate estimations of K_{leg} and improved power to detect change in smaller samples.

Conflict of interest statement

The authors declare no conflict of interest. This study was supported by a Non-Government Centre Support Grant and Perth Children's Hospital Foundation Grant ID 9632. Annie Chappell is the recipient of an Australian Government Research Training Program Scholarship.

Chapter 9 General discussion, limitations, and future directions

9.1 Discussion

In clinical practice, children and adolescents with CP, GMFCS Levels I and II, can run[15] and have goals to improve their running. Running is a fundamental movement skill[37] which is used in everyday activities and facilitates participation in play, sport and recreational activities[35]. Skilled running requires high force production during a rapid, cyclical movement[52]. Therefore, improving lower limb power generation, including the function of the stretch-shortening cycle, should in theory translate into improved running performance and thereby into improved participation in physical activity. There is a paucity of published evidence on interventions specific to improving the skill of running in youth with CP. This body of work described the running of youth with CP and evaluated the effectiveness of a task-specific, low-load plyometric running training program in improving power generation and leg stiffness in running in children and adolescents with CP.

A systematic review of the literature related to running in people with CP was undertaken, as such a review had not been identified in the literature. The review had two aims: 1) to review what is known about how people with CP run, and 2) to review what is known about the effect of interventions on improving running in people with CP. The systematic review was published and subsequently updated. The review established the growing interest in running in people with CP in the context of participation and function. There were few papers reporting the kinematics and kinetics of running in people with CP and no information regarding the effect of intervention on the biomechanics of running in recreational runners with CP. The updated systematic review identified increased interest in running in people with CP; however, there remains relatively little information in the research literature regarding the biomechanics of running in people with CP and whether these can be improved with intervention. This doctoral work is therefore the first to examine the effect of a task-specific, low-load plyometric training intervention on the biomechanics of running in children and adolescents with CP.

A randomised controlled trial was undertaken, and the outcomes related to function and participation were reported in a paper on which the doctoral candidate was a co-

investigator (Appendix 1). This study was the first to use the HiMAT[191] in children with CP, and to evaluate a skill-based running program in this population. The study reported that the intervention was effective for the achievement of running goals and to improve participation in school activities but did not report concurrent changes in running biomechanics.

Extending the outcomes of the RCT beyond the function and participation outcomes was required to understand the mechanisms by which changes are made to function. This is important to inform and optimise interventions to improve running skill. As power generation is reported to be critical for the emergence of independent running skill[93] and is directly related to running speed[56], determining the potential to improve power generation during running is important for training running skill in children and adolescents with CP, and was the focus of this doctoral work.

Firstly, it was important to understand how running biomechanics in children with CP differ from those who are typically developing. Such understanding allows for the creation of a problem list, identifying and prioritising potential goals of intervention. Secondly, an analysis of changes made through intervention provides clinicians with information regarding the potential for normalising running gait. If normalisation of running biomechanics is unlikely, clinicians may instead choose to strengthen effective compensation strategies, while being mindful of the potential for abnormal tissue stresses to contribute to injury[56]. However, if normalisation of running biomechanics in children with CP is possible, this should be the goal of intervention. Such information was not available in the research literature prior to this study. Therefore, an analysis of the biomechanics of running in children with CP was undertaken utilising the structure of the randomised controlled trial to report on the effect of a low-load plyometric intervention on the derived variables of interest. This work has been reported in Chapters 5, 6, 7 and 8. The analysis had a focus on power generation and the function of the stretch shortening cycle due to the importance of power generation strategies for mechanical efficiency[68]. Using the stretch shortening cycle during running reduces the metabolic energy requirement by harnessing elastic energy to augment power produced by muscle contraction[82]. Running economy is a predictor of running performance[68], and therefore power generation and the function of the stretch shortening cycle can be considered fundamental elements of skilled running.

The use of 3D motion analysis is the gold standard for analysis of power generation but has

potential for error, especially through the representation of the body by a mathematical model. It was advisable to establish a method which would reduce modelling error by realistically modelling joint constraints in children with CP, thus maximising precision, as the CP population is heterogenous, with a high degree of intra-participant variability[12, 23]. A published paper from this thesis (Chapter 4.3) reported that customised joint translation boundaries specified in an inverse kinematic model can reduce the intra-participant variability of kinematic and kinetic data compared to a model without joint constraint boundaries, which may reflect improved precision. Furthermore, a known-groups analysis utilising functional groups GMFCS Level I and GMFCS Level II indicated that the reduction of intra-participant variability was not the result of removing biological variability, but instead due to the reduction of error from the data-processing workflow. These findings also indicate that the observed kinetics of running in people with CP aged 9-18 years validate the use of GMFCS levels I and II as a classification of gross motor function. This is the first time an examination of specified 3D joint translation constraints was applied to running gait in a cohort of people with CP. The findings may be applied to future investigations of running in people with CP.

The biomechanical model recommended in the Section 4.3 was then utilised to process biomechanical data used in the calculation of joint power generation in the lower limb during running in children and adolescents with CP. The fundamental requirement for running is the ability to generate enough power to launch the body into a flight phase, which is achieved primarily by the ankle plantarflexors[122]. Early increases in running speed are likewise achieved primarily through an increase in power generation by the ankle plantarflexors[83]. The ankle plantarflexors generate power through both muscle contraction and through the mechanism of the stretch shortening cycle, which improves efficiency[82]. Therefore, an analysis of ankle power generation and the function of the stretch shortening cycle during running were undertaken using baseline data. This analysis, detailed in two papers (one published and one prepared for publication), is novel in the context of the CP population.

The first study of baseline data, presented in Chapter 5, found that children and adolescents with CP generated less ankle power than typically developing children, which was more pronounced in GMFCS Level II than Level I. At slower speeds, youth with CP used a proximal strategy to compensate for a distal deficit (i.e. reduced ankle power generation), by increasing hip flexor power in swing. This strategy was reflected in an increased cadence

and reduced stride length. This strategy is normally employed by people who are neurotypical only after optimum stride length is reached[240]. Youth with CP therefore used a normal strategy to increase velocity but used it too early in their speed range. This interpretation infers that an ankle power deficit limited maximum running velocity.

The second study of baseline data, presented in Chapter 6, found that children and adolescents with CP had reduced leg stiffness when jogging and running compared to typically developing peers, but that leg stiffness increased rapidly with increasing speed. The underlying mechanisms were found to be different in GMFCS Levels I and II. Children in GMFCS Level I had reduced ankle power generation at push-off compared to typically developing children, which would reduce peak ground reaction force in stance. This was magnified in children in GMFCS Level II, who had a greater ankle power generation deficit than children in GMFCS Level I. A normal level of leg stiffness could have been achieved by matching the ground reaction force with an appropriate amount of leg shortening, which primarily occurs as knee flexion[290]. Youth in GMFCS Level I had excessive leg shortening on landing while jogging, which resulted in low leg stiffness, but were able to achieve normal leg stiffness when running and sprinting. Youth in GMFCS Level II had more difficulty matching leg shortening to the ground reaction force. They had excessive leg shortening in jogging which resulted in low leg stiffness, but rapidly reduced leg shortening on landing with increasing speed. Youth with CP in GMFCS Level II therefore used a normal strategy to accommodate increased velocity but used too much, too early in their speed range. It appears that sprinting speed was limited in GMFCS Level II because minimum leg shortening was reached at slower speeds compared to GMFCS Level I or typically developing youth.

Overall, results of the studies presented in Chapters 5 and 6 support previous reports that running function is better in children in GMFCS Level I than those in Level II[9] and suggest that this may be related to the extent of deficit in ankle power generation. This deficit may be due in part to an interruption of the mechanism of the stretch-shortening cycle, as reflected by leg stiffness. Leg stiffness is measured in the weight-acceptance phase of stance, which corresponds to the eccentric phase of the stretch-shortening cycle. Low leg stiffness suggests that energy is being dissipated through excessive joint flexion, rather than recycled via tendon recoil. Loss of elastic energy may result in decreased power generation at push-off. Decreased power generation at push-off may result in reduced height of the flight phase and reduced peak GRF in the next stance phase. Low leg stiffness and a deficit

in ankle power generation are therefore inter-related. Children in both GMFCS Level I and Level II used normal strategies to increase speed – increasing hip flexor power and increasing leg stiffness – but used the strategies at much slower speeds than would be expected in the TD population.

The RCT was undertaken to examine the effect of a task-specific, low-load plyometric running intervention on children and adolescents with CP. The intervention sought to improve running goal attainment, running biomechanics and participation. The first paper, which is included as Appendix 1, reported that the intervention group had better running-related goal attainment and better school participation compared to the control group at follow-up. In the biomechanical domain, there were few between-group differences at follow-up. Youth in GMFCS Level I in the intervention group responded differently to those in GMFCS Level II in the intervention group. Children classified as GMFCS Level II are more likely to have bilateral impairment and more likely to have gait impairment affecting the knee than those in GMFCS Level I[291, 292]. These differences mean that, in broad terms, youth in GMFCS Level II may have more complex running gait impairments to be addressed by intervention, compared to those in Level I.

The two studies of biomechanical data, presented in Chapters 7 and 8, found that participants in GMFCS Levels I and II in the intervention group increased leg stiffness during submaximal hopping by 100% and improved running speed. During jogging, children in the intervention group in GMFCS Level I had improved leg stiffness due to a reduction in excessive change in leg length. For participants in GMFCS Level I, the intervention had no significant effect on ankle power generation, hip power generation or propulsion strategy, and effect sizes were small. These results suggest that participants in GMFCS Level I may have learned to use the stretch-shortening cycle more effectively during jogging, which should improve efficiency. In other words, it suggests that while the total ankle power generated was the same, a larger proportion was elastic energy provided by recoil of the Achilles tendon, thus reducing metabolic energy consumption. As we did not measure oxygen consumption, the evaluation of metabolic efficiency before and after intervention is an avenue for further investigation.

For youth in GMFCS Level II, the intervention resulted in reduced hip power generation and reduced cadence compared to the control group, with large effect sizes. The intervention resulted in no significant between-group effects in ankle power generation or propulsion strategy, although effect sizes for propulsion strategy were moderate (SPS=1 *d*=0.8; SPS=2

d=0.6; SPS=3 *d*=0.3). The reduction in hip power generation at the same speed suggests a reduction in proximal compensation and a normalisation of power generation strategy in the intervention group. Although the between-group difference was not significant, participants in GMFCS Level II in the intervention group did have a significant within-group increase in ankle power generation during running, which was not seen in the control group. Participants in GMFCS Level II also had greater rGRF than the control group at follow-up, which was not statistically significant but had moderate effect size for jogging and running. Again, change in leg length was greater (closer to typically developing values) in the intervention group at follow-up, which while not statistically significant had moderate effect size for jogging and running. These observations support the hypothesis that the distal deficit is starting to be corrected with a corresponding reduction in proximal compensation. Lack of statistically significant between-group findings, despite the differences in within-group findings and some moderate effect sizes, suggest the study may have been under-powered for changes in biomechanical variables. A larger, multi-centre trial could clarify our understanding of the mechanisms by which running is improved by intervention in people with CP.

