Curtin School of Allied Health

Aquatic high intensity interval training in non-athletes and disability populations

Julie Depiazzi

000-0001-5561-3427

This thesis is presented for the Degree of
Master of Philosophy (Physiotherapy)
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Declaration

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgement has been made.

This thesis contains no material which has been accepted for the award of any other degree or diploma in any university.

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 study received human research ethics approval from the Curtin University Human Research Ethics Committee (EC00262), Approval Number # HRE20170282

Signature: ..... Date: .....

Abstract

This thesis comprises two studies: a systematic review and a pilot randomised controlled trial. This review aimed to evaluate what is known regarding aquatic high intensity interval exercise training and its effect on aerobic capacity and body composition in populations that are not athletic. This knowledge was then used to develop, implement and evaluate the feasibility of this type of training for young people with cerebral palsy (CP).

Study one: systematic review and meta-analysis

Objective: In a non-athletic population, to (1) investigate the effectiveness of high intensity interval training (HIIT) in an aquatic environment (A-HIIT) on aerobic performance, strength and body composition, (2) report on the safety of this approach, and (3) describe the risk of bias and quality of reporting for the included studies.

Design: Systematic review and meta-analysis.

Method: A systematic search was undertaken of six databases: PubMed, MEDLINE, CINAHL, EMBASE, SPORTDiscus and the Cochrane Library. Trials were eligible for inclusion if they compared the effect of A-HIIT in a non-athletic population with a control group that received no exercise training. Data were extracted independently by two reviewers and meta-analyses were undertaken using a random effects model. Risk of bias was assessed using Cochrane's risk of bias tool. All studies were graded using the Physiotherapy Evidence Database (PEDro) and the Consensus on Exercise Reporting Template (CERT).

Results: We reported on eight studies across 13 papers. These studies included 377 participants (A-HIIT *n* = 212; control *n* = 165), the majority of whom were female (89%), with the mean age ranging from 21.7 to 69.0 years (A-HIIT) and 21.7 to 69.8 years (control). Those who completed A-HIIT programs demonstrated greater aerobic performance (standardised mean difference [SMD] 0.69, 95% confidence interval [CI] 0.39 to 0.98); I^2 = 0%; *n* = 191) and improved lower limb muscle strength (SMD 0.30, 95% CI 0.04 to 0.56; I^2 = 0%; *n* = 237). No differences were seen in measures of body composition or adverse events. The (mean ± standard deviation [SD]) PEDro and CERT scores were 4.9 ± 1.5 and 15.1 ± 2.1, respectively.

Conclusion: In a non-athletic population, A-HIIT was safe and appears to have improved aerobic performance and lower limb strength. The exercise intervention was well described, and monitoring and reporting of exercise intensity in water was feasible.

Study two: pilot randomised controlled trial

Objective: To investigate the feasibility of novel A-HIIT for adolescents with CP at GMFCS (Gross Motor Function Classification System) level II.

Design: Pilot randomised controlled trial.

Method: Following initial assessments, participants were randomised to usual care or 10 weeks of twice-weekly A-HIIT. Each class comprised 10 standardised intervals of one minute exercise separated by one minute rest. Participants' heart rates were monitored using telemetry with high intensity exercise defined as the attainment of ≥80% of maximum heart rate.

Measures: Primary outcomes related to the feasibility of the protocol to progress to a definitive trial. Measurements collected before and after the intervention period comprised peak rate of oxygen uptake, lean muscle mass, pain and health-related quality of life. Participant feedback was obtained.

Results: Of 46 participants approached, 17 consented, resulting in a recruitment fraction of 37% (95% CI 23 to 52). Twelve completed baseline assessments (mean age 14 years 7 months, SD 2 years 0 months; 5 males). Heart rate data were available for 1180 stations of the 1190 stations completed (99%, 95% CI 99 to 100). High intensity exercise was achieved in 1111 stations (93%, 95% CI 92 to 95). All randomised participants completed the study and reported that the intervention was fun, improved function and provided friendship opportunities. There were no major adverse events or exacerbation of pain.

Conclusion: A-HIIT in adolescents with CP at GMFCS level II is feasible. Recruitment was challenging, but those who agree to participate are likely to complete a trial. Uncertainty remains on the efficacy of the intervention, highlighting the need for a large definitive trial.

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Statement of originality

This thesis is presented for the degree of Master of Philosophy (Physiotherapy) at Curtin University, Western Australia. Studies were undertaken between October 2016 and November 2020, through the School of Physiotherapy and Exercise Science at Curtin University, in association with the Physiotherapy Department and the Department of Respiratory Medicine at Perth Children's Hospital, Western Australia.

This research project was developed in association with my supervisors, who have been involved in editing both the thesis and all associated publications.

All material presented in this thesis is original.

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Acknowledgement of Country

We acknowledge that Curtin University works across hundreds of traditional lands and custodial groups in Australia, and with First Nations people around the globe. We wish to pay our deepest respects to their ancestors and members of their communities, past, present, and to their emerging leaders. Our passion and commitment to work with all Australians and peoples from across the world, including our First Nations peoples, are at the core of the work we do, reflective of our institutions' values and commitment to our role as leaders in the Reconciliation space in Australia.

Contents

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Abbreviations

A-HIIT	aquatic high intensity interval training
CI	confidence interval
CERT	Consensus on Exercise Reporting Template
CP	cerebral palsy
CPET	cardiopulmonary exercise testing
DXA	dual energy X-ray absorptiometry
GMFCS	Gross Motor Function Classification System
HIIT	high intensity interval training
HR	heart rate
HRpeak	peak heart rate achieved during an exercise interval
HRQoL	health-related quality of life
MD	mean difference
PCH	Perth Children's Hospital
PEDro	Physiotherapy Evidence Database
PedsQL™	Pediatric Quality of Life Inventory v4.0
RCT	randomised controlled trial
SD	standard deviation
SMD	standardised mean difference
VO_{2max}	maximum rate of oxygen consumption
VO_{2peak}	peak rate of oxygen uptake

Work arising as part of this thesis

Published papers

Depiazzi J, Forbes R, Gibson N, Smith N, Wilson A, Boyd R, Hill K. The effect of aquatic high-intensity interval training on aerobic performance, strength and body composition in a non-athletic population: systematic review and meta-analysis. *Clinical Rehabilitation* 2019; 33: 157–170. https://doi.org/10.1177/0269215518792039

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Platform presentations/Posters

A pilot randomised controlled trial: can a water based exercise program improve aerobic capacity in adolescents with cerebral palsy. [Presentation]. Telethon Kids Institute and Respiratory Medicine Perth Children's Hospital Symposium. Rottnest Island. November 2019.

High intensity interval training in an aquatic environment is feasible in adolescents with cerebral palsy: pilot randomised controlled trial. [Poster]. Australasian Academy of Cerebral Palsy and Developmental Medicine conference. Perth. March 2020.

High intensity interval training in an aquatic environment does not increase pain in adolescents with cerebral palsy. [Presentation]. Australasian Academy of Cerebral Palsy and Developmental Medicine conference. Perth. March 2020.

Adolescents with cerebral palsy can participate in high intensity aquatic exercise. [Presentation]. Grand round invitation, Perth Children's Hospital. Perth. October 2020.

High intensity interval training in an aquatic environment is feasible in adolescents with cerebral palsy: pilot randomised controlled trial. [Poster]. Child and Adolescent Health symposium. Perth. November 2020.

High intensity interval training in an aquatic environment does not increase pain in adolescents with cerebral palsy. [Presentation]. Child and Adolescent Health symposium. Perth. November 2020.

Chapter 1:
Introduction

1 Introduction

This chapter describes the background and rationale for this program of research. The research questions are outlined, and the novelty and significance are described. Components of this research have been published (see Work arising as part of this thesis for details).

1.1 Background

1.1.1 Aquatic high intensity interval training in non-athletes

Decreased general aerobic performance has been associated with shorter life expectancy, increased cardiometabolic risk, and reductions in activity participation, exercise enjoyment, and health-related quality of life.[1-3] While the health benefits of exercise have been well established, both healthy and clinical populations report difficulty in training at an exercise dose sufficient to maintain or improve aerobic performance.[4-6] Barriers to exercise such as boredom, deconditioning, pain and movement difficulties can be further compounded by environmental factors such as location and weather.[7, 8] To promote participation in exercise at sufficient dose to maintain or improve aerobic performance, there is a need to explore novel approaches to exercise prescription and the environment in which it is conducted.

One novel approach to exercise prescription may include changing the training method from continuous to interval-based exercise, and also modifying the exercise environment from land to water. High intensity interval training (HIIT) is an exercise method that involves interspersing periods of high levels of exercise intensity with periods of rest or less exertion. In healthy young adults, HIIT has been shown to

increase muscle oxidative potential and endurance capacity within a 2-week training period.[9] It is also time efficient with similar adaptions in skeletal muscle and exercise performance seen in 2.5 hours of HIIT versus 10.5 hours of conventional continuous training in these populations.[10] An advantage of an interval training rest period is relief from unpleasant symptoms of exertion such as exhaustion, fatigue and pain, in both clinical and healthy populations.[11-13] These rest periods likely contribute to the greater ratings of perceived enjoyment for HIIT compared to continuous exercise in some studies, despite HIIT programs producing higher ratings of perceived exertion and participants achieving higher training work rates.[12, 14]

In addition to utilising interval training principles, barriers to exercise may be further reduced by changing the exercise environment from land to water. An aquatic training environment is used by health professionals for maintaining or improving aerobic performance, while limiting some uncomfortable symptoms of exercise exertion such as joint pain.[15, 16] It is believed that the properties of an aquatic environment such as buoyancy, hydrostatic pressure, viscosity and warmth reduce joint loading and minimise impediments to exercise found in land-based training such as pain, and balance and movement difficulties.[17, 18] In lessening the impact of these impediments, the training dose that is tolerated is likely to be greater, making vigorous training intensities more feasible and, in turn, improving the likelihood of increasing aerobic performance.[18-21] Training in an aquatic environment is also reported to be fun and enjoyable and is therefore more likely to promote ongoing exercise participation.[22, 23]

The effectiveness of interventions that combine HIIT with the benefits offered by an aquatic environment (i.e. aquatic HIIT, or A-HIIT) has not been systematically reviewed in a non-athletic or clinical population.