Overall, these results suggest that running improvement in children and adolescents with CP relies on a complex interaction that includes both increasing ankle power generation and the optimisation of leg stiffness. The capacity for ankle power generation needs to be increased through a hierarchy of ankle power generation activities, such as bounding on a leg sled with progression to a trampette. Concomitantly, learned optimisation of the eccentric phase of the stretch-shortening cycle with appropriate leg shortening (i.e. knee control) through cyclical low-load plyometric activities may improve running efficiency. Further investigation is required to clarify the length of intervention required, the different training requirements of youth in GMFCS Level I versus Level II and any unique training requirements of youth with bilateral impairment versus unilateral impairment. In addition, the development of a valid and reliable running classification system may assist in targeting intervention to the needs of subgroups within the CP population.

9.2 Limitations

The power calculation for the study was based on clinical outcomes which may have resulted in the biomechanical analysis being under-powered to detect changes in power generation or leg stiffness, especially in the sub-groups GMFCS Levels I and II. Furthermore,

the heterogeneity of the CP population resulted in large inter-participant variability. This increases the possibility of failing to find a significant difference between intervention and control groups, where one exists (type II error). The modelling study, reported in Chapter 4, aimed to develop a model which most accurately represented joint movement in children with CP in GMFCS Level I and II to minimise the risk of a type II error. In addition, despite analysing multiple variables, the *p*-value was not adjusted but retained at 0.05 for significance. This may have increased the chances of a type I error, or reporting a significant difference which is not a true difference, however the chance of a type II error was considered the greater risk in this body of work. Some proponents may consider the effect size of the changes when considering the need for alpha level adjustment. Although some between-group differences were identified in biomechanical variables at follow-up, these results need to be interpreted carefully, especially the sub-group analysis of GMFCS Levels I and II. The sub-group analyses need to be verified with a much bigger sample.

Natural gross motor maturation has the potential to cause a confounding effect, both over the 12 weeks between baseline and follow-up, and in the range of ages of participants from nine years to 18 years. Typically developing children can be expected to have a reasonably mature running pattern by nine years[91], but maturation time frames can be different in children with CP[17]. In addition, the effect of puberty on leg stiffness and power generation is another possible confounding factor[71]. These limitations were minimised by randomly stratifying the participants into groups according to age and gender, and by providing a parallel control group. In addition, joint power was normalised for height and weight. The systematic review, reported in Chapter 3, did not identify any evidence that the intervention should be tailored differently for children at age nine years compared to age 18 years.

The doctoral candidate was one of the interventionists, introducing a potential risk of bias. The risk of bias was minimal as the doctoral candidate was one of six physiotherapists who all received the same training prior to commencing the study and were overseen by a study supervisor who was not involved in the collection or analysis of biomechanical data. In addition, the collection of 3DGA data is an objective process with few opportunities for bias to occur. No instructions or comments about running technique were given to the participants during testing and the instructions regarding speed were standardised.

The systematic review update identified that the number of papers in the research area has doubled during the period of enrolment and preparation of the research design. However,

the update failed to identify any studies reporting biomechanical outcomes of an intervention, such that the new information would not have changed the adopted research design.

9.3 Future directions

This body of work has examined elements primarily from Body Structure and Function and Activity areas of the International Classification of Functioning, Disability and Health. The thesis has focussed on the impairment level to inform understanding of changes in activity and participation and provides a basis for future research focus and directions. There remain gaps in knowledge which may form the basis for further investigations such as those outlined in the following paragraphs.

This thesis focussed on kinetics because of the importance of power generation for running. Changes in kinetics following the running intervention implies possible changes in kinematics, but this data has not yet been reported. Future work should describe the kinematics of running in children and adolescents with CP, including the multi-segmental kinematics of the foot. In addition, it is yet to be reported how running barefoot differs from shod running in people with CP. This information would add to the understanding of how changes in power generation during running are achieved.

Chapters 7 and 8 reported changes in power generation and leg stiffness that were different in GMFCS Level I compared to Level II. Further investigation is required to determine whether interventions for running skill should be tailored for CP subgroups, such as GMFCS Levels I and II, or bilateral and unilateral CP. This information would aid clinicians in tailoring intervention to individual participants. This doctoral work did not initially plan to analyse by sub-group, but it became apparent that such an analysis would be necessary for a thorough understanding of the emerging picture. The biomechanical analysis was therefore potentially underpowered and further analysis by sub-group should be undertaken in a much larger cohort.

Chapter 8 reported improvements in leg stiffness in GMFCS Level I which implies a greater contribution of elastic energy to power generation and thus improved efficiency. Future work should investigate oxygen consumption during running before and after intervention to expand our understanding of changes in the stretch-shortening cycle in the CP population. In addition, further analysis is required to determine which impairments (e.g.

spasticity, contracture, weakness) are related to running function and which impairments change following the running intervention.

Chapters 7 and 8 reported changes in power generation and leg stiffness immediately following participation in the intervention. Further analysis is required to determine whether changes in running function are maintained in the medium- to long-term following cessation of the running training. Future investigations should attempt to determine the optimal dose for intervention in terms of days per week, number of weeks and session intensity. This is important for ensuring that intervention is cost-effective.

Further analysis of the data should aim to determine whether changes in running biomechanics are related to functional outcomes or goal attainment. For example, are those participants who showed the most increase in power generation at the ankle those who improved the most on the HiMAT or the MPST? Or were changes in leg stiffness more closely related to functional outcomes or goal attainment? Are the HiMAT and MPST sensitive to change in this population?

This body of work was undertaken in children and adolescents with CP. It may serve as a model for the investigation of running intervention in other paediatric and adult populations, such as people with Down Syndrome, Developmental Coordination Disorder, Autism Spectrum Disorder or Acquired Brain Injury (ABI). The influence of intellectual disability upon both the assessment process and the motor learning aspect of the intervention may require adaptations to be made, for example, a longer intervention period may be necessary for participants with a diagnosed intellectual disability or behavioural challenges. Children with acquired brain injury (that is, injury acquired after the age of two years) have both similarities and differences to children with CP (that is, injury acquired before the age of two years), both have decreased walking gait speed compared to TD children, but children post-traumatic brain injury have more variable step length and increased step time compared to children with CP[293]. The intervention analysed in this body of work has been effective in adults post-ABI[294], and its effectiveness in children with ABI is an avenue worth exploring in future work.

9.4 Conclusion

Running skill is a facilitator to an active lifestyle which promotes health and wellbeing, and is important for participation in play, sport and recreational activities. Children with CP do

not run as well as their typically developing peers and this body of work has shown that part of the limitation is due to the biomechanics of their running. The contribution of ankle plantarflexor power to forward propulsion in running is reduced in young people with CP and is related to GMFCS level. This deficit appears to be compensated in part by increased hip flexor power generation during swing, but maximum speed is reduced. Children with CP also have atypical leg stiffness profiles which implies that the function of the SSC is sub-optimal. Leg stiffness profiles are different for GMFCS Level I and Level II. A low-load plyometric intervention to improve the skill of running in children and adolescents with CP is effective for running-related goal attainment and can effect change in running biomechanics. This body of work has reported improvements in ankle power generation for running in young people with CP in GMFCS Level II and improved leg stiffness in GMFCS Level I.

This body of work may form a foundation for further investigation of running training in people with CP, including an investigation of how plantarflexor spasticity and contracture relate to ankle power generation or SSC function, the optimal dose of intervention, how long improvements are maintained following intervention, and whether intervention should be differently tailored for children in CP subgroups. This work could also be replicated in other neurodevelopmental diagnostic groups.

Appendices

Appendix 1 The effect of a running intervention on running ability and participation in children with cerebral palsy: a randomized controlled trial

PREFACE: The first paper published as a result of the investigations that form the body of word described in this thesis, which reported participation and field measures of the randomized controlled trial, was published with Dr. Gibson as the first author. This paper was published as "Gibson, N., Chappell, A., Blackmore, A. M., Morris, S., Williams, G., Bear, N., & Allison, G. (2018). The effect of a running intervention on running ability and participation in children with cerebral palsy: a randomized controlled trial. *Disability and rehabilitation*, *40*(25), 3041-3049." https://doi.org/10.1080/09638288.2017. 1367426. The manuscript is reproduced as it appears in print below.

Abstract

Purpose: To evaluate effects of a running intervention on running ability and participation in children with cerebral palsy (CP).

Materials and Method: Children with CP (9 to 18 years) with Gross Motor Function Classification System levels I-III, were randomly assigned to a 12-week running intervention or usual care. Primary outcomes included improvement in running ability (assessed by Goal Attainment Scaling), high- level mobility (assessed by the High-Level Mobility Assessment Tool) and participation (assessed by the Participation and Environment Measure for Children and Youth). Secondary outcomes were aerobic and anaerobic fitness and agility. Blinded assessments took place at baseline and 12 weeks. Regression analysis adjusting for baseline differences was used to determine between-group differences.

Results: Forty-two participants (mean age 12.5 years, SD 2.8 years; 15 female) completed the study. Statistically significant group differences at 12-weeks were found for improvements in running ability (86% treatment group versus 0% control group achieved or exceeded their running goals, $p<0.001$), and participation in the school environment (Participation and Environment Measure mean difference 1.18: 95%CI 1.00 to 1.39, $p=0.045$).

Conclusion: A 12-week individualized running training skills intervention results in achievement of running ability goals and participation in the school environment in children with CP.

Keywords: rehabilitation, pediatric neurological injury, goal-directed training, goal attainment, high- level mobility, motor skill

Introduction

Children with disabilities are less physically active than their typically developing peers[1]. Evidence suggests that without intervention to increase physical activity, sedentary children are more likely to become physically inactive adults, placing them at risk of poor health[2]. Encouraging the development of positive physical activity habits in children helps establish patterns that continue into adulthood[3].

Participation in physical activities depends considerably on proficiency in motor skills[4]. Running is a fundamental motor skill achieved in early childhood. It is the cornerstone for many individual and team sports and recreational activities[5]. Children with CP classified at Gross Motor Function Classification System (GMFCS) Level I and II show limitations in high level mobility and physical activity relative to their typically developing peers[2,6]. Although some children with CP have some capacity to run, they experience limitations of their running ability, particularly with speed and agility[7,8] and they may avoid or discontinue membership in sports and recreational activities due to these deficits in their running ability[9].