1.1.2 Aquatic intensity interval training in adolescents with cerebral palsy

Of particular interest is the novel use and potential benefits of an A-HIIT training method in populations that present with impairments that limit the intensity that can be reached during land-based exercise. One example of such a population is children and adolescents with cerebral palsy (CP). This clinical population is the focus for the pilot randomised controlled trial (RCT) conducted within this body of work.

In Australia, a child is born with CP every 15 hours, making it the most common physical disability in childhood.[24] CP describes a disorder of posture and movement caused by a non-progressive lesion to the developing brain.[24] It is a heterogeneous disorder and is associated with various types and degrees of motor impairment affecting posture, gross motor and fine motor function, oromotor function, and cognitive function.[24, 25] In addition to physical challenges, children with CP have decreased aerobic capacity compared to their typically developing peers and this can be associated with lower levels of physical activity, increased cardiometabolic risk and shortened life expectancy.[25-29] Improvements in aerobic capacity in children with CP using land-based exercise training have been demonstrated, but barriers to the training dose tolerated and long-term participation include pain during exercise, fatigue, poor balance and difficulties with co-ordination.[26, 29, 30]

The paucity of studies investigating the addition of HIIT to the aquatic environment for children with CP means that a number of challenges need to be considered prior to conducting a definitive RCT.[31] For example, the ability to measure the 'real-time' heart rate responses of participants in water to titrate high intensity is unknown, as is the tolerability of this type of training for this population, and the effect of potential

physical barriers such as pain. Further, the feasibility of quantifying improvements in aerobic capacity following aquatic interventions in this population using gold standard measures, namely peak rate of oxygen uptake measured during a laboratory-based cardiopulmonary exercise test, is unclear, making prospective sample size calculations challenging. These factors highlight the need for a pilot study prior to undertaking a definitive trial.[32]

1.2 Research questions

This thesis explores: (1) the design and efficacy of A-HIIT programs; (2) the ability and tolerability of adolescents with CP to participate in A-HIIT; and (3) the feasibility of measuring outcomes, including work rate in the aquatic environment. It encompasses two studies.

Study 1:

Study 1 began as an exploration of what was known about the efficacy and design of A-HIIT programs in young people with disability. Due to the paucity of published information, it became evident that the systematic review of the literature had to be broadened to a relatable population, that being non-athletes. Therefore, the aims of Study 1 were:

1) To determine in non-athletes the effect of A-HIIT on changes in aerobic capacity and body composition

2) To determine any safety concerns with this type of training in non-athletes

3) To utilise the information gathered from this systematic review to inform the design and measurement of Study 2.

Study 2:

Study 2 was a pilot RCT that sought to explore the ability and tolerability of adolescents with CP who are ambulant to exercise at high intensity in water.[33] The main aim of Study 2 was to determine if the protocol in its current format was feasible to progress to an RCT without modification. Factors that were evaluated to make this decision were:

1) recruitment fraction
2) tolerability of intervention
3) fidelity of telemetry heart rate monitoring in an aquatic environment
4) tolerability and feasibility of outcome assessments
5) domains including completion rates, missing data, estimates, variances and confidence intervals for the difference between the control and intervention groups for clinical and participant-reported measures
6) barriers and enablers of study completion.

1.3 Significance and novelty

The benefits of exercise across all populations on physical health and health-related quality of life have been well documented.[1-3] Barriers to exercise participation continue to exist, however, in both healthy and clinical populations, including boredom, deconditioning, pain and movement difficulties.[7, 8] The potential of A-HIIT programs to overcome some of these barriers to allow training at higher work rates has not been systematically explored in non-athletic and/or clinical populations. A-HIIT programs

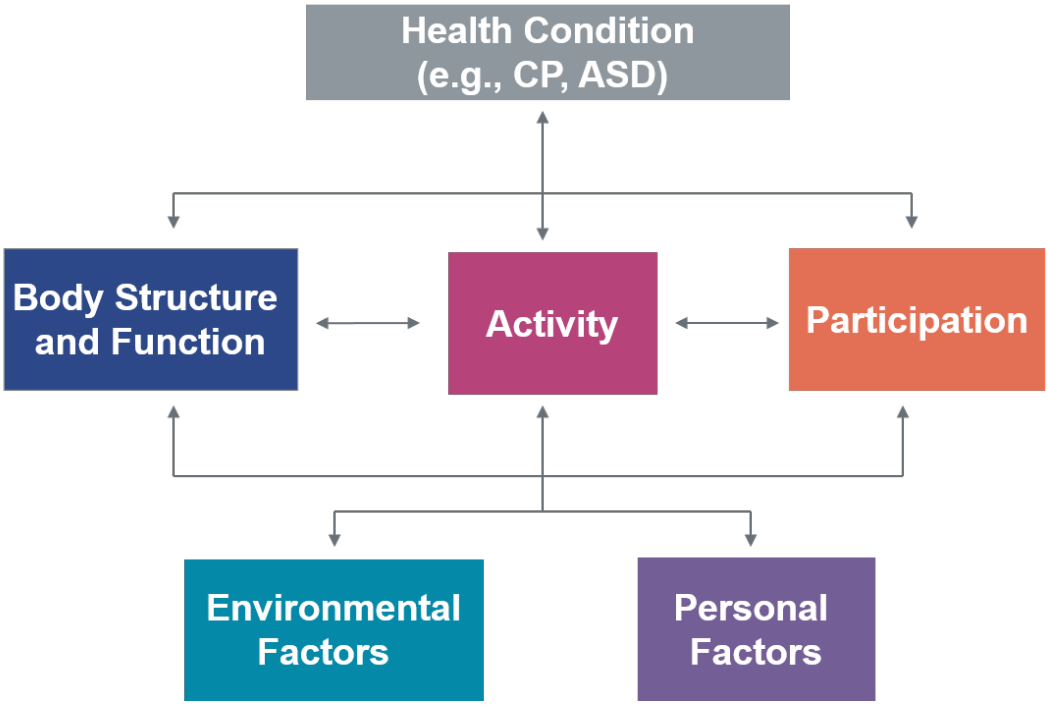
have the potential to make exercise more feasible and tolerable for populations that find it difficult to achieve effective exercise intensities and/or who have symptomatic barriers to exercise such as pain and fatigue.[21] This is especially relevant for children and adolescents with disabilities like CP who are expected to adhere to ongoing, often intensive, physiotherapy programs from an early age and may have a perception that exercise is difficult and unenjoyable,[26] contributing to long-term health-related concerns.

Registry studies in Australia and Sweden demonstrate that despite vast improvement in medical care, the survival of young people with CP has not improved in the past 40 years.[34] Although CP is not a progressive condition, chronic diseases of adulthood (e.g. diabetes, cardiovascular disease) that lead to morbidity and mortality are more prevalent in people living with CP regardless of their level of independence.[29, 34, 35] A recent study undertaken in Western Australia found that people with CP who died of causes related to ageing (e.g. cancer, cardiometabolic disease) died at a younger age then their typically developing peers.[34] Similar studies in the United Kingdom and the United States also report a higher prevalence of chronic diseases in adults with CP than adults without CP, such as diabetes (9.2% vs 6.3%), stroke (4.6% vs 2.3%) and arthritis (31.4% vs 17.4%).[29, 36]

In people with CP, health-related concerns begin early in life. In adolescents with CP, lower aerobic capacity is associated with lower levels of physical activity and increased cardiometabolic risk.[28] In the general population, increasing aerobic capacity is associated with improvements in cardiometabolic health, but intervention studies in children with CP aimed at showing improvements in aerobic capacity are lacking.[2, 28, 37] This is in part due to intrinsic challenges and limitations to vigorous exercise

participation for these children, such as pain and fatigue, and difficulties in conducting a robust study to measure change in this cohort.[26, 30, 37]

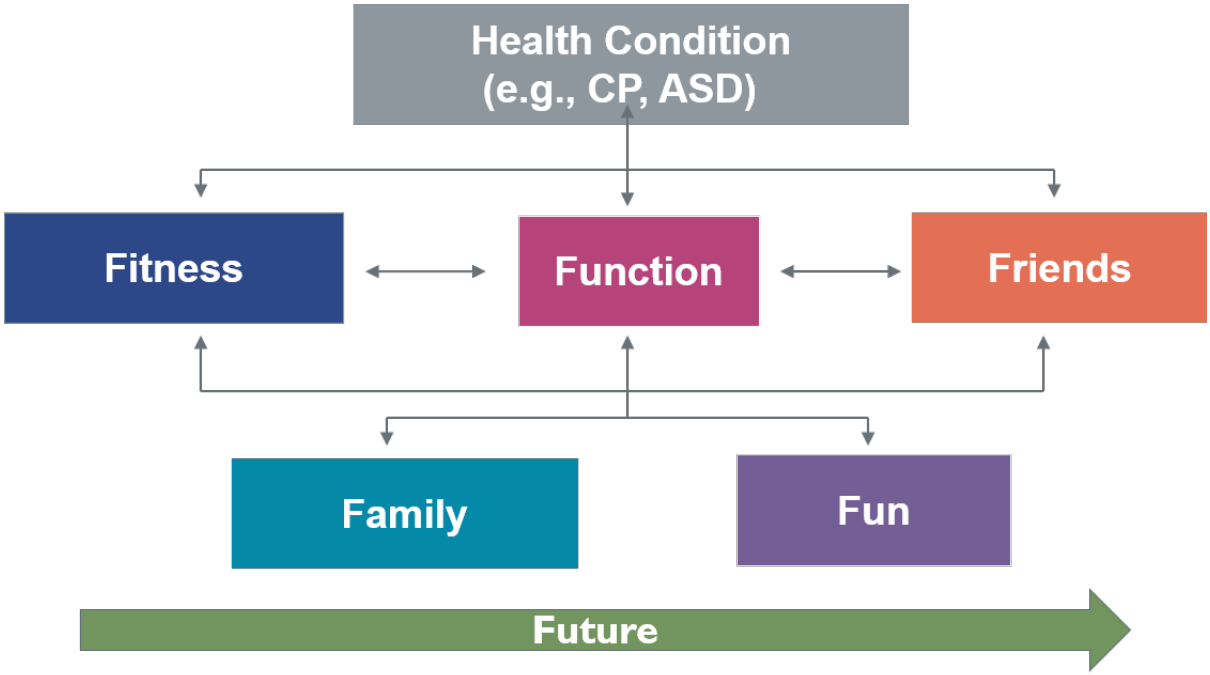
The importance of managing preventable chronic disease from childhood in people with CP by increasing physical activity is conceptualised in the framework of the World Health Organisation's International Classification of Functioning, Disability and Health (ICF) (Figure 1-1.1: The ICF model: interaction between components of the International Classification of Functioning, Disability and Health).[38] The ICF framework illustrates how body structure and function, activity, participation, and environmental and personal factors are inter-related and equally influence health and functioning.