Despite the considerable focus on addressing walking limitations in children with CP, running training has received little attention despite its importance for play and participation[10]. Intervention specifically focused at training the components and skills of running are important for a number of reasons. Firstly, determinants of motor change and acquisition of skill in CP require therapy and training[11]. Secondly, running is often identified as a goal of intervention for school-aged children because many school activities are focused around running. Thirdly, although many interventions have been shown to improve muscle strength, anaerobic and aerobic capacity and even walking capacity in CP, few have demonstrated a translation of these gains into higher level mobility (such as running), or increases in participation[12,13].

Specificity of learning principles states that learning is optimized by practice that approximates the target skill[14]. This suggests that to improve running ability, the intervention has to include running. A targeted running skills training program for adults

with acquired brain injury (ABI) and has been shown to increase participation in meaningful recreational and leisure activities in that population[15].

As both ABI and CP are non-progressive brain injuries, a running training program of this kind may also be effective for children with CP. However, there are clear differences between children with CP and adults with ABI which may influence the application of this program to children. The adult ABI population includes a high representation of young males, which may partly account for the running intervention appeal and success. In adults, motivation to acquire the skill is largely under the control of the participant, but in children, ability to access an intervention is influenced by family capacity. It is likely that children who access a program regularly and maintain their skills over time are those whose families support them to do this. Further, in contrast to people who experience an ABI in early adulthood, children with CP have not experienced typical motor skill acquisition. Children with CP will need to learn running skill for the first time rather than re-attain a skill learnt during typical motor development. Because of these differences between adults with ABI and children with CP, and because parental influences and lifestyle factors are known to influence physical activity levels in children[3], a program incorporating a family directed motivational aspect is likely to optimize outcome.

This study aimed to determine if a running skill acquisition and training intervention could improve running ability, general high-level mobility and participation in children with CP. Secondary aims were improving running capacity that included aerobic and anaerobic fitness and agility. We hypothesized that children in the intervention group would improve in running ability, general high-level mobility and improved levels of participation; and that these would exceed the levels of these outcomes attained with standard care.

Materials and Methods

Participants

To be included in the study, participants had to be aged 9 -18 years; have CP and be classified as GMFCS Level I-III; be able to walk 10 meters unassisted; be willing to attend at least two after-school intervention sessions per week for the duration of 12 weeks; and be willing to commit to completing two additional exercise sessions (at home or in a gym) per week for the 12 weeks.

Children were excluded from the study if they had a medical condition that precluded participation in a vigorous exercise programme; had surgery in the last 6 months; were unable to complete the pre-intervention assessments due to the lack of understanding the instructions; or had cognitive or behavioural challenges that may have interfered with intervention delivered in a group setting.

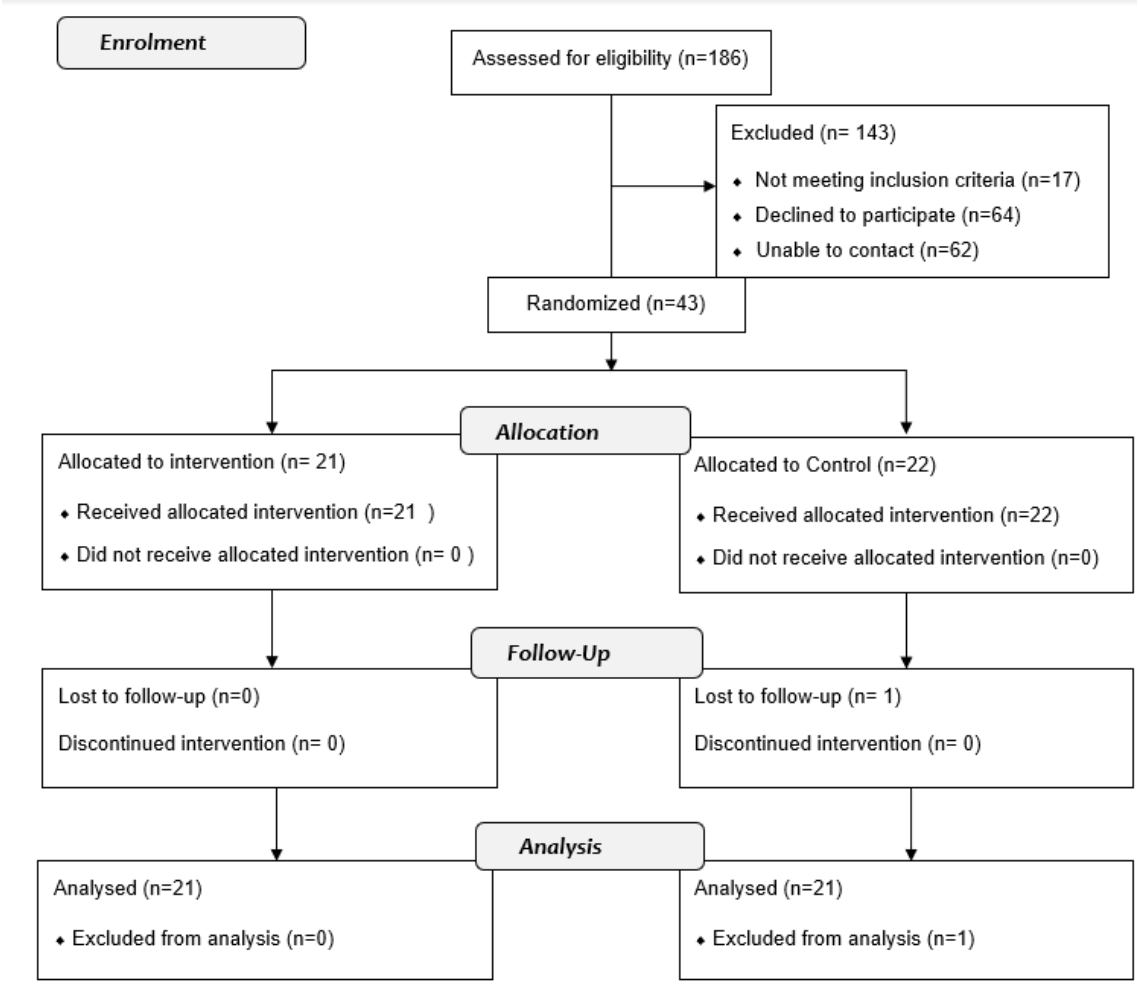
This intervention aimed to change running ability. Due to the lack of suitable research to determine effect size, power was calculated based on published data from the 10-m shuttle run test measuring running fitness[16]. With a significance level of 0.05, a *d* of 0.65, and a power of 80%, 30 participants were required for each group. Due to logistical constraints, 43 children (15 female) were recruited for this study from a local non-government therapy center.

Study design

The study was approved by the Ethics Committees of Princess Margaret Hospital, Perth, Western Australia and Curtin University, Perth, Western Australia. The trial was prospectively registered with Australian and New Zealand Clinical Trials Register, Trial Number ACTRN12614000467639. Informed consent was obtained from participants where applicable and their parent/guardian.

This study was conducted between January 2015 and May 2015 and utilized a randomized controlled trial design. On the basis of age (less than 13 years or 13 years and over) and High-Level Mobility Assessment Tool score (HiMAT, score up to 25 or over 25), participants were stratified into four blocks, and then randomly assigned to the intervention and control groups using computer-generated random numbers, by a team member who was not involved in recruitment or intervention. It was assumed that children with similar levels of high-level mobility function at a similar age would have a similar potential to respond to the intervention. The children and their parents were not told which group they were in until after all their baseline assessments were completed. The study design is illustrated in Figure 1.

Figure 1: Consort Flow Diagram



Intervention

The intervention is described in more detail in Table 1. Participants assigned to the intervention group received the running training intervention for 12 weeks at a community therapy organization. The intervention sessions were conducted after school hours and at two metropolitan locations so that participants could pick the location and day most suitable to them. The intervention was based on a running skill program for adults with neurological injury described by Williams & Schache and Schache et al., and is essentially an individualized program of exercises focused on running skill, conducted in a group setting. The intervention included a hierarchical series of exercises that targeted the three main muscle groups responsible for forward progression and achievement of a flight phase in running, i.e. the ankle plantar flexors, hip flexors and hip extensors[17,18]. The individually tailored exercises were chosen by the therapist after viewing slow motion sagittal and frontal video footage of the participant's running gait and determining the abnormalities affecting the acquisition of typical running skill. The program was underpinned by motor

control principles of which practice was a key component[18]. Participants were required to attend twice a week for one hour per session where they received individually tailored neuromuscular control exercises that were delivered in a group setting by a physiotherapist trained in the running program intervention. They were also given a home program to complete on another two days per week.

Therapists conducting the running training intervention underwent a two-day induction in the principles and methods of the training intervention developed by Williams and Schache[243]. In addition, the therapists were trained in motivational interviewing and asked to motivate participants to identify and act on increased opportunities for physical activity participation in the school and community. Participants in the treatment group received a weekly email newsletter from the study coordinator (NG) indicating the purpose and intent of the exercises the children were practicing. This reinforced the importance of mass of practice and application outside of the face-to-face group sessions.

Participants assigned to the control group received their usual therapy provided by their community therapist. The usual care was monitored with a questionnaire and not altered for the duration of the study.

Table 1: Running in cerebral palsy - Description of Intervention

Item	Details
1. Exercise equipment	Warm up only: exercise bike, elliptical Intervention: Total Gym® leg sled or similar, trampette, stairs, resistance bands, single step, cones, agility ladder
2. Qualifications and training	Two senior and three base-grade physiotherapists (PTs) with experience in working with school-aged children with cerebral palsy (CP) delivered the program. All therapists involved in delivering the program had undertaken 15 hours training in rehabilitation of running for people with neurological impairment. All PTs also undertook eight hours training in motivational interviewing delivered by a clinical psychologist.
3. Individual/Group	Individualised programs were established and progressed at individual rates. The individualised program was delivered and performed in a group setting twice per week and at home programme provided to be performed twice per week.
4. Supervision	Participants were supervised in a ratio of 1PT:3participants. The participants were taught the exercises individually and once performing the exercise correctly, allowed to practice independently.

5. Adherence to exercise	Progress notes were completed at the end of each session, including the level of difficulty of each exercise and number of repetitions, or time spent doing the exercise. Attendance was recorded for each participant and reported as a number of sessions out of 24 possible sessions. Home program exercises were prescribed weekly and home exercise diary sheets were collected at the end of each week.
6. Motivation strategies	Participants were encouraged with verbal feedback about their technique, both what was done well and what changes needed to be made. Participants were given a time or number of repetitions to aim for. Exercises were incorporated into games where possible. Participants were also encouraged to 'buddy up' with another group participant to encourage each other. Each participant had a home program, with diary sheets issued and collected weekly to encourage adherence. Participants received a weekly newsletter that highlighted the rationale and purpose of different aspects of the exercises, for example, what an exercise was trying to achieve related to the skill of running, the importance of frequency of practice, the importance of practicing the exercises properly, the importance of getting the correct 'dose' of the exercise prescribed etc. Participants who did not attend a session without informing PTs of the reason were called by one of the PTs the next day to encourage attendance. The PTs used motivational interviewing techniques to encourage participants to explore options for adhering to home prescribed exercises and for exploring and promoting physical activity in the community.
7a. Decision rule(s) for determining progression	Once the participant was consistently able to perform the exercise with good technique they were progressed to the next level. Speed and quality of movement were prioritised over load as the focus was on ballistic movement necessary for running.
7b. How program was progressed	The program incorporates a series of hierarchically challenging 'pre-runner' and 'runner' activities. [See Williams & Schache 2010 and Schache et al., 2014 for more detail on the activities][17,18]. Participants are prescribed relevant activities to address the running gait impairments demonstrated for that individual. The individually tailored exercises are derived by the therapist viewing slow motion observational sagittal and frontal video footage of the participant's running gait and determining the abnormalities affecting the acquisition of typical running skill. The exercises/activities target the three main muscle groups responsible for forward progression when walking and running, i.e. the ankle plantar flexors, hip flexors and hip extensors. Activities were not progressed from 'pre-runner' to the 'runner' activities until the individual could achieve a flight phase during running gait. Exercises were

	progressed once good form/technique was demonstrated on the starting activity. For pre-runners, progression of propulsion and running exercises occurred on the Total Gym® leg sled until the highest level was reached. The exercises were then performed on the trampette until the participant was able to perform the exercises overground. For simulation of leg turnover and appropriate foot contact alignment an activity termed the "claw" exercise was utilised [See Williams & Schache 2010 and Schache et al., 2014 for more detail][17,18]. For participants whose motor control did not enable good technique, activities such as the "claw" were either broken down into components, or facilitated with therapist handling until the participant was able to perform a cycle with good technique. This was progressed by decreasing therapist facilitation and eventually adding resistance (for e.g., with resistance bands). Once the participant progressed from a pre-runner to a runner and could run with good technique overground, slopes were added and the distance or speed increased depending on the individual goal. From slopes, participants progressed to agility exercises. These began with simple cutting/side-stepping exercises and progressed in complexity. Once a reasonable level of agility was attained, sport-specific skills relevant to the individual participant's interest were introduced.
8. Exercises	Please refer to Williams & Schache 2010 and Schache et al., 2014 for the types of exercises utilised to address different running impairments[17,18]. Each session followed the following structure: 1. Warm up (usually on a stationary bike, elliptical or cross trainer): 5 minutes 2. Individualised pre-runner or runner exercises/activities: 50 minutes 3. Passive and active stretches: 5 minutes
9. Home programme	All participants received a home program to be performed twice weekly, which contained individually tailored exercises that had been learnt with the PTs and which they could perform independently with good technique. Participants were asked to complete the home program on two additional days to the two training days that they attended.
10. Non-exercise components	N/A
11. Type and number of adverse events	One participant sustained a sprained ankle, which required first aid and resolved within one week with full return to activity.
12. Setting	Two locations were utilised: - A public oval with sports clubroom - A community service provider with an adjacent public reserve
13. Exercise	Participants were asked to attend two one hour sessions

intervention	per week with the home program performed another two times per week, for a total of 12 weeks.
14a. Generic/Tailored	Each participant received an individually tailored program based on a core group of hierarchical exercises.
14b. How the exercises are tailored	The exercises were tailored for each participant according to the identified impairments impacting their ability to run and by level of difficulty. The PTs progressed the exercises according to the participant's response. Adjunct exercises were added by the PTs if necessary, for example hip abductor strengthening exercises were added if the participant could not stabilise the pelvis while performing the exercises.
15. Decision rule for starting level	Participants were started at the most challenging level they could perform with good technique.
16a. Adherence/Fidelity	All PTs undertook 15 hours training covering the theoretical and practical underpinnings of the exercise program. PTs met for 10 minutes following each session to discuss issues experienced by individuals in the group and find solutions. The principle investigator/senior physiotherapist visited each site where the group program was being delivered to ensure the integrity of the program was being maintained, and to troubleshoot any intervention queries from the administering therapists. This procedure reinforced similar administration of the intervention between PTs at each site and adherence to the program. Additionally, all PTs delivering the program met on two further occasions and presented a total of 6 case presentations of intervention delivery and progression of the activities. These case presentations were chaired by the principle investigator and aimed to reinforce and ensure the intervention strategies and principles were being adhered to.
16b. Intervention delivered as planned?	The intervention was delivered as planned. Median number of sessions attended out of a possible 24 training sessions in the 12-week intervention was 16 (66.7%), with the minimum 4 (16.7%) and maximum 21 (87.5%).

Outcome Assessments

All participants attended two study assessments, at the beginning and end of the 12-week period. These assessments were performed at a university laboratory. Two research assistants who were blinded to group allocation conducted the assessments.

Running is the ability to generate a flight phase consistently between alternating foot strikes[131]. Running skill refers to proficiency in the motor skill of running[19]. **Achievement of running ability**, or improvement towards acquisition of running skill was evaluated using Goal Attainment Scaling (GAS**)** incorporating achievement of components of running skill

within the formulation of the goal. The GAS is validated for use in CP intervention trials and provides a criterion-referenced measure of progress over time towards previously defined individualised goals[20,21]. Goals were set in close collaboration with the child, the child's parents, and the community physiotherapist prior to randomisation. Goals were scored on a 5-point scale with -2 set as the baseline score.

An example of a GAS goal related to running skill derived from a participant's goal articulated as "be able to run faster" would be formulated as: Goal 1: to be able to complete a 50-meter sprint in *x seconds* less time than baseline time trial. The therapist would then perform observational video analysis using slow motion capture of running to identify the key biomechanical abnormality of the running technique to both develop the therapist-derived goal and the focus of the intervention to achieve the goal. For the example above, the therapist might determine that a running impairment limiting the ability to run faster is inability to generate a flight phase off the affected leg, so the concomitant goal, 2: "to be able to bound *x cm* off the more affected leg". Where *x* in Goal 1 and Goal 2 is determined as the expected change from the baseline score and then a 5-point scale is determined using the baseline score, the expected goal attainment and levels greater than the expected goal attainment. The GAS score was converted to a weighted T-Score with 50 (SD=10), indicating goal attainment[22].

Improvements in high- level mobility were measured using the High-Level Mobility Assessment Tool (HiMAT)[23]. The HiMAT was developed to quantify high-level mobility outcomes following ABI and has been validated in children with ABI and CP[24]. It consists of 13 items on a hierarchical mobility continuum and includes walking forwards, walking backwards, bounding, running, hopping and stair climbing. Measures obtained on each item are scored and summed for a total score out of 54, with higher scores indicating better mobility performance. In children with ABI a 2-point improvement or 4-point deterioration indicates a clinically significant difference with 95% confidence[24].

Change in participation was assessed using the Participation and Environment Measure for Children and Youth (PEM-CY)[25]. The PEM-CY is a parent-report instrument that examines participation and environment factors that affect the participation of children across three settings: home, school, and community. Parents were asked to rate their child's involvement in 25 activities across the three settings. Examples of activities are "computer and video games" at home, "classroom activities" at school, and "neighborhood outings" in

the community. For each activity, the parent was asked how often their child participated in 1 or more activities of this type and the question was rated using an 8-point scale, from never to daily. In addition, they were asked to rate their child's involvement in each of the activities using a 5-point scale, from minimally involved to very involved, and whether they wanted their child's participation to change in this type of activity. The clinically meaningful difference for PEM-CY has yet to be determined but it has good intra and inter-rater reliability[26].

Running capacity includes running-related aerobic fitness, anaerobic fitness which is related to running velocity and power generation and agility, which is the ability to change direction or velocity rapidly in response to a stimulus[27].

Aerobic fitness was evaluated using the 10-metre shuttle run test (SRT) validated for children with CP classified at GMFCS Level I or Level II[16]. The test has been shown to be sensitive to change in children with CP with a change of more than 0.84 minute (one level) for the SRT-GMFCS I and of more than 0.50 minute (half level) for SRT-GMFCS II attributed to clinically meaningful improvement[28].

Anaerobic Fitness was measured using the Muscle Power Sprint Test (MPST) administered as per the guidelines by Verschuren et al.[29]. Participants were instructed to complete six 15-metre runs at a maximum speed with 10 seconds rest between each run[29]. Power output for each 15-metre sprint was derived using the sprint time (seconds) and the participant's body mass (kilogram) such that power (Watts) for each run was determined by

$$\text{Power (Watts)} = (\text{mass} * \text{distance}^{2})/(\text{time}^{3})$$

The minimal clinically important difference for the MPST in children with CP is a change greater than 18Watts (Mean Power)[30].

Agility was measured using the 10 × 5-Meter Sprint Test. This is a timed continuous sprint test whereby the participant has to make nine fast turns after finishing every five meters with no rest between each turn. The time taken to execute the 10 x 5-meter sprints yields a valid measure of agility in children with CP[295]. The minimal clinically important difference for the 10 × 5-Meter Sprint Test is a decrease in time of more than 3.2 seconds[30].

Statistical Analysis

Group characteristics and outcome measures are described using mean and standard deviations for continuous variables and frequencies and proportions for categorical variables. Outcome measures were assessed for normality and log transformations were applied for skewed distributions. When log transformations were not successful non-parametric tests were applied to the data and the data described using medians and ranges. Within-group differences were completed using paired t tests for the normally distributed and log transformed data. The non-parametric equivalent test used was the Wilcoxon signed rank test.

Between-group differences were analysed using regression method where group allocation and the baseline score included the regression equation. The group coefficient represents the estimated difference between the two treatment groups, adjusted for baseline score. For skewed data that was able to be adequately transformed the regression coefficients were back transformed (exponential), with the interpretation in terms of percent change. This occurs as back transformation results in ratios of the geometric mean. Any back transformed coefficient with a 95% confidence interval crossing the value 1, indicated a non-significant result.

The GAS was examined for change in score from baseline and examined between groups using Fishers exact test. Clinical difference changes were examined for the HiMAT, SRT, MPST and the 10x5m sprint. Change in scores was examined for each individual and the proportion exhibiting a clinically meaningful change was compared between the two groups.

Alpha was set at 0.05. All analysis was completed in STATA version 13.1 (StataCorp, Texas).