ASD = autism spectrum disorder; CP = cerebral palsy

Figure 1-1.1: The ICF model: interaction between components of the International Classification of Functioning, Disability and Health framework[38]

This has been further interpreted by leading disability researchers and clinicians into the six ‘F-words’ for childhood disability which include ‘Fitness’ as a focus for therapeutic management of children with disability (Figure 1-1.2).[39] This model also considers that for a therapeutic activity to be feasible in young people, it must not only be physiologically achievable, it also needs to be age appropriate, encourage friendships and be fun.



ASD = autism spectrum disorder; CP = cerebral palsy

Figure 1-1.2: The F-words in childhood disability overlayed on the International Classification of Functioning, Disability and Health framework[39]

This body of work investigates the design, efficacy, tolerability and safety of A-HIIT programs. Using this knowledge, a study was designed, which if scaled up, would evaluate the effect of A-HIIT on aerobic capacity in adolescents with CP. The goal of this thesis is to demonstrate that a novel A-HIIT program aimed at promoting fitness in children with disability is feasible and can encompass all aspects of the ICF framework.

Chapter 2:
Literature review

2 Literature review

This literature review comprises five parts.

Part 1 informs the current scope of work on the benefits of aquatic high intensity interval training (A-HIIT). Part 2 describes the disorder of cerebral palsy (CP) and its prevalence. Part 3 explores the limitations related to exercise participation for those who have CP, and the impact of this on morbidity, mortality and health-related quality of life. Part 4 describes previous research into land-based exercise in children with CP. Part 5 explains the reasons why a pilot study design was used to investigate the implementation of A-HIIT in adolescents with CP, and Part 6 summarises this gap in knowledge.

Chapter 3 presents a systematic review of the literature that has reported on the efficacy of A-HIIT programs in non-athletes. Therefore, the scope of this chapter is limited to a narrative review that presents an argument for the *potential* of A-HIIT as a modality to improve aerobic capacity in children with CP.

2.1 Aquatic high intensity interval training in young and clinical populations

Independently, high intensity interval training (HIIT)[40] and aquatic therapy[41, 42] interventions have demonstrated improvements in aerobic capacity for clinical and young populations. In adolescents, a 2015 systematic review suggests land-based HIIT interventions can improve aerobic capacity (mean difference [MD] 2.6 mL/kg/min, 95% confidence interval [CI] 1.8 to 3.3; $p < 0.001$).[43] Further, adolescent participants have reported a preference for HIIT compared to traditional continuous training

programs.[22] Studies investigating the benefits of HIIT combined with an aquatic environment have been limited to adult clinical populations for outcomes related to pain,[44] and cardiometabolic[45] and musculoskeletal health.[21] While these studies show the potential for A-HIIT to be beneficial in clinical populations, the appropriate design and measurements, feasibility of implementation, and tolerability of these types of interventions in children and populations with disability remains unknown.

2.2 Cerebral palsy: definition, incidence, motor impairment and healthcare cost

Cerebral palsy is a non-progressive disorder of posture or movement resulting from a defect or lesion of the brain that is acquired early in life.[24] It can be defined as follows:

> Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems.[46] (p.9)

It is the most common cause of childhood disability, affecting between 1.5 and 3.8 children per 1000 births.[47]

CP is a heterogeneous disorder. Children present with gross motor impairments that can be graded using the Gross Motor Function Classification System (GMFCS), ranging from mild (i.e. independently ambulant without aids; GMFCS level I) to severe (i.e. dependent on a wheelchair and others for mobility; GMFCS level V) (Figure 2-1)[48] The

GMFCS is based on self-initiated movement. Distinctions between levels are based on functional limitations, the need for handheld mobility devices (e.g. walker) and, to a much lesser extent, quality of movement.[48]

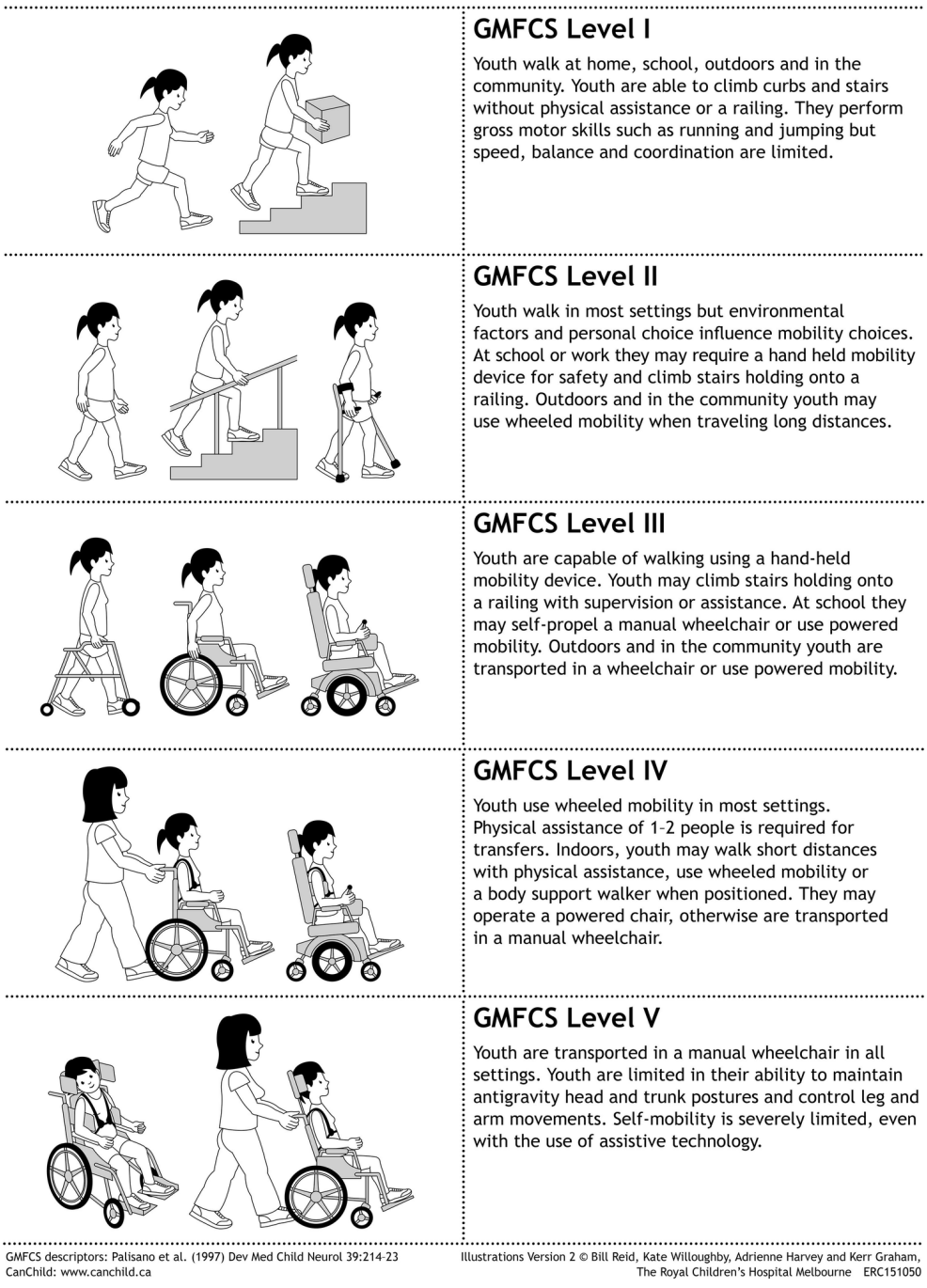


Figure 2-1: Description and illustration of the Gross Motor Function Classification System (GMFCS) – expanded and revised for adolescents between 12 and 18 years[48]

There are around 36,000 people living with CP in Australia, with 58% of these classified as GMFCS level I to II.[49] In young children, early intervention health costs are significant. The Centers for Disease Control and Prevention in the United States reported that, of all children enrolled in Medicaid in 2005, those with CP required the highest expenditure and had 10 times higher medical costs compared to children without CP.[50] Adults living with CP, regardless of GMFCS level, are reported to have a higher proportion of comorbidities such as heart disease and stroke than their peers,[29, 35] leading to earlier mortality.[34] In 2018, the total cost to the Australian economy for Australians living with CP was A$5.2 billion.[49]

2.3 Reduction in aerobic capacity in adolescents with cerebral palsy

While the disorder of posture and movement with CP is non-progressive, secondary limitations such as reduction in aerobic capacity can increasingly impact health-related quality of life with age. Aerobic capacity is a performance measure indicative of cardiovascular fitness; it is the ability of the heart and lungs to deliver oxygen to muscles, and the muscles' ability to extract it. In children, it is reported as the peak rate of uptake of oxygen (VO_{2peak}) measured during a maximal exercise test and is commonly expressed as mL per kilogram per minute (mL/kg/min).

In children and adolescents with CP, aerobic capacity has been found to be reduced when compared with their typically developing (TD) peers.[26, 37, 51, 52] This reduction in aerobic capacity is evident not only in those with severe motor impairment (GMFCS IV and V), but also those with relatively mild motor impairments (GMFCS I and II).[27] Specifically, earlier work has reported that children and adolescents with GMFCS II CP have a mean VO_{2peak} of (mean ± standard deviation) 33.9 ± 7.8 mL/kg/min, which is

less than that measured in their typically developing peers, being 41.0 ± 7.2 mL/kg/min ($p < 0.001$).[27] Other studies in children with GMFCS I and II CP have reported a range of values for VO_{2peak} (32.7 mL/kg/min to 42.0 mL/kg/min[53-55]), but it is consistently reported as lower than their typically developing peer control group. This consistency in data across international studies suggests the reduction in aerobic capacity cannot be contributed to differences in local testing practice or therapeutic interventions.

The reason for this reduced aerobic capacity in children with GMFCS I and II CP is unknown, but is likely due to a combination of primary limitations related to neurological function, such as respiratory muscle spasticity and joint contracture,[56] and secondary limitations related to immobility such as deconditioning from reduced participation in moderate to vigorous intensity physical activity.[26, 27, 56] Furthermore, the difference in aerobic capacity between those with and without CP appears to increase with age, giving weight to the argument that the cause is likely to be secondary to the primary impairment in neurological function.[57]

Improving aerobic capacity through vigorous exercise is possible in young people with CP, but is challenging even for those children who are independently ambulant.[26, 58-60] Body and structure impairments, such as hypertonia, muscle weakness, and muscle and joint contracture, are known to cause inefficiencies in movement and posture resulting in higher energy costs, poor gait efficiency, excessive fatigue and fear of falling.[61, 62] These can impact a child's independence and therefore activity participation.[61, 63] In addition, children with CP often have comorbidities, including visual and visuospatial problems, and balance and navigational difficulties.[64, 65] Cognitive, attentional and executive functioning problems such as impulsivity and poor judgement are also commonly seen in children with CP.[64] These additional challenges may limit parental

willingness for their children to participate in land-based activities that rely on competency in these abilities to ensure safety.[63]

Pain in young people with CP is highly prevalent and has been well documented in cross-sectional studies and systematic reviews.[30, 66-68] In adolescents with CP, up to 74% (95% CI 69% to 79%; *n* = 667) self-report weekly pain, with 45% of those reporting pain during physiotherapy.[30] Females are more likely to describe pain (odds ratio [OR] 1.28, 95% CI 1.12 to 1.47; *n* = 3545).[68] The most frequent source of pain is in the lower extremities, with pain prevalence increasing with age, even between early childhood and adolescence (4 to 18 years) (OR 1.07, 95% CI 1.06 to 1.09).[30, 67, 68] Importantly, musculoskeletal pain has been found to be a major predictor of emotional and behavioural issues in this population, which can further limit exercise participation.[69] It is likely that this increase in pain, reduction in health-related quality of life and decreased physical activity participation with age are partly responsible for the reduced aerobic capacity noted by adolescence.[57]

2.4 Interventions to improve aerobic capacity in cerebral palsy

Data supporting the effectiveness of interventions to improve aerobic capacity in young people with CP are scarce. This is due to the very small number of randomised controlled trials (RCTs), inadequate methodological design and reporting of studies, and difficulties in both recruitment and measuring outcomes to assess efficacy. The focus of this section is a critical review of studies that have attempted to improve aerobic capacity in children and adolescents with CP using land-based exercise training given there are no RCT investigating aerobic capacity change as the primary outcome in aquatic studies.

In 2011, a core set of exercise tests for evaluating the effects of an exercise training program in children and adolescents with CP was developed using a Delphi study and expert recommendations.[70] One of these tests is cardiopulmonary exercise testing (CPET), which is considered a gold standard.[37, 70] To date, it appears there are only three studies conducted in adolescents with CP (age greater than 13 years) that have investigated change in aerobic capacity after exercise interventions compared to a non-exercising control group, using the CPET.[59, 71, 72] These studies all explored the effect of continuous land-based exercise, such as cycling, walking uphill, treadmill ambulation and arm cranking, performed 2 to 4 times per week at a moderate intensity work rate (<80% heart rate reserve, <75% maximum heart rate, or <65% VO_{2peak}). No safety concerns were reported.

In these studies, reported improvements in VO_{2peak} in the exercise group, relative to their baseline measures (i.e. within group differences), varied from 9% to 22%. These results need to be interpreted with caution due to the possible influence of confounders.

In the first study, Slaman et al[71] recruited and randomised 57 participants, of whom 19 (33%) were young people with GMFCS II CP. They reported an improvement in VO_{2peak} of (mean difference; MD) 195 mL/min (95% CI 57 to 333) over and above any change seen in the control group, which was statistically significant at 6 months. A number of factors need to be considered in interpreting this outcome. Firstly, measures of VO_{2peak} collected 3 months following completion of the supervised exercise program component demonstrated no between-group difference and large variability in response to the intervention (MD between-groups 89 mL/min, 95% CI −99 to 277). The difference between-groups were only demonstrated after those in the exercise group participated in a further 3 months of motivational counselling on activity and

participation (not exercise intervention). This second 3-month phase of motivational intervention following exercise intervention did not meet the guidelines for exercise prescription to improve aerobic capacity.[73] Therefore, it is unlikely that the reported improvements in VO_{2peak} in the intervention group can be attributed solely to the supervised 3-month exercise intervention program.

Secondly, in the study by Slaman et al [71]16% of the CPET performed in the study did not meet criteria for a maximal test and were excluded from analysis. The reasons for this included lack of motivation or an inability to achieve maximal test criteria using an arm crank method. These factors highlight the difficulties of undertaking maximal exercise testing in CP populations, and, in particular, reaching a maximal test with the arm ergometer. As the CPET data from those unable to perform maximal exercise testing were excluded from the final analysis, results may be biased towards participants who have fewer physical limitations and therefore are able to undertake the cycle ergometer CPET.

Finally, the attrition rate in the study was 29% in the exercise group and 24% in the control groups. This increases the likelihood of a type II statistical error, and also reduces the protection afforded by randomisation to ensure that the groups were balanced for important confounders.

The two other studies of exercise interventions had small samples, with 20[72] in one study and 13[59] in the other. In the second study by Nsenga et al[72] undertook a cycling study and reported a 22% improvement in VO_{2peak} in 10 participants with CP involved in the exercise training group. The authors did not report between-group analyses comparing the exercise group and the control group, who were not randomised but were

balanced in terms of clinical characteristics. Using online software and data reported in the manuscript, the between-group difference (in favour of the exercise group) was estimated to be MD 7.7 mL/kg/min, 95% CI 2.5 to 12.9; $p < 0.05$; $n = 20$.

In the third study, Unnithan et al[59] undertook an uphill walking exercise intervention over 12 weeks and assessed aerobic capacity improvement using an arm crank ergometer method in 13 randomised participants. They reported a 19% improvement in VO_{2peak} in their exercise training group ($n = 7$). Again, using online software and data reported in the manuscript, the between-group difference (in favour of the exercise group) was estimated to be MD 2.8 mL/kg/min, 95% CI −3.53 to 9.13; $p = 0.35$; $n = 13$. While it is likely the study is underpowered for between group differences, measuring improvements of a lower limb–based training method using an upper limb CPET may have also influenced results.

Another important difference noted between all the studies was that when comparing pre-training (i.e. baseline) measures of VO_{2peak}, Unnithan et al[59] reported much lower VO_{2peak} values when compared to Nsenga (20.8 mL/kg/min versus 43.4 mL/kg/min). Slaman et al[71]did not report VO_{2peak} values adjusted for weight and therefore cannot be compared. This difference between Unnithan et al[59] and Nsenga et al[72] may be explained by the different CPET methods used to assess aerobic capacity (arm crank versus cycle ergometer).[59, 72] In children, treadmill and cycle ergometer protocols can yield a 10% difference in VO_{2peak}.[74] The influence of an arm cranking protocol on VO_{2peak} in children is unclear, but in adults, VO_{2peak} measured with arm cranking has been reported as 29% less than with cycling protocols.[75]

Mobility characteristics of the participants recruited across the studies also differ. The Unnithan et al study had less mobility than those in the Nsenga et al study (GMFCS II and III versus GMFCS I and II CP). Slaman et al included adolescents with GMFCS levels 1 to IV, however 89% were GMFCS I or II CP. Given that aerobic capacity across GMFCS levels is reported as significantly different,[76] it is difficult to compare improvements across studies when there is heterogeneity in gross motor capacity and physical function of participants.

In addition to these studies investigating aerobic exercise interventions in adolescents with CP measured through a CPET, two studies have specifically used HIIT methods. One RCT by Van Wely et al[77] implemented a 4-month program aimed at improving muscle strength and anaerobic fitness in 49 children aged 13 years and under,[77] which is a younger sample than the population of interest for this thesis (adolescents). The HIIT aspect of the supervised training involved three sets of slaloms or running. No mention was made of the intensity of the HIIT or the work: rest ratio. Training took place twice a week for 2 months, followed by once a week for a further 2 months. Aerobic capacity was reported as a secondary outcome and was measured using a CPET. On completion of the training, no between-group differences in VO_{2peak} were reported (0.3mL/kg/min, CI −3.0 to 3.7; n = 33).[77] Of note, only 67% of participants contributed to VO_{2peak} data post intervention period due to lack of motivation or timing constraints, which may have impacted outcomes.

The other study using HIIT as an exercise method and reporting change in aerobic capacity measured during a CPET, was a small case series by Lauglo et al.[78] The exercise program consisted of treadmill walking or running (± physical assistance) and the prescription was individualised to each participant's ability. Intervals consisted of

1.5 to 4 minutes of exercise at 85% maximal heart rate for a maximum of 16 minutes high intensity exercise. HIIT bouts were interspersed with individualised rest periods of moderate intensity exercise. Training took place two to four times per week for 24 sessions. Eight children of 20 enrolled (mean age 14; range 13-16 years) were able to complete a peak CPET treadmill test for data. Participants demonstrated stable VO_{2peak} during two measures collected during a baseline 'run-in' period, and a 10% improvement in VO_{2peak} after training (median interquartile range 37.3 [31.0 to 40.1] mL/kg/min to 41.0 [36.6 to 48.5] mL/kg/min; $p < 0.01$; $n = 8$). A further 10 participants were able to complete a standardised submaximal CPET with oxygen uptake measures at baseline. This study is difficult to replicate because the children had a range of physical functional ability (GMFCS levels from I to IV) and, therefore, the exercises and HIIT training they could perform were disparate and not standardised. It is important to note that six (30%) adolescents did not complete the study. The reasons for this are unclear and enjoyment of participation was not reported. The magnitude of improvement in VO_{2peak} was consistent with improvements reported in other land based HIIT studies in healthy adolescents (MD 2.6 mL/kg/min, 95% CI 1.8 to 3.5; $p < 0.001$).[43, 79, 80]

In summary, while improvement in aerobic capacity through exercise training (as measured by CPET) has been reported in adolescents with CP, evidence for this is weak. The heterogeneity of physical impairment, the lack of standardisation in exercise programs, and difficulty completing gold standard measures of exercise capacity has hindered the confidence in the efficacy of these programs in young people with CP.[37, 76] There is a need for well-designed prospective controlled studies investigating alternative training approaches that are both effective and enjoyable. Of particular

interest is the novel use and potential benefits of an A-HIIT training method in populations that present with impairments that limit the intensity that can be reached during land-based exercise. Robust studies are required on both the design and efficacy of A-HIIT programs generally and the feasibility of adapting them in populations with disability.