Results

Demographic information and baseline values for participants are presented in Table 2. The two groups were well matched in terms of gender, age, height, and weight. The treatment group was the only group to include a child classified as GMFCS III. Baseline outcome measures were similar between the two groups except for MPST peak and mean power; however, neither of these differences were statistically significant ($p>0.05$).

Table 2: Participant Characteristics in Treatment and Control Groups

	Control n =21 n(%)	Treatment n=21 n (%)
Gender Male Female	13 (61.9) 8 (38.1)	14 (66.7) 7 (33.3)
GMFCS I II III		
	12 (60.0) 9 (40.0) 0 (0.0)	12 (57.1) 8 (38.1) 1 (4.8)
Topographical involvement		
Unilateral Bilateral	13 8	12 9
	Mean (SD)	Mean (SD)
Age (years)	12.5 (2.8)	12.4 (2.7)
HiMAT Score	25.7(10.6)	25.8(12.2)
Height (cm)	152.6 (15.6)	152.5 (14.0)
Weight (kg)	51.1 (19.5)	47.8 (15.6)

cm=centimetres; GMFCS=gross motor classification system; HiMAT= high- level mobility assessment; kg=kilograms; n=number; SD=standard deviation

Median number of sessions attended out of a possible 24 training sessions in the 12-week intervention was 16 (66.7%), with the minimum 4 (16.7%) and maximum 21 (87.5%).

Table 3 reports the mean differences between time 1 (T1=post-test) and time 0 (T0=baseline) for the outcome measures for the treatment and control groups. Group differences are reported using beta coefficient and 95% confidence intervals. GAS T-Score (22.4: 95% CI 16.2 to 28.5), and the average school frequency of participation (1.18: 95%CI 1.00 to 1.39) were the only outcomes to demonstrate a statistically significant difference. The change in GAS score for the treatment group was more than two standard deviations greater than the change for the control group.

Table 3: Between-group differences and 95% confidence intervals using regression analysis controlling for baseline

Outcome	Difference T1-T0 Mean (SD)		Group differences Coefficient (95% CI) [2]	P value
	Control	Treatment		
GAS T-Score	2.38(4.36)	24.76(13.27)	22.4(16.2, 28.5)	<0.001*
HiMAT	1.7 (5.4)	2.5 (5.7)	0.8 (-2.7, 4.3)	0.651
SRT level	0.6 (1.4)	1.6 (2.3)	1.0 (-0.2, 2.2)	0.110
SRT (number)	7.0 (12.9)	16.1 (22.4)	8.8 (-2.7, 20.3)	0.131
SRT (min)	0.7 (1.3)	1.6 (2.5)	0.9 (-0.3, 2.2)	0.142
MPST Peak Power (W)	21.4 (57.4)	14.1 (53.7)	-6.0 (-40.3, 28.2)	0.723
MPST Mean Power (W)	25.6 (58.9)	16.5 (50.5)	-6.8 (-40.6, 27.0)	0.687
10x5 Sprint sec	0.2 (7.9)	-0.5 (4.7)	-1.3 (-5.4, 2.8)	0.535
PEM-CY HOME Ave Frequency (0-7)	0.0 (0.8)	-0.3 (0.8)	-0.2 (-0.9, 0.4)	0.440
PEM-CY HOME % of activities (0-100)	-1.2 (9.6)	-4.3 (13.4)	-3.3 (-11.5, 4.8)	0.409
PEM-CY HOME Ave involvement (1-5)	- 0.2 (0.7)	-0.1 (0.8)	0.2(-0.3, 0.6)	0.421
PEM-CY HOME % desired change (0-100) [1]	0.6 (14.8)	-9.9 (29.7)	0.8 (0.5, 1.3)	0.403
PEM-CY SCHOOL Ave Frequency (0-7) [1]	0.0 (1.4)	0.7 (1.2)	1.2(1.0, 1.4)	0.045*
PEM-CY SCHOOL % of activities	11.2 (30.8)	5.3 (14.1)	0.1 (-1.0, 9.8)	0.984
PEM-CY SCHOOL Ave involvement (1-5)	-0.5 (1.1)	0.1 (0.9)	0.4 (-0.2, 1.1)	0.186
PEM-CY SCHOOL % desired change [1]	-12.3 (37.4)	-9.6 (41.3)	1.1(0.6, 2.2)	0.662
PEM-CY COMMUNITY Ave Frequency (0-7)	-0.2 (0.9)	-0.2 (1.3)	0.1(-0.6, 0.8)	0.872
PEM-CY COMMUNITY % of activities	-10 (17.1)	1.6 (21.2)	7.8 (-4.3, 19.8)	0.198
PEM-CY COMMUNITY Ave involvement (1-5)	-1.4 (1.1)	-0.3 (1.5)	0.8 (-0.0, 1.7)	0.060
PEM-CY COMMUNITY % desired change [1]	-7.3 (40.9)	3.6 (30.8)	1.6 (0.8, 3.2)	0.201

[1] requiring log transformation for analysis - back transformed coefficients; [2] Adjusted for baseline; * significant

Ave=average; CI=confidence interval; GMFCS = Gross Motor Functional Classification System; HiMAT=High-Level Mobility Assessment Tool; min=minute; MPST=Muscle Power Sprint Test; n=number; PEM-CY= Participation and Environment Measure Child and Youth; SD=standard deviation; sec=seconds; SRT= shuttle run test; T0= time 0 (baseline); T1=time 1 (post-test); W= watts

Table 4 shows the percentage of participants who showed clinically meaningful changes in

outcomes between the groups. For the treatment group all but one participant made some improvement in their GAS. There were 86% of participants achieving their goal in the treatment group versus none in the control group (difference in proportions of 86%: 95% CI 71% to 100%, p<0.001).

Table 4: Number (and percent) of participants in each group achieving clinically significant change

Outcome	Clinically significant change	Control n(%)	Treatment n(%)
*GAS	+2	0 (0)	7 (33.3)
	+1	0 (0)	3 (14.3)
	0	0 (0)	8 (38.1)
	-1	4 (19)	2 (9.5)
	-2	17 (81)	1 (4.8)
HiMAT	+2	7 (33.3)	12 (57.1)
	-4	3 (15.0)	3 (14.3)
Functional Strength	>9	7 (33.3)	6 (28.6)
MPST mean power	>18W	9 (45.0)	10 (47.6)
10x5 sprint	<-3.2 sec	4 (19)	4 (20)
Modified SRT	GMFCS 1>0.85 min	4 (30.8)	7 (53.8)
	GMFCS II/III>0.5 min	5 (62.5)	6 (75.0)
	overall	9 (42.9)	13 (61.9)

GAS clinically and statistically significant difference between groups at time point 1 (12 weeks post), *p<0.001 using Fischers exact test

GAS= Goal Attainment Score; GMFCS – Gross Motor Functional Classification System; HiMAT=high-level mobility assessment; min=minute; MPST=muscle power sprint test; n=number; sec=seconds; SRT= shuttle run test; W = Watts

Discussion

The *a priori* aim of this study was to determine if a goal-directed, 12-week running skills intervention could improve running ability, general high-level mobility and participation in children with CP. The rationale for this study was underpinned by improving running skill to mediate positive responses in participation. Significantly increased achievement of running-related goals (as measured by the GAS) and increased frequency of participation at school were demonstrated, despite there being no significant differences in other running and high- level mobility measures at the ICF body structure and function level[4].

The majority of goals set by the participants were related to being able to participate fully in running-related tasks. Specific attention was paid to determining goals as part of the intervention as goal-directed interventions are evidence-based in CP[31]. Participant-set goals were augmented by therapist-derived running skill goals required in order to attain the

participant goal. The high retention rate of study participants and the high adherence of attendance in the intervention group showed that participants regarded this as an important and valuable intervention.

The reported higher frequency of participation in school activities at the completion of the intervention suggests an increased confidence to participate, at least initially in a safer environment such as school. We are unable to report a clinically important change based on the PEM-CY as this value is unknown. However, this result suggests that frequency of participation in the school environment is modifiable in this population with an additive contribution from improving running ability. The improvement to participants over time—as a group or individually—may yield clinically meaningful improvements but what this value is for a clinically meaningful change in the PEM-CY requires further investigation.

There were no differences in participation reported in the community settings. It may require more time for participants to develop the confidence to translate participation into community areas. In addition, our positive effects on participation were seen within 12 weeks, whereas for other studies this was not achieved at all, or not achieved until 12 months[31]. Whilst this improvement in participation is promising, further investigation is required to determine whether changes are maintained or improved, as previous studies have shown that improved participation immediately following an intervention period are not maintained 6-12 months later[13,31]. Considered together, the improvement in GAS and school participation suggest that it is the improvements in running ability that had a positive effect on increasing participation.

Despite the importance of running for participation for children and adolescents with CP, it has received relatively little attention. There have been a few small studies that have mentioned running, but they tend to mention it as part of a suite of outcomes (particularly as a measure of fitness), rather than training the motor skill of running, or running gait intervention itself[13,32,33]. Whilst improvements in fitness and agility are reported, translation of improvements into participation have either not been assessed[32,33] or not been demonstrated[13].

Our results demonstrated that the individualized targeted running intervention adapted from William and Schache adult program[34] was effective at achieving running-specific goals of each of the participants in a cost-effective group environment. The high rate of goal

attainment adds evidence both to the effectiveness of the intervention program in children with CP, and to the validity of the conceptual framework presented by Williams and Schache[241] and warrants further exploration.

It is interesting to note that both the significant outcomes in this study were participation-related, with no detected differences observed in outcome measures at the ICF body structure and function level in this study. Whilst a significant difference in GAS goal achievement was demonstrated, there was no significant difference in high level mobility as measured by the HiMAT. This demonstrates the high variability of outcome of respondents in this domain and suggests that some, but not all children will make a clinically important improvement in motor skill in a program such as this one. Differences in attainment of skill may be underpinned by differences in motor learning of individuals. Some individuals may require more practice and therefore a longer time in the program to acquire new motor skills[14]. These factors should be considered in evaluating duration of a program such as this one for individual skill attainment.

Failure of our study to detect between-group differences in aerobic and anaerobic fitness and agility could be attributed to our study being under-powered for detecting change in these outcomes. This is not surprising given the intervention activities in our study were primarily addressing the acquisition of running skill (i.e., the activities were underpinned by a skills development framework) rather than fitness or agility.

For training running fitness and agility, the individual must have sufficient motor ability to undertake an appropriate level of training to affect these domains. It may be that the 12-week intervention time was not sufficient to effect a change in both the acquisition of running ability and also effectively address the domain of fitness and agility. Therapists could consider motor skills training before undertaking running fitness and agility training. It is important to note that running goals were achieved in the absence of significant improvements in these other measures of physical capacity, underpinning the importance of tailoring goal setting to the individual, even when participating in a group environment.