2.5 The need for a pilot study

2.5.1 What is a pilot randomised controlled trail?

Pilot studies can be considered a subset of feasibility studies, with a pilot RCT defined as a type of feasibility study that represents a small-scale version of a planned larger trial[32, 81] (Figure 2-2).

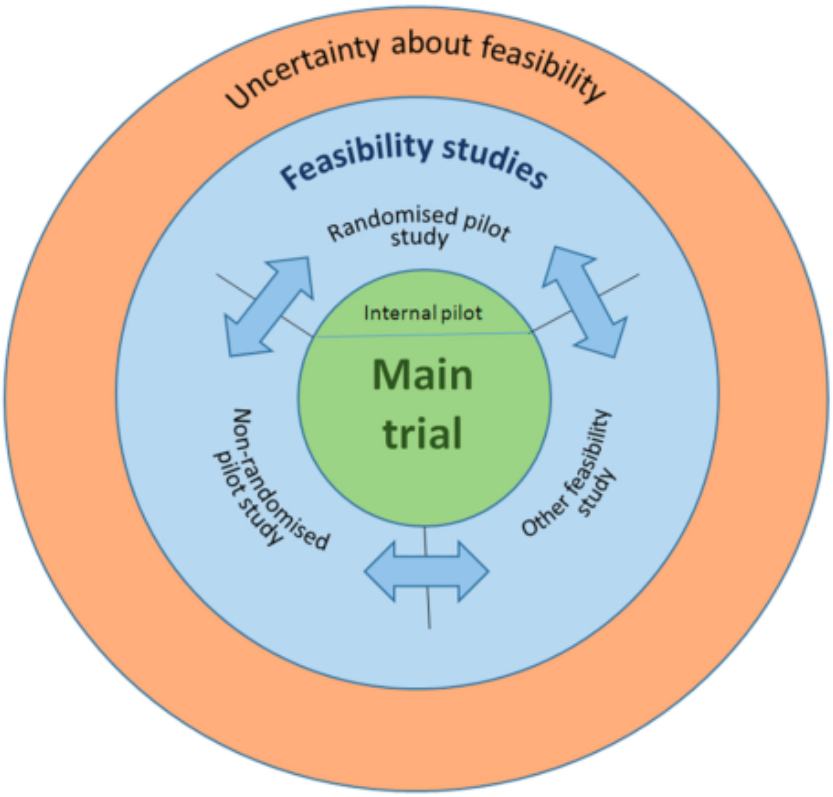


Figure 2-2: Pilot study conceptual framework[32]

The terms pilot study and feasibility study have been traditionally intertwined, and often used to disguise trials that were designed to test effectiveness but were underpowered. Reviews have been conducted on the use of pilot studies reported in medical journals. A 2001 review across seven major medical journals and 90 articles found that pilot studies often had an inappropriate emphasis on hypothesis testing with poorly reported methodology. Only four articles in this review stated the pilot study was conducted in preparation for a definitive study.[82] This review was repeated across the same journals 7 years later, with a large proportion (81%) still focused on hypothesis testing.[83] Kaur et al[84] in 2017 reviewed 191 pilot studies published in a rehabilitation journal over 30 years and noted that only 60% were testing feasibility for a future trial and less than 4% had a registered study protocol. These themes of inadequate reporting and bias towards hypothesis testing and discussion of significance have been repeated in other reviews.[85, 86]

In 2004, Lancaster et al[82] defined the objectives of conducting a pilot study: to test the study protocol, the data collection, the randomisation procedure, the recruitment and consent procedures, the acceptability of the intervention, and the feasibility of using selected outcome measures. There is increasing importance placed on completing feasibility studies prior to undertaking larger RCTs. This, in part, is due to increasing competition for grant funding and demand for less risk in research design before commitment of considerable human and monetary resources.[84] For new and establishing researchers, it also has the potential to improve the quality of the definitive trial research and minimise design flaws.[87] In 2015, the importance of these types of trials was reinforced with the development of a dedicated medical journal: *Pilot and Feasibility Studies*.[88]

In 2016, a conceptual framework for pilot studies and a CONSORT (Consolidated Standards of Reporting Trials) reporting statement were published.[89] The influence of past CONSORT statements on the quality of published manuscripts has been investigated with a positive association with improved trial reporting.[90] Similar reviews to the ones previously described will need to be repeated in future to determine if current limitations in methodology, intent and reporting improve with clear reporting guidelines, and what impact this has on the number of published pilot studies.

2.5.2 Why we do we need a pilot study?

When commencing this program of research, the theoretical rationale for the benefits of trialling A-HIIT in children with CP was evident, but there were a number of unknown feasibility factors relating to this proposed intervention. For example, the following factors were unknown:

- Could high intensity exercise be achieved in an aquatic environment in children with CP?
- Could children with CP complete all the planned outcome assessments?
- Could the exercise intensity be accurately reported in the aquatic environment in these children?
- Was the burden of intervention frequency acceptable to families?

The systematic review in non-athletes could not answer the number of unknown factors that might influence efficacy of A-HIIT in disability populations. The gap in empirical

evidence on the ability to achieve and measure HIIT in an aquatic environment (intervention fidelity) and limited published data on aerobic capacity outcomes in disability populations, raises questions about the feasibility of conducting the proposed research.

2.6 Summary

In healthy young adults and athletes, HIIT programs have demonstrated greater improvements in aerobic capacity than moderate continuous exercise training.[80] Aquatic exercise programs are reported to be more fun and less likely to exacerbate musculoskeletal pain than land-based programs.[18, 22] Together, A-HIIT has the potential to allow higher work rates to be achieved, which in turn may facilitate improvements in aerobic capacity without exacerbating unwanted symptoms and encourage ongoing participation in exercise. A robust understanding of program design and the effectiveness of these types of interventions may enable the successful expansion of these programs into clinical populations, specifically, populations with disability such as adolescents with CP, who are known to have additional limitations in aerobic capacity that can impact on morbidity and mortality.

Specically, although adolescents with GMFCS II CP are ambulant, physical impairments and limitations such as pain and fatigue reduce tolerable options for ongoing commitment to improving and sustaining aerobic capacity. Further, exercise enjoyment and limitation of exercise barriers such as pain and fatigue are essential to encourage ongoing particpation in programs and the prevention of chronic disease.

Therefore, we determined a priori that two studies were required.

1) A systematic review on the reporting and efficacy of A-HIIT programs in non-athletic populations to develop a robust study design and

2) A comprehensive pilot study to investigate the feasibility of a supervised A-HIIT program before answering the definitive trial question: In adolescents with CP at GMFCS level II, does a 10-week A-HIIT program change aerobic capacity (primary outcome), lean muscle mass, pain and social interaction (secondary outcomes), over and above usual care?

Chapter 3:
Systematic review and meta-analysis

3 Systematic review and meta-analysis

This chapter reports on a systematic review and meta-analysis undertaken to explore:

1) The efficacy of high intensity interval training (HIIT) in an aquatic environment (A-HIIT) on aerobic capacity, strength and body composition

2) The design of A-HIIT programs that have been previously described in non-athletic populations

3) Any safety concerns of A-HIIT programs that have been previously reported in non-athletic populations.

This study has been published as:

Depiazzi J, Forbes R, Gibson N, Smith N, Wilson A, Boyd R, Hill K. The effect of aquatic high-intensity interval training on aerobic performance, strength and body composition in a non-athletic population: systematic review and meta-analysis. *Clinical Rehabilitation* 2019; 33: 157–170. https://doi.org/10.1177/0269215518792039

This chapter presents this publication in a thesis format and includes a sensitivity analysis (Section 3.2.5) that was not included in the publication.

3.1 Methods

This systematic review was undertaken and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analysis (PRISMA) guidelines[91] and registered with the international prospective register of systematic reviews

(ID CRD42017079492; https://www.crd.york.ac.uk/PROSPERO). Screening to determine study eligibility, assessment of risk of bias and data extraction were completed independently by two reviewers (JD and RF), with discrepancies resolved through discussion or by a third reviewer (KH).

3.1.1 Search strategy

Six databases were searched including the Cochrane Central Register of Controlled Trials, PubMed, CINAHL, SPORTDiscus, MEDLINE and EMBASE, from their inception until December 2017. The search strategy comprised the following Medical Subject Headings (MeSH) or keywords: (i) aquatic exercise *or* aquatic *or* water exercise *or* hydrotherapy *or* immersion *or* head out aquatic *or* water run *or* swim, (ii) *and* high intensity interval training *or* interval training *or* intermittent exercise *or* plyometric. An example of the search terms for PubMed can be found in Appendix 1. Additionally, the reference lists of systematic reviews and included papers were searched by hand for further possible studies. Thesis titles were searched through ProQuest. Duplicates were removed using Endnote (Endnote X7, Thomson Reuters, USA). Authors were contacted by email for additional data as required.

3.1.2 Eligibility criteria

Titles, abstracts and, where necessary, full text were screened to determine eligibility. Studies were included for analysis if they met the following criteria: (i) were published as full papers; (ii) used either a randomised controlled trial (RCT) design or other study design in which outcomes were compared between groups of participants who were recruited at the same time, where one group received A-HIIT and the other did not undergo exercise training (control); (iii) recruited participants aged ≥6 years, with or

without diagnosis of a health condition; (iv) described the intervention as an exercise program that was conducted in water that met our a priori definition of A-HIIT. Specifically, for this review, A-HIIT was defined as exercise in water consisting of at least three cycles of work:rest. In order for the training to be considered 'high intensity', the work rate prescribed during the work interval needed to elicit: (i) a heart rate greater than 75% of the predicted maximum heart rate maximum *or* (ii) a rate of oxygen uptake (VO_2) greater than 75% of VO_2 at peak or maximal capacity (VO_{2peak} and VO_{2max}), *or* (iii) a rate of perceived exertion (RPE) of 15 or more on the Borg exertional scale,[92] *or* (iv) reports of the intensity being 'hard' or requiring 'all out' exertion. Separate parameters needed to be described for a 'rest' period. Studies were excluded if: (i) the participants were described as athletes, (ii) A-HIIT was conducted for <2 weeks and/or comprised <6 sessions of exercise training, or (iii) the A-HIIT was not supervised or monitored for exercise intensity.