Limitations

The decision to include children with GMFCS III is a limitation of this study. The original intervention was designed for adults with an acquired neurological injury with inclusion of any individual that could walk 10 meters unassisted[34]. When transferring this criterion to children with CP in this study we opted to include children with GMFCS III who could walk 10 meters unassisted. We were only able to recruit one participant that met this additional criterion and articulated a higher-level mobility goal. However, this was a pragmatic clinically based study and many children with GMFCS III want to improve their mobility. We used GAS to individualise the outcome and the intervention and the three primary muscle groups for forward propulsion in walking are the same for running, so it seems reasonable to include GMFCS III if they can walk independently. However, attainment of running in children of GMFCS III is unlikely within a 12-week intervention time frame.

Whilst the positive improvements in the goal attainment of running and school participation frequency were seen by 12 weeks, a limitation of this study is there was no follow-up to see if participants maintained their improvements in running ability and participation levels. This was a proof of concept study with the primary aim being to determine whether the intervention would effect a change in running skill ability. Further work is required to determine whether these gains are maintained.

Conclusion

In considering the results of this study within the World Health Organization International Classification of Functioning[4] our findings show that access to a program specifically targeted at training running, delivered in a group context at school age is important in enhancing activities (running) and participation at school. Rehabilitation strategies to improve high- level mobility capacity should therefore be given a greater priority particularly when the benefits of participation in school-related physical activities are essential for promoting ongoing skill development and social integration.

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Appendix 2 Statement of author contribution

I, Annie Chappell, was responsible for the following:

- Conceptualisation and design of the biomechanical research;
- Biomechanical data collection, processing and interpretation of results;
- Data analysis under the advisement of a statistician;
- Contribution to the delivery of the intervention;
- Preparation and editing of the manuscript; and
- Correspondence with the journal.

to the listed publications:

Chappell, A., Allison, G. T., Williams, G., Gibson, N., & Morris, S. (2020). The effect of a running training intervention on ankle power generation in children and adolescents with cerebral palsy: A randomized controlled trial. *Clinical Biomechanics*, 76, https://doi.org/10.1016/j.clinbiomech.2020.105024.

Chappell, A., Gibson, N., Williams, G., Allison, G. T., & Morris, S. (2019). Propulsion strategy in running in children and adolescents with cerebral palsy. *Gait & posture*, *70*, 305-310.

Chappell, A., Gibson, N., Morris, S., Williams, G., & Allison, G. T. (2019). Running in people with cerebral palsy: A systematic review. *Physiotherapy theory and practice*, *35*(1), 15-30.

Chappell, A., Liew, B., Murphy, A. T., Gibson, N., Allison, G. T., Williams, G., & Morris, S. L. (2019). The effect of joint translation constraint on within-participant variability of kinematics and kinetics during running in cerebral palsy. *Clinical Biomechanics*, *63*, 54-62.

and to these manuscripts prepared for submission:

Chappell, A., Allison, G.T., Gibson, N., Williams, G. and Morris, S. A comparison of leg stiffness in running between typically developing children and children with cerebral palsy. Submitted for publication.

Chappell, A., Allison, G.T., Gibson, N., Williams, G. and Morris, S. The effect of running intervention on leg stiffness in youth with cerebral palsy: a randomised controlled trial. Submitted for publication.

Further, I, Annie Chappell, contributed the following to the publication Gibson, N., **Chappell, A.**, Blackmore, A. M., Morris, S., Williams, G., Bear, N., & Allison, G. (2018). The effect of a running intervention on running ability and participation in children with cerebral palsy: a randomized controlled trial. *Disability and rehabilitation*, *40*(25), 3041-3049:

- Delivery of the intervention; and
- Review and editing of the manuscript.

Annie Chappell

I, as a co-author and thesis supervisor, endorse that this level of contribution by the candidate indicated above is appropriate:

Dr Susan Morris	
Professor Garry T Allison	
Dr Noula Gibson	
Dr Gavin Williams	

Appendix 3 Recruitment letter

The Centre for Cerebral Palsy
PO Box 61, Mount Lawley,
[redacted]

[Insert Date]

Dear Parent,

You have been given this letter from your physiotherapist as they have identified your child as potentially suitable for involvement in a running and lifestyle group for children with cerebral palsy.

This study is looking at teaching running and high level mobility skills to children with cerebral palsy. Your therapist has indicated that your child may be suited to this study as they have expressed goals around running skills, and/or of increased physical activity and participation.

This is a joint study with Princess Margaret Hospital. We are investigating a three-month programme that teaches young people with CP to run and make running and activity a part of their weekly routine. The study will tell us what improvements this running programme makes to running ability, activity levels and social participation of young people with CP.

The running training is based on a successful training regime in adults who have acquired a brain injury. We have modified the programme for children and young people and wish to see if it is beneficial in children with CP. The running training will occur at two sites (Coolbinia and Currambine) 6 days per week either before or after school, and you can choose to attend whichever sites/days suit your family. This study will train your child to run with a goal in mind. At the end of the running training study we expect that your child will have achieved a substantial running orientated goal such as running in a school event, lap-a-thon or community fun run. A more detailed information sheet of this research has been included with this letter. If you would like to be involved, or have further questions, please contact:

Annie Chappell, Senior Physiotherapist, The Centre for Cerebral Palsy
[redacted]
or
Noula Gibson, Research Coordinator Physiotherapist, Princess Margaret Hospital
[redacted]

Following this letter, you may receive a phone call to formally invite you to participate in this study. Should you wish not to be contacted, please contact Annie Chappell on the above details before [Insert date 2 weeks from date of letter].

Thank you very much for your time and we look forward to receiving your response.

Kind regards,

Annie Chappell

Senior Physiotherapist
School Age Intervention Services
The Centre for Cerebral Palsy

Noula Gibson PhD
Research Coordinator Physiotherapist
Department of Physiotherapy, Princess Margaret Hospital

Appendix 4 Information sheet for parents

INFORMATION SHEET FOR PARENTS

A running training and lifestyle programme for children with cerebral palsy

Why are we doing the study?
Running is the cornerstone for many sports and games. School fun runs, lap-a-thons, daily fitness, and general sport all require running. Yet running can be a big challenge for children and young people with cerebral palsy (CP), and many set a goal for themselves to learn to run.

Running is a skill that can be taught. But it takes training and practice. This is a three-month programme that teaches children and young people to run. It shows them how to make running and keeping fit a part of their ordinary weekly routine.

We are doing this study to help young people with CP participate with their schoolmates in running activities and learn to keep themselves active and fit. We want to find out exactly how this programme helps these children so that we can offer the best possible running programme to children with CP.

Who is carrying out the study?
This study is a joint initiative of The Centre for Cerebral Palsy (TCCP), Princess Margaret Hospital for Children (PMH) and Curtin University of Technology. The researchers are Dr Noula Gibson and Annie Chappell. Annie is using the information from this study as part of her PhD. Both are senior physiotherapists who have worked for many years with children and young people with CP.

What will the study tell us?
The study will tell us what improvements this running programme makes in the lives of children and young people with CP.

Does my child have to take part?
No, your child does not have to take part in this study. Whether you decide to take part or not, it will not affect the services you receive from PMH and TCCP.

What will you be asked to do if you decide to take part in this study?
You will be asked to support your child in attending at least two after-school running sessions per week for 12 weeks. You will also be asked to support your child in doing at least two other home-based or gym-based sessions over this period. Your child's programme will run in either Term 3 or Term 4 of this school year.

What does my child need to do to be in the study?
If you agree for your child to participate your child will be asked to select a sealed envelope which will allocate your child by chance to one of two groups. The difference between the two groups is that if your child is allocated to group 1, they will not start the running group for three months, but we will still test their fitness, strength and agility in that three months. Your child will form part of the group that we compare the results of the running to. If at the end of the three months your child wants to try the running group for three months they can. If your child is allocated to group 2 they will start the running group

straight away.

Your child will be expected to attend at least two after-or before-school sessions per week for 12 weeks. At these sessions, your child will do strengthening exercises, pre-running and running drills, and agility exercises. Your child is welcome to bring along a buddy to these sessions

Your child will also be expected to do a gym or home exercises programme 2 or 3 days per week.

For both groups there will also be assessments at the start of the study, 3 months later, 6 months later, and 9 months later. Each time there will be two assessment sessions when your child will be given tests of fitness, strength and agility as well as an assessment of their participation. Each assessment session will take about 90 minutes. One or both of the assessment sessions will take place at Curtin University of Technology, where your child's running will be analysed in a motion analysis lab. This involves placing markers on the legs and hips and asking the child to run over a force plate embedded in the floor. Your child will also be asked to wear an activity monitor for three days. This is a small wrist watch sized device that counts how active your child is in the day. It can calculate the amount of energy they burn through the day.

Is there likely to be a benefit to my child?
Yes. It is very likely that your child will become fitter during the programme and that they will be able to participate more in school and recreational activities, particularly sports.

Is there likely to be a benefit to other people in the future?
Yes. The information we gain from this study will be used to make the programme as good as possible for children and young people in the future who want to learn to run.

What are the possible risks and/or side effects?
There may be some trips, falls and minor injuries. Experienced physiotherapists will supervise the programme to ensure that it is safe and they will also tailor it individually for each child.

What are the possible discomforts and/or inconveniences?
The programme requires several hours per week for 12 weeks. It also requires the support of the whole family to help the child develop weekly habits of being active and fit.

Where is your information kept?
Your child's assessments are kept in their personal medical record at The Centre for Cerebral Palsy. We will also keep results of your assessments using your initials only and these will be kept in a locked filing cabinet at The Centre for Cerebral Palsy, which is reserved for research study results. A copy of the assessments with your initials only will also be stored in a locked storeroom and in the computer in the Physiotherapy Department, at Princess Margaret Hospital for fifteen years. This is because the Chief Investigator, Noula Gibson is part of Physiotherapy Department, at Princess Margaret Hospital and is supporting this research. Access to all this information is protected by a password known to the researcher, Noula Gibson. After fifteen years this information at PMH will be destroyed by erasing electronic information and shredding hard copies of study documents. Your child's clinical record at The Centre for Cerebral Palsy will remain for seven years.

What about my privacy?
Your child's results will be confidential. Your child's name will not be kept with the research

results. We will use an identification number instead of your child's name. When we publish this study, we will not use any children's names.

Who has approved the study?
The Human Research Ethics Committee at PMH has approved this study.

Who to contact for more information about this study:
If you would like any more information about this study, please do not hesitate to contact one contact one of the research team. They are very happy to answer your questions.

Name	**Title**	**Contact Number**
Noula Gibson	Chief Investigator	[redacted]
Annie Chappell	Senior Physiotherapist	

Who to contact if you have any concerns about the organisation or running of the study?