3.1.3 Bias and quality of reporting assessment

Bias was assessed using the Cochrane risk of bias assessment tool.[93] Each study was graded for the following domains: sequence generation; allocation concealment; blinding (participants, personnel, outcome assessors); incomplete outcome data; selective reporting; and other bias, such as inequity in gender distribution. Pre-determined criteria were established, and for each study a rating of 'low', 'high' or 'unclear' risk was assigned for each domain. Assessment of publication bias was planned through visual interpretation of funnel plot asymmetry if greater than 10 studies were eligible for inclusion in the meta-analysis.[94]

Given its familiarity among physiotherapists, studies were also graded using the Physiotherapy Evidence Database (PEDro) Scale. This scale reports on the quality of methodological reporting and is widely used in exercise intervention reporting analysis.[95] Items pertaining to internal validity and quality of reporting were scored for 10 criteria by assigning 'yes' a score of 1 and 'no' a score of 0. Where data were unavailable or unclear, the item was assigned a score of '0'. For this review, studies scoring ≤3 were considered to be of low methodological quality.

Quality of reporting of the exercise intervention was determined using the Consensus on Exercise Reporting Template (CERT).[96] The CERT is a 16-item checklist designed to improve the reporting of exercise programs. It comprises seven categories: materials, provider, delivery, location, dosage, tailoring and compliance.[96] Scoring items as 1 if they did report on these categories, versus 0 if they did not report on these categories, allowed for a possible total score of 19. Where there was ambiguity in reporting, the item was scored a 0 (Appendix 2). Where two or more papers reported on the same study intervention, the CERT score reflects the combined reporting of the exercise intervention across all studies.

3.1.4 Data extraction

Data were extracted on study design, participant characteristics, content and dosage of the exercise intervention. Baseline and post-intervention outcomes were extracted by two independent reviewers (JD and RF) and checked for accuracy by a third and/or fourth reviewer (KH or NG). Where possible, intention to treat data were extracted for post-intervention measurements.

3.1.5 Outcome measurement selection

Aerobic performance was defined as any measure of exercise capacity or tolerance that had been collected during a standardised exercise test. Peak lower limb muscle strength or force were defined as measures collected during a maximal voluntary isometric or isokinetic contraction by a dynamometer (seated or handheld) or load cell. Where multiple strength measures were taken, only a single lower limb muscle per intervention group was used, with the quadriceps or knee extension as a first preference. Body composition was defined as any measure of lean muscle mass, lean body mass or fat mass obtained by skin fold measures, BodPod® or dual energy X-ray absorptiometry. Data extracted on safety related to adverse events were grouped as serious (death) and minor (requiring health professional assessment, advice and/or treatment).

3.1.6 Synthesis of results

Meta-analyses were performed using Review Manager (RevMan) (Version 5.3. Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2014). As data for outcomes related to aerobic performance, strength and body composition were reported using different units of measurement, the size of the effect was estimated as the standardised mean difference (SMD) with corresponding 95% confidence intervals (CI). To calculate the SMD, post-intervention means and standard deviations (SD) were used. Where studies reported change scores rather than post-intervention measures, post-intervention means were estimated using baseline measures. Where only pre-intervention SD were available, pooled SD of baseline measures were utilised for post-intervention measures. For these meta-analyses, an SMD of 0.2 to 0.49 was considered a small effect size, 0.5 to 0.79 was considered a medium effect size, and greater than or

equal to 0.8 was considered to be a large effect.[97] Heterogeneity was assessed using the I^2 statistic with values >50% deemed to be high. For all analyses, an inverse-variance weighted random effects model was used. Data pertaining to safety were reported as count data (e.g. adverse events) and were assessed using risk difference.

Sensitivity analyses were planned that involved the exclusion of studies meeting the following criteria: (i) non RCT (risk of selection bias), (ii) loss to follow-up rate ≥15% (risk of attrition bias), (iii) PEDro scores ≤3 (low score of methodological quality), (iv) training frequency ≤2 per week, and (v) duration of training ≤8 weeks. Subgroup analyses were planned with studies grouped according to the average age of participants (≤18 versus >18 years).

3.2 Results

3.2.1 Included studies

A total of 2559 records were identified. Thirteen papers met inclusion criteria. Data for quantitative analysis were extracted from eight different studies reported over these 13 papers[98-110] (Figure 3-1).

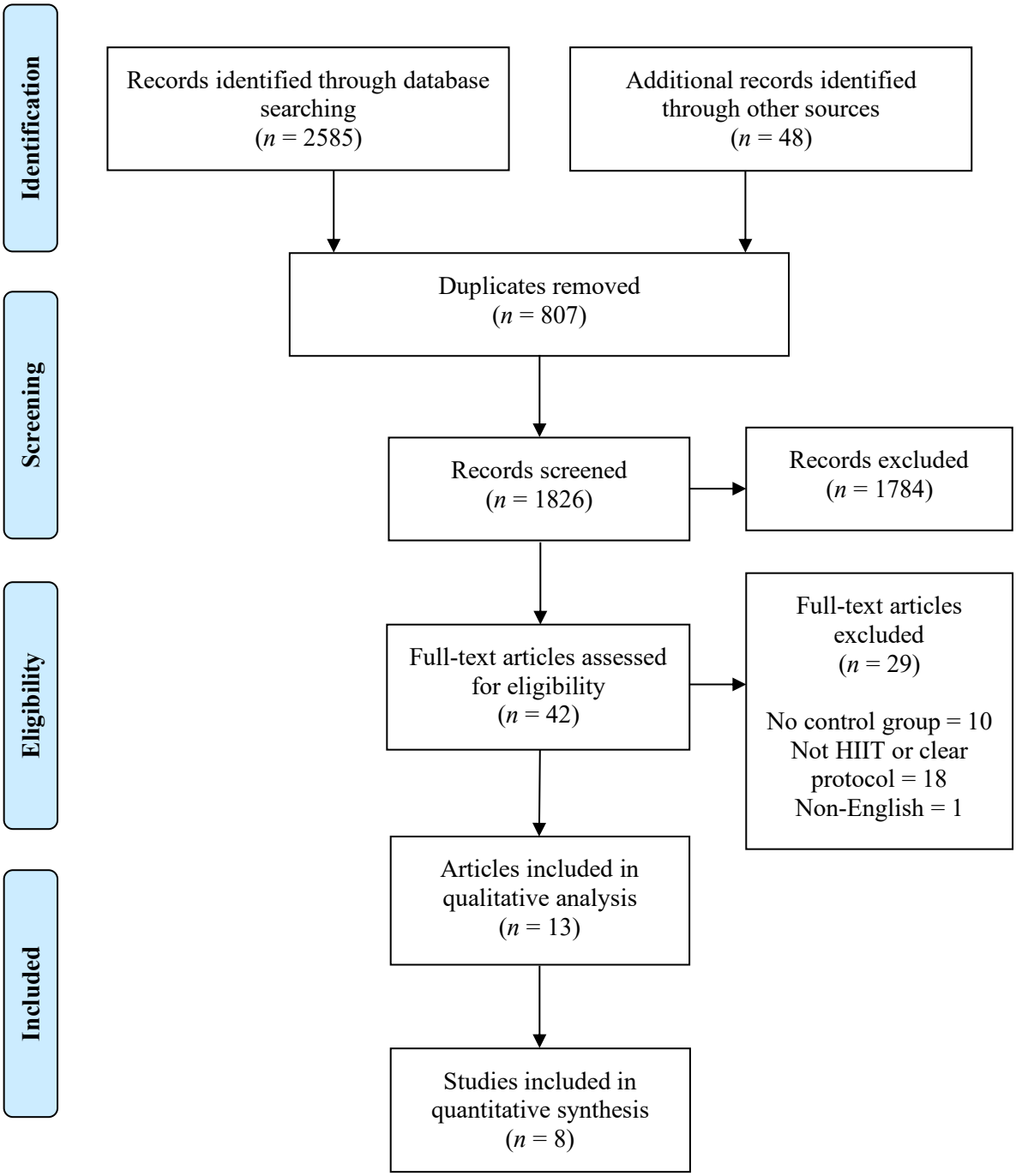


Figure 3-1: PRISMA flow diagram

3.2.2 Participants

Data were extracted on 377 participants (A-HIIT n = 212, control n = 165) with the mean participant age ranging from 21.7 to 69.0 years for A-HIIT and 21.7 to 69.8 years for controls (Table 3-1). Participants were predominantly female (89%). Using self-reported activity levels, more than 50% of participants described themselves as sedentary. Two studies included participants with medical conditions: mild hypertension[104] or knee osteoarthritis.[110] No participant reported undertaking routine aquatic exercise or regular HIIT prior to participating in the study.

Table 3-1: Study design and participant characteristics

Author and year	Study design	Intervention groups	N	Male: Female	Rx Age mean (SD+)	C Age mean (SD+)	Population type	Normal activity level
Bento 2015	RCT	A-HIIT, C and Rx B	Total = 67 A-HIIT = 25 C = 19 Rx B = 23	0:67	65.5	66.2	Healthy	A/S
Broman 2006	RCT	A-HIIT, C	Total = 29 A-HIIT = 18 C = 11	0:29	69.0 (4.0)	69.8 (3.5)	Healthy	A
Mohr 2014*, Connolly 2016, Mohr 2015, Nordsborg 2015	RCT	A-HIIT, C and Rx B	Total = 62 A-HIIT = 21 C = 20 Rx B = 21	0:62	44.0 (5.0)	45.0 (4.0)	Clinical	S
Moreira 2013*, Moreira 2014	RCT	A-HIIT, C	Total = 108 A-HIIT = 64 C = 44	0:108	58.6 (6.7)	59.3 (6.1)	Healthy	S
Rebold 2013	RCT	A-HIIT, C	Total = 25 A-HIIT = 13 C = 12	17:8	21.7 (1.5)	21.7 (1.5)	Healthy	A
Waller 2017*, Munukka 2016	RCT	A-HIIT, C	Total = 87 A-HIIT = 43 C = 44	0:87	64.0 (2.0)	64.0 (2.0)	Clinical	A
Hamer 1990	RCT	A-HIIT, C	Total = 20 A-HIIT = 12 C = 8	20:0	22.7 (3.1)	25.1 (4.8)	Healthy	S
Michaud 1995	Exp C	A-HIIT, C	Total = 23 A-HIIT = 16 C = 7	2:14	32.0 (6.8)	32.0 (6.8)	Healthy	S

A = active or not stated; A-HIIT = aquatic high intensity interval training; C = control; Rx B = alternative intervention; S = sedentary; + = where data available; * Article of primary outcome interest – papers reporting on the same intervention are grouped and intervention referred to by the author indicated by *

3.2.3 Quality assessment

3.2.3.1 Risk of bias – Cochrane assessment

Of the 13 papers included in this review, one was rated as being at high risk of selection bias (Figure 3-2).[103] All papers were rated at high risk of performance bias and one study was rated as being at high risk of detection bias.[110] Four studies were rated at high risk of attrition bias with a loss to follow-up of ≥15%.[98, 99, 101, 103] A further study did not report on withdrawals.[109] A total of 32 participants withdrew from the included studies (8.5%), 22 from the A-HIIT groups. Reasons for withdrawal included unrelated death, poor tolerability, unrelated medical conditions, family and work commitments, or loss of interest. All papers were rated as being unclear in terms of reporting bias. The other potential source of bias related to gender inequity with five of the studies recruiting only female participants.[98, 99, 104, 106, 110] The number of studies included in the meta-analysis was insufficient to determine publication bias using funnel plots.[94]

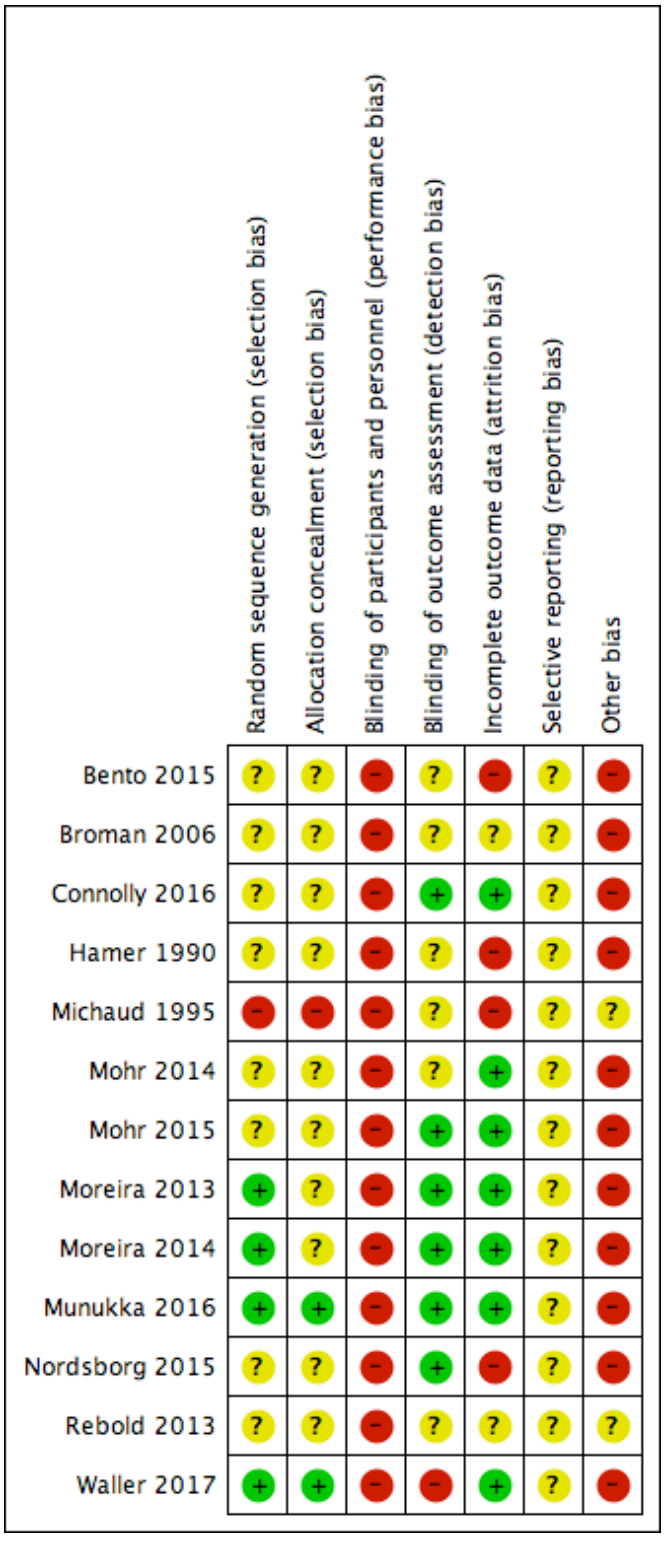


Figure 3-2: Risk of bias assessment demonstrating low risk (green), high risk (red) and unclear risk (yellow)

3.2.3.2 Quality of methodological reporting – PEDro Scale

Seven of the eight studies were RCTs (Table 3-2). The mean PEDro score was 4.9 ± 1.5 (mean ± SD) for all papers, with two rating as 'low' quality with scores ≤3.[103, 109]

Study	1	2	3	4	5	6	7	8	9	10	11	Total
Bento 2015	1	1	0	1	0	0	0	0	0	1	1	**4/10**
Broman 2006	1	1	0	1	0	0	0	0	0	1	1	**4/10**
Connolly 2016[a]	1	1	0	1	0	0	0	1	0	1	1	**5/10**
Hamer 1990	1	1	0	1	0	0	0	0	0	1	1	**4/10**
Michaud 1995[x]	1	0	0	1	0	0	0	0	0	1	1	**3/10**
Mohr 2014[a]	1	1	0	1	0	0	0	1	0	1	1	**5/10**
Mohr 2015[a]	1	1	0	1	0	0	0	1	0	1	1	**5/10**
Moreira 2013[c]	1	1	1	1	0	0	1	1	0	1	1	**7/10**
Moreira 2014[c]	1	1	1	1	0	0	1	0	0	1	1	**6/10**
Munukka 2016[b]	1	1	1	1	0	0	1	1	1	1	1	**8/10**
Nordsborg 2015[a]	1	1	0	1	0	0	0	0	0	1	1	**4/10**
Rebold 2013	1	1	0	0	0	0	0	0	0	1	1	**3/10**
Waller 2017[b]	1	1	0	1	0	0	0	1	1	1	1	**6/10**

Scale of item score: 0 = absent/unclear, 1 = present. The PEDro Scale criteria are: (1) specification of eligibility criteria, (2) random allocation, (3) concealed allocation, (4) prognostic similarity at baseline, (5) subject blinding, (6) therapist blinding, (7) assessor blinding, (8) greater than 85% follow-up of at least one key outcome, (9) intention to treat analysis, (10) between-group statistical comparison for at least one key outcome, (11) point estimates and measures of variability provided for at least one key outcome. [a, b, c] = papers describing same interventional study; [x] = non RCT

Table 3-2: PEDro summary

3.2.3.3 Quality of reporting the intervention – CERT

Analysis of the quality of reporting of the exercise intervention using the CERT is detailed in Table 3-3. The CERT score (mean ± SD) was 15.1 ± 2.1 with all articles scoring high for description of exercise, surroundings, motivation, and exercise intensity monitoring and progression. Of the six studies that reported attendance to the exercise classes, the mean attendance was >85%. Of the four studies that reported on adherence to the prescribed exercise intensity, the mean adherence to intensity was >90% (Table 3-4).

Table 3-3: Consensus on Exercise Reporting Template (CERT) assessment

Article	Item																		
	1	**2**	**3**	**4**	**5**	**6**	**7a**	**7b**	**8**	**9**	**10**	**11**	**12**	**13**	**14a**	**14b**	**15**	**16a**	**16b**	**Total score /19**
Bento 2015	0	0	1	0	1	1	1	1	1	0	0	0	1	1	1	1	1	0	0	**11**
Broman 2006	1	1	0	0	1	1	1	1	1	0	0	1	1	1	1	1	1	1	1	**15**
Hamer 1990	1	1	0	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	1	**18**
Michaud 1995	1	0	0	0	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	**15**
Mohr 2014	0	1	1	1	1	1	1	1	1	0	0	1	0	1	1	1	1	1	1	**15**
Moreira 2013	1	0	0	1	1	1	1	1	1	0	1	1	1	1	1	1	1	1	1	**16**
Rebold 2013	1	0	0	0	1	1	1	1	1	1	1	0	1	1	1	1	1	1	0	**14**
Waller 2017	1	0	1	1	1	1	1	1	1	1	0	1	1	1	1	1	1	1	1	**17**

1 = yes; 0 = no

Each item describes: (1) exercise equipment, (2) personnel expertise, (3) individual or group, (4) supervised or unsupervised, (5) adherence to exercise, (6) motivation strategies, (7a and b) when and how exercise progressed, (8) exercise replication, (9) home program, (10) non-exercise components, (11) type and adverse events, (12) setting, (13) exercise intervention, (14a and b) exercise generic or individually tailored, (15) starting level, (16a and b) adherence measurement and delivered per protocol

Table 3-4: Summary of characteristics of the intervention

Author	Super-vised	Intensity monitoring	Define intensity	Training sessions (no.)	Total class time (min)	HIIT sessions (no.)	Description of HIIT (no. sets of work:rest in seconds)	Type of water training	Other training group	Water temp.	Mean 85% attendance	Mean 90% adherence to intensity
Bento 2013	NR	BORG RPE	RPE 14–16 in last 4 weeks of training	3x/week x 12 weeks = 36	60	12	3 sets of 40s (work): 20s (active recovery)	fast reps of resisted movement	land resistance	28–30°C	NR	NR
Broman 2006	NR	BORG CR 10; Polar® HR	>75% MHR	2x/week x 8 weeks = 16	44	16	3 sets of 10 min cycles of 15–40s (work): 10–20s (rest)	DWR with buoyancy jacket	N	27°C	Y	Y
Mohr* 2014	Y	work rate; HR	'all out' freestyle swimming	3x/week x 15 weeks = 45	15–25	45	6–10 sets of 30s (work): 120s (passive recovery)	freestyle swim	low intensity continuous swim AND land football	NR	Y	Y
Moreira* 2013	Y	BORG CR 10; Polar® HR	week 15–24 BORG >8 (>80% MHR)	3x/week x 24 weeks = 72	50–60	30	4–5 sets of 10–15s (work): 90–100s (rest)	aerobic activity	N	30–31°C	Y	NR
Rebold 2013	Y	BORG RPE; Polar® HR; work rate	80–95% max work rate	2x/week x 8 weeks = 16	17	16	8 sets of 20s (work): 10s (rest)	treadmill running with weighted vest	N	26–28°C	NR	NR
Waller* 2017	Y	BORG RPE; Polar® HR; biochemistry; work rate	'all out' using full ROM; BORG 15–18	3x/week x 16 weeks = 48	60	42	5 stations (x2), 30–45s (work): 30–45s + 45s (rest/ transition)	progressive resistance training	N	30–32°C	Y	Y