If you have any concerns or complaints regarding this study, you can contact the Director of Medical Services at PMH (Telephone No: (08) 9340 8[redacted]). Your concerns will be drawn to the attention of the Ethics Committee who is monitoring the study.

What to do next if you would like your child to take part in this research:

If you would like to take part in this research study, please read and sign the consent form provided.

THANK YOU FOR YOUR TIME

Appendix 5 Older child information sheet

INFORMATION SHEET FOR YOUNG PEOPLE (AGED 12 AND OVER)

A running group for children with cerebral palsy

Why are we doing the study?
For many sports and games, you need to be able to run.

Running is not easy for many kids with CP. That's why they set it as a goal for themselves.

You can learn to run. People can teach it to you. But you need plenty of practice. This running group lasts for 3 months. If you do it, you will learn how to run and keep fit.

We are doing this study to help young people with CP join in with their schoolmates in sports and games and learn to keep themselves active and fit. We want to offer the best possible running group to young people with CP.

Who is carrying out the study?
The researchers are Dr Noula Gibson and Annie Chappell. They work at The Centre for Cerebral Palsy (TCCP) and Princess Margaret Hospital for Children (PMH). Both are physiotherapists who have worked for years with many children and young people with CP.

Do I have to take part?
No, you do not have to take part in this study. Whether you decide to take part or not, the other services you receive from PMH and TCCP will go on just the same.

What will I have to do if I take part in this study?
If you agree to participate you will be allocated by chance to one of the two study groups. The difference between the two groups is that if you are in group 1, you will not start the running group for three months but we will still test your fitness, strength and agility in that three months. If at the end of the three months you want to try the running group for three months you can. If you are in group 2 you will start the running group straight away.

In the running group you will have to attend at least two after- or before-school sessions per week for 12 weeks. At these sessions, you will do exercises and practise your running. You are welcome to bring along a buddy to these sessions

You will also have to do a gym or home exercises programme 2 or 3 days per week.

For both groups we will test your fitness three times: one at the beginning of the study, we will test you again in 12 weeks time. You will also be asked to wear an activity monitor for three days. This is a small wrist watch sized device that counts how active you are during the day.

If you are in group 2 that starts the running group first we will also assess your fitness again at 6 months from when you started in this study to see if you have been able to keep your fitness up.

Will the running group help me?
Yes. If you do the exercises for 3 months, then you will become fitter. We hope this will

enable you to join more in school sports and games.

If I join the study, will it help others?
Yes. We will use the information we gain from this study to make the programme as good as possible for children and young people in the future who want to learn to run.

What are the risks?
You might have some trips and falls. The physiotherapists will supervise you and make sure the programme is safe for you to do.

Where is my information kept?
Your assessments are kept in your personal medical record at The Centre for Cerebral Palsy. We will also keep results of your assessments using your initials only and these will be kept in a locked filing cabinet at The Centre for Cerebral Palsy. Whenever we do a study, we keep our results in this locked cabinet. A copy of the assessments with your initials only will also be stored in a locked storeroom and in the computer in the Physiotherapy Department, at Princess Margaret Hospital for fifteen years. This is because the Chief Investigator, Noula Gibson is part of Physiotherapy Department, at Princess Margaret Hospital and is supporting this research. Access to all this information is protected by a password known to the researcher, Noula Gibson. After fifteen years this information will be destroyed by erasing electronic information and shredding hard copies of study documents. Your clinical record at The Centre for Cerebral Palsy will remain for seven years.

Will you tell other people my results?
Yes, we will share the results of this study with other people. But we will not tell them the names of any of the children in the study.

What if I change my mind?
You can withdraw at any time and your treatment from Princes Margaret Hospital or The Centre for Cerebral Palsy will not be affected in any way. If you withdraw you will be offered the same care as you usually receive through The Centre for Cerebral Palsy.

Who has approved the study?
The Ethics Committee at PMH has approved this study.

Who to contact for more information about this study:
If you would like any more information about this study, please contact us. We are very happy to answer your questions.

Name	**Title**	**Contact Number**
Noula Gibson	Chief Investigator	
Annie Chappell	Senior Physiotherapist	

I want to join the study. What to do now?
If you would like to take part in this research study, please talk to your parents. You will need to sign a consent form and so will your parents. Once you have done this, we can include you in our study.

Appendix 6 Information sheet for younger children

INFORMATION SHEET FOR CHILDREN (AGED UNDER 12)

A running group for children with cerebral palsy

What is the running group for?

For many sports and games, you have to run. But running is not easy for many kids with CP.

You can learn to run. People can teach it to you. But you need plenty of practice. This running group lasts for 3 months. If you do it, you will learn how to run and keep fit.

Who are we?

We are Noula Gibson and Annie Chappell. We are physiotherapists. We work at The Centre for Cerebral Palsy and Princess Margaret Hospital for Children. There will be a few other physiotherapists too.

Do I have to take part?

No, you do not have to take part in this study if you don't want to.

What will I have to do if take part?

You will come to two after-school sessions per week for 12 weeks. At these sessions, you will do exercises and practise your running. You can bring along a buddy to these sessions

You will also do exercises at home or in a gym 2 or 3 days a week.

We will test your fitness three times: one before you start the running group, one at the end of the running group, and a third time six months after you started the group.

Will the running group help me?

Yes. If you do the exercises, you will get fitter.

If I join the group, will it help others?

Yes. We will make the group as good as possible for children with CP who want to learn to run.

Will I hurt myself?

You might have some trips and falls. The physiotherapists will look after you and make sure the programme is safe.

Will you tell other people my results?

Yes, we will share the results of this study with other people. But we will not tell them your name.

I want to join the running group. What to do now?

Tell your mum or dad that you want to join the group.

THANK YOU FOR YOUR TIME

Appendix 7 Parent consent form

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service

FORM OF CONSENT

for Parents/Guardians

PLEASE NOTE THAT PARTICIPATION IN RESEARCH STUDIES IS VOLUNTARY AND SUBJECTS CAN WITHDRAW AT ANY TIME WITH NO IMPACT ON CURRENT OR FUTURE CARE.

I…….. have read

Given Names Surname

the information explaining the study entitled

A running training and lifestyle programme for children with cerebral palsy

I have read and understood the information given to me. Any questions I have asked have been answered to my satisfaction.

I agree to allow

……

(full name of participant and relationship of participant to signatory)

to participate in the study.

I understand my child may withdraw from the study at any stage and withdrawal will not interfere with routine care.

I agree that research data gathered from the results of this study may be published, provided that names are not used.

Dated..... day of..... 20.....

Child's Signature.....

(Where appropriate)

Parent or Guardian's Signature.....

I,..... have explained the above to the

(Investigator's full name)

signatories who stated that he/she understood the same.

Signature.....

Appendix 8 Child consent form

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service

CONSENT FORM

for children

You don't have to join this research if you don't want to. It is voluntary. You can stop if you want to. Either way, you will still receive just the same therapy and care from The Centre for Cerebral Palsy in the future.

I..... have read
Given Names Surname

the information about the study called

A running training and lifestyle programme for children with cerebral palsy

I have understood the information given to me.
I have had the chance to ask any questions I wanted.
If I asked any questions, somebody answered them for me.

I understand I can stop if I want to.
If I stop, I will still receive just the same therapy and care from The Centre for Cerebral Palsy in the future.

I agree that the researchers can share the information about me with other doctors and therapists, as long as my name is not used.

Dated..... day of..... 20.....

Signature.....

I,..... have explained the above to the

(Investigator's full name)

signatory who stated that he/she understood the same.

Signature.....

Appendix 9 Adverse event form

RUNNING GROUP

REPORT FORM FOR ADVERSE EVENTS

Please notify Chief Investigator [redacted] of all adverse events ASAP on [redacted]

Date: _____

Venue: _____

Physiotherapist(s): _____

Child: _____

What happened?

_____

What action was taken?

_____

FOLLOW-UP

Date: _____

Outcome

Signed: _____

[redacted] **notified:** ☐ Yes **Date notified: _____**Signed:_____

RUNNING IN CEREBRAL PALSY ADVERSE EVENT REPORT FORM
Patient's Initials: Patient ID:

2. List all the adverse events below.

AE No	Visit No	ADVERSE EVENT List each symptom separately	Date of Onset (dd/mm/yy)	Date of Resolution (dd/mm/yy) or √ - the box if continuing	Character of Event (√ - tick only one)	Severity 1 = Mild 2 = Moderate 3 = Severe 4 = NA	Treatment 1 = None 2 = Basic first aid needed 3 = Required medical intervention	Relationship Intervention 1 = Unrelated 2 = Possible 3 = Probable 4 = Definite	* Is the event classified as the SAE? (Yes / No)	Other Comments	Signature
				❑	❑ Intermittent ❑ Single event	1 2 3 4	1 2 3	1 2 3 4			
				❑	❑ Intermittent ❑ Single event	1 2 3 4	1 2 3	1 2 3 4			
				❑	❑ Intermittent ❑ Single event	1 2 3 4	1 2 3	1 2 3 4			
				❑	❑ Intermittent ❑ Single event	1 2 3 4	1 2 3	1 2 3 4			
				❑	❑ Intermittent ❑ Single event	1 2 3 4	1 2 3	1 2 3 4			

Investigator's Signature / / (dd/mm/yy)

Appendix 10 Home program sheet

Record the activity type and the length of time you participated in that activity for the week. Bring this sheet with you to all your running group classes.

Name:_____ DATE:_____ WEEK:_____

EXERCISE/ACTIVITY	MON	TUES	WED	THURS	FRI	SAT	SUN

OTHER COMMENTS:

Appendix 11 Ethical Approval

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service

Our Ref: 2014055EP

Dr Noula Gibson
Pysiotherapy
PMH, Roberts Road
Subiaco 6008

Dear Dr Gibson

HUMAN RESEARCH ETHICS COMMITTEE (HREC)
HREC REF 2014055EP
STUDY TITLE A running, training and lifestyle program for children with cerebral palsy

The ethics application for the project referenced above was reviewed by the PMH Human Research Ethics Committee (HREC) at its meeting on 17/07/2014. It has been approved and the following documents have been approved for use in this project.

Scientific Protocol Form 4B, version 1 01/05/2014
Attendance Sheet, Running Group
Information Sheet for Parents, 01/05/14
Information Sheet for children over 12, 01/05/14
Information Sheet for Children under 12, 01/05/14
Form of Consent - Parents/Guardians, 01/05/14
Consent Form for children, 01/05/14
Exercise Diary Sheets, 01/05/14
Adverse Event Form, Appendix 8, 01/05/14

Approval of this project from PMH HREC is valid to and on the basis of compliance with the 'Conditions of HREC Approval for a Research Project' (attached).

[Note: If additional sites are recruited prior to the commencement of, or during the research project, the Coordinating Principal Investigator is required to notify the HREC. Notification of withdrawn sites should also be provided to the HREC in a timely fashion.

A copy of this ethical approval letter must be submitted by all site Principal Investigators to the Research Governance Office or equivalent body or individual at each participating institution in a timely manner to enable the institution to authorise the commencement of the project at its site/s.

This letter constitutes ethical approval only.

This project cannot proceed at any site until separate site authorisation has been obtained from the CE, or delegate, of the site under whose auspices the research will be conducted at that site.

The PMH HREC is registered with the Australian Health Ethics Committee and operates according to the NHMRC National Statement on Ethical Conduct in Human Research and International Conference on Harmonisation – Good Clinical Practice.

The HREC's Terms of Reference, Standard Operating Procedures, membership and standard forms are available from http://www.pmh.health.wa.gov.au/development/resources/ethics.htm or from the Ethics Office. Should you have any queries about the HREC's consideration of your project, please contact Ethics Office.

Please quote the above trial number 2014055EP on all correspondence associated with this trial.

Yours sincerely

Dr Mark Salmon
Executive Director
Medical Service

1/08/2014

* The Ethics Committee is constituted, and operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Research Involving Humans

Government of **Western Australia**
Department of **Health**
Child and Adolescent Health Service

CONDITIONS OF HREC APPROVAL FOR A RESEARCH PROJECT

The following general conditions apply to the research project approved by the Human Research Ethics Committee (HREC) and acceptance of the approval will be deemed to be an acceptance of these conditions by all investigators involved in the research project:

1. The responsibility for the conduct of projects lies with the Coordinating Principal Investigator (CPI), all correspondence should be signed by CPI.
2. Projects that do not commence within 12 months of the approval date may have their approval withdrawn and the project closed. The CPI must outline why the project approval should stand.
3. The submission of an application for HREC approval will be deemed to indicate that the investigator/s and any sponsor recognises the approving HREC is registered with the National Health and Medical Research Council (NHMRC) and that it complies in all respects with the National Statement on Ethical Conduct in Human Research and all other national and international ethical requirements. **The HREC will not enter into further correspondence on this point.**
4. A list of attendance at a specific meeting is available on request, but no voting records will be provided.
5. The CPI will notify the HREC of his or her inability to continue as CPI and will provide the name and contact information of their replacement. Failure to notify the HREC can result in the project being suspended or approval withdrawn.
6. The CPI will notify the HREC of any departures of named investigators. The CPI will also notify the HREC if any new investigators and/or sites join the project that will utilise the HREC's approval.
7. The CPI will inform the HREC about any changes to the project. The CPI is responsible for submitting any amendments to the approved documents listed on the approval letter, or any new documentation to be used in the project. Any new or amended documentation should be submitted in a timely manner and cannot be implemented at any participating site until they have received HREC approval.
8. The CPI is responsible for reporting adverse events, indicating whether or not the project should continue. Reporting requirements are as per the WA Health Research Governance and Single Ethical Review Standard Operating Procedures. Additional reports other than those outlined that are submitted to the HREC will be returned without acknowledgement. The HREC can request additional reporting requirements as a special condition of a research project.
9. Where a project requires a Data Safety Monitoring Board (DSMB) it is the CPI's responsibility to ensure this is in place before the commencement of the project and the HREC notified of this. All relevant reports from the DSMB should be submitted to HREC.
10. For projects where the site is acting as the sponsor (ie. investigator initiated project) it is the responsibility of the CPI to report serious and unexpected drug/device reactions, as well as other reactions/events to the Therapeutic Goods Administration (TGA). Please refer to TGA website for further information and the relevant forms (see http://www.tga.gov.au/pdf/clinical-trials-guidelines.pdf p71 for medications or p77 for devices).
11. If this project involves the use of an implantable device a properly monitored and up to date system for tracking participants is to be maintained for the life of the device in accordance with the National Statement section 3.3.22 (g).

12. The investigator is responsible for notifying the Therapeutic Drugs Administration of a device incident in accordance with the National Statement section 3.3.22 (g).

13. An annual report on an approved research project will be required on the anniversary date of the project's approval. HREC approvals are subject to the submission of these reports and approval may be suspended if the report is not submitted.

14. The HREC has the authority to audit the conduct of any project without notice. Exercise of this authority will only be considered if there are grounds to believe that some irregularity has occurred, if a complaint is received from a third party or the HREC decides to undertake an audit for Quality Improvement purposes.

15. The HREC can conduct random monitoring of any project. The CPI will be notified if their project has been selected. The CPI will be given a copy of the monitor's report along with the HREC and Research Governance Office (RGO) at each site.

16. Complaints relating to the conduct of a project should be directed to the HREC Chair and will be promptly investigated according to the Committee's complaints procedures.

17. CPI are reminded that records of consent or authorisation for participation in a project form part of the Acute Hospital Patient Record and should be stored with that record in accordance with the WA *Health Patient Information Retention and Disposal Schedule (Version 2) 2000.* A copy of the 'Participant Information Sheet' should also be included in the medical records as part of informed consent documentation.

18. The duration of HREC approval for a project is 3 year (with the option of 5 years) from the date of approval. The date of approval expiry is stipulated in the HREC approval letter.

19. If the project is to continue beyond the stipulated approval expiry date a request for an extension should be submitted prior to that expiry date. One extension of 3 years can be granted but approval beyond this time period may necessitate further review by the HREC.

20. Once the approval period has expired, the CPI is required to submit a final report. If the report is not received within 30 days the project will be closed and archived. An outstanding final report could impact on the CPI's ability to apply for approval for future projects.

21. If a project is suspended or terminated by the CPI, or a project sponsor, the CPI must immediately inform the HREC and the RGO at each site of this and the circumstances necessitating the suspension or termination of the project. Such notification should include information as to what procedures are in place to safeguard participants.

22. If a project fails to meet these conditions the HREC will contact the investigator(s) to request they rectify the identified issues. If, after being contacted by the HREC, the issues are not addressed the HREC approval will be withdrawn. The HREC will notify the RGO at each site within WA Health that work may no longer be conducted in relation to the project other than that concerning the participants safety.

Appendix 12 Reciprocal ethical approval

Curtin University

Memorandum

To	Professor Gary Allison, Physiotherapy
From	Professor Peter O'Leary, Chair Human Research Ethics Committee
Subject	Protocol Approval **HR 219/2014**
Date	4 December 2014
Copy	Mrs Leanne (Annie) Chappell, Physiotherapy Dr Susan Morris, Physiotherapy Dr Noula Gibson, PMH Dr Gavin Williams, Epworth Healthcare

Office of Research and Development
Human Research Ethics Committee

TELEPHONE 9266 2784
FACSIMILE 9266 3793
EMAIL hrec@curtin.edu.au

Thank you for your application submitted to the Human Research Ethics Committee (HREC) for the project titled "*The effect of a running and lifestyle intervention in children and adolescents with cerebral palsy.*". The Committee notes the prior approval by PMH HREC (2014055EP) and has reviewed your application consistent with Chapter 5.3 of the *National Statement on Ethical Conduct in Human Research*.

- You have ethics clearance to undertake the research as stated in your proposal.
- The approval number for your project is **HR 219/2014**. *Please quote this number in any future correspondence*.
- Approval of this project is for a period of four years **04-12-2014** to **17-07-2017**.
- Annual progress reports on the project must be submitted to the Ethics Office.
- **It is your responsibility, as the researcher, to meet the conditions outlined above and to retain the necessary records demonstrating that these have been completed. See: Western Australian University Sector Disposal Authority (WAUSDA).**
- If you are a Higher Degree by Research student, data collection must not begin before your Application for Candidacy is approved by your Faculty Graduate Studies Committee.
- The following standard statement **must be** included in the information sheet to participants: *This study has been approved by the Human Research Ethics Committee of (PMH HREC) and Curtin University* **219/2014.**

Applicants should note the following:

It is the policy of the HREC to conduct random audits on a percentage of approved projects. These audits may be conducted at any time after the project starts. In cases where the HREC considers that there may be a risk of adverse events, or where participants may be especially vulnerable, the HREC may request the chief investigator to provide an outcomes report, including information on follow-up of participants.

The attached **Progress Report** should be completed and returned to the Secretary, HREC, C/- Office of Research & Development annually.

Our website https://research.curtin.edu.au/guides/ethics/non_low_risk_hrec_forms.cfm contains all other relevant forms including:

- Completion Report (to be completed when a project has ceased)
- Amendment Request (to be completed at any time changes/amendments occur)
- Adverse Event Notification Form (If a serious or unexpected adverse event occurs)
- Western Australian University Sector Disposal Authority (WAUSDA)

Yours sincerely

Professor Peter O'Leary
Chair Human Research Ethics Committee

Curtin University

HREC Progress Report

The Form is to be completed and returned to *the Secretary, Human Research Ethics Committee, c/- Office of Research & Development.* hrec@curtin.edu.au

If a Form C Co-ordinator, approved your application please submit your completed form to your school Form C Co-ordinator.

Annual completion of this form fulfils researchers' obligations under section 5.5.5 of the National Statement on Ethical Conduct in Human Research.

All questions must be answered or the Form will not be processed.

Approval Number:	
PROJECT TITLE:	

1	**Please confirm the project is proceeding exactly as specified in the protocol.**	**YES☐**	**NO☐**
If NO, please provide details_____ *(Attach additional comments on a separate sheet of paper if necessary)*			
2	**Have any ethics related issues emerged in regard to this project since you received Ethics' Committee approval? (e.g. breach of confidentiality, harm caused, inadequate consent or disputes on these).**	**YES☐**	**NO☐**
If yes, please provide details_____ *(Attach additional comments on a separate sheet of paper if necessary)*			
3	**Have any ethics related issues in regard to this project been brought to your attention by others? (e.g. study respondents, organisations that have given consent, colleagues, the general community etc).**	**YES☐**	**NO☐**
If yes, please provide details _____ *(Attach additional comments on a separate sheet of paper if necessary)*			
4	**Please outline the progress made to date.** **(e.g. Number of participants recruited; Data collected / analysed; Papers published)**	**YES☐**	**NO☐**
(Attach additional comments on a separate sheet of paper if necessary)			
5	**Please detail what arrangements have been made for the ongoing storage and security of the research records in accordance with the Western Australian University Sector Disposal Authority (WAUSDA).** http://uim.curtin.edu.au		
(Attach additional comments on a separate sheet of paper if necessary)			

Investigator:		**Signature:**	
		Date:	
Co-Investigator:		**Signature:**	

School/Department:			
Head of Area: *Or equivalent*		**Signature:**	
Date:			

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