Author	Super-vised	Intensity monitoring	Define intensity	Training sessions (no.)	Total class time (min)	HIIT sessions (no.)	Description of HIIT (no. sets of work:rest in seconds)	Type of water training	Other training group	Water temp.	Mean 85% attendance	Mean 90% adherence to intensity
Hamer 1990	Y	carotid palpation +/– HR telemetry	week 6–8 HR >80% @VO_{2max}	3x/week x 8 weeks = 24	25–44 + cool-down	9	3–5 sets of 30–600s (work): 30–150s (rest)	running in 1 metre of water (with shoes)	N	28°C	Y	Y
Michaud 1995	NR	RPE 1–5; Polar® HR; cadence	RPE 4/5; ≥75% @ VO_{2max}	3x/week x 8 weeks = 24	40–70	21–24	1–10 sets of 30–420s (work): 30s (rest)	DWR with buoyancy jacket	N	27–29°C	Y	NR

BORG = RPE 6–20; BORG CR 10 = RPE 0–10; class time = number of minutes spent exercising; DWR = deep water running; HIIT sessions = number of training sessions that met HIIT criteria; HR = heart rate; intensity monitoring = type of monitoring used; MHR = maximum heart rate; mean 85% attendance = group mean attendance to training sessions in those included for analysis; mean 90% adherence = group participant mean of at least 90% adherence to study intensity parameter; N = no; NR = not reported or unclear; RPE = rate of perceived exertion; ROM = range of motion; supervised = A-HIIT sessions supervised; VO_{2max} = maximum rate of oxygen uptake; Y = yes; * = multiple papers describing the study

3.2.4 Characteristics of the A-HIIT program

Characteristics of the intervention, exercise intensity and dosage are summarised in Table 3-4. Pre-intervention measures for outcomes included in this meta-analysis were balanced between groups. A variety of exercises were described including freestyle swimming, deep water vest running, underwater treadmill running, and fast-paced resisted movement. The majority of studies monitored exercise intensity using heart rate monitors such as Polar® and/or a rate of perceived exertion such as the Borg Scale[111]. The total number of exercise sessions ranged from 16 to 72, with some studies employing a participant familiarisation and/or progressive exercise design before reaching meta-analysis criteria for high intensity. Work:rest ratios were heterogeneous, with work periods ranging from 10 to 600 seconds and rest periods from 10 to 150 seconds. Four of the studies had work periods that were greater than rest periods by the end of the intervention period.[98, 99, 101, 109]

3.2.5 Effect of A-HIIT

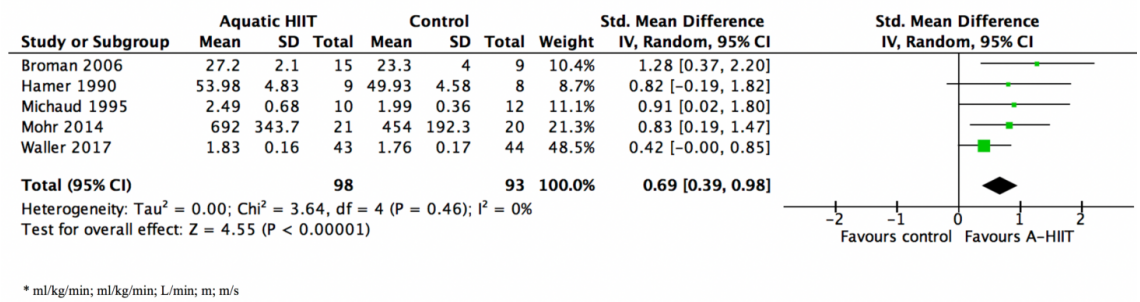
3.2.5.1 Aerobic performance

Five studies reported on aerobic performance. Of these, three expressed data as VO_{2peak} or VO_{2max}[99, 101, 103], one expressed data as a distance (Yo-Yo IE1 test[104]) and one expressed data as a speed (UKK 2 km walking test[110]). The effect of A-HIIT, over and above a control group, on aerobic performance was moderate, with an SMD of 0.69 (95% CI 0.39 to 0.98). This analysis demonstrated low heterogeneity ($I^2 = 0\%$) (Figure 3-3a).

Sensitivity analyses that involved the exclusion of studies based on selection bias (Figure 3-3b), attrition bias (Figure 3-3c) and low methodological quality (Figure 3-3d)

had little effect on the result. Similarly, removal of studies with training frequency of ≤2 per week[99] (Figure 3-3e) or ≤8 weeks of training duration (Figure 3-3f)[99, 101, 103] did not change the effect of A-HIIT on aerobic performance.

3-3a: Effect of A-HIIT versus control on aerobic performance



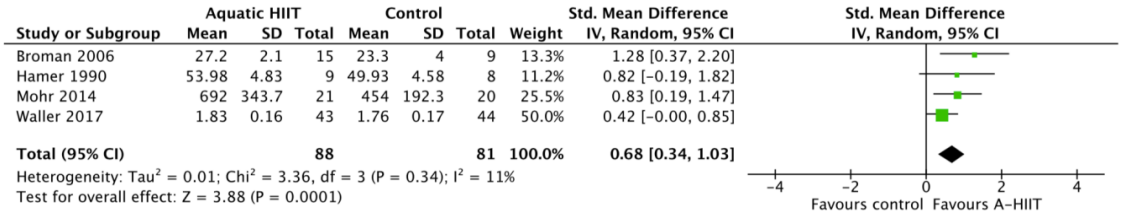
Study or Subgroup	Aquatic HIIT Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Broman 2006	27.2	2.1	15	23.3	4	9	10.4%	1.28 [0.37, 2.20]
Hamer 1990	53.98	4.83	9	49.93	4.58	8	8.7%	0.82 [-0.19, 1.82]
Michaud 1995	2.49	0.68	10	1.99	0.36	12	11.1%	0.91 [0.02, 1.80]
Mohr 2014	692	343.7	21	454	192.3	20	21.3%	0.83 [0.19, 1.47]
Waller 2017	1.83	0.16	43	1.76	0.17	44	48.5%	0.42 [-0.00, 0.85]
Total (95% CI)			**98**			**93**	**100.0%**	**0.69 [0.39, 0.98]**

Heterogeneity: Tau² = 0.00; Chi² = 3.64, df = 4 (P = 0.46); I² = 0%
Test for overall effect: Z = 4.55 (P < 0.00001)

Std. Mean Difference IV, Random, 95% CI
−2 −1 0 1 2
Favours control Favours A-HIIT

* ml/kg/min; ml/kg/min; L/min; m; m/s

3-3b: Sensitivity analysis for effect of A-HIIT versus control with removal of studies with high selection bias

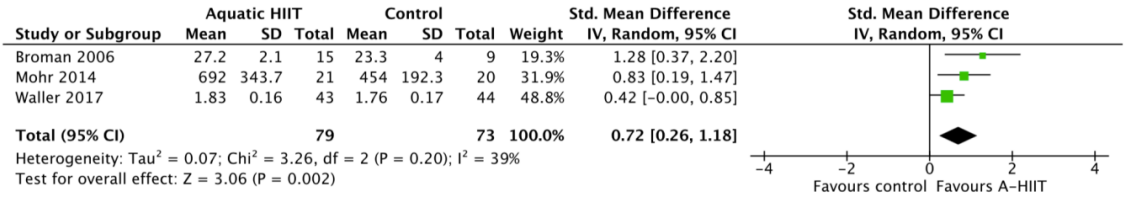


Study or Subgroup	Aquatic HIIT Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Broman 2006	27.2	2.1	15	23.3	4	9	13.3%	1.28 [0.37, 2.20]
Hamer 1990	53.98	4.83	9	49.93	4.58	8	11.2%	0.82 [-0.19, 1.82]
Mohr 2014	692	343.7	21	454	192.3	20	25.5%	0.83 [0.19, 1.47]
Waller 2017	1.83	0.16	43	1.76	0.17	44	50.0%	0.42 [-0.00, 0.85]
Total (95% CI)			**88**			**81**	**100.0%**	**0.68 [0.34, 1.03]**

Heterogeneity: Tau² = 0.01; Chi² = 3.36, df = 3 (P = 0.34); I² = 11%
Test for overall effect: Z = 3.88 (P = 0.0001)

Std. Mean Difference IV, Random, 95% CI
−4 −2 0 2 4
Favours control Favours A-HIIT

3-3c: Sensitivity analysis for effect of A-HIIT versus control with removal of studies with high attrition bias

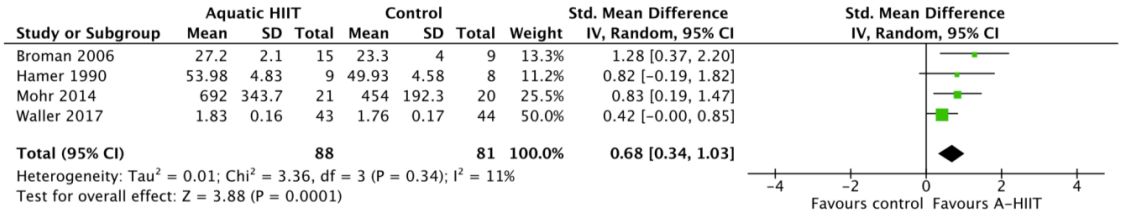


Study or Subgroup	Aquatic HIIT Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Broman 2006	27.2	2.1	15	23.3	4	9	19.3%	1.28 [0.37, 2.20]
Mohr 2014	692	343.7	21	454	192.3	20	31.9%	0.83 [0.19, 1.47]
Waller 2017	1.83	0.16	43	1.76	0.17	44	48.8%	0.42 [-0.00, 0.85]
Total (95% CI)			**79**			**73**	**100.0%**	**0.72 [0.26, 1.18]**

Heterogeneity: Tau² = 0.07; Chi² = 3.26, df = 2 (P = 0.20); I² = 39%
Test for overall effect: Z = 3.06 (P = 0.002)

Std. Mean Difference IV, Random, 95% CI
−4 −2 0 2 4
Favours control Favours A-HIIT

3-3d: Sensitivity analysis for effect of A-HIIT versus control with removal of studies with low methodological quality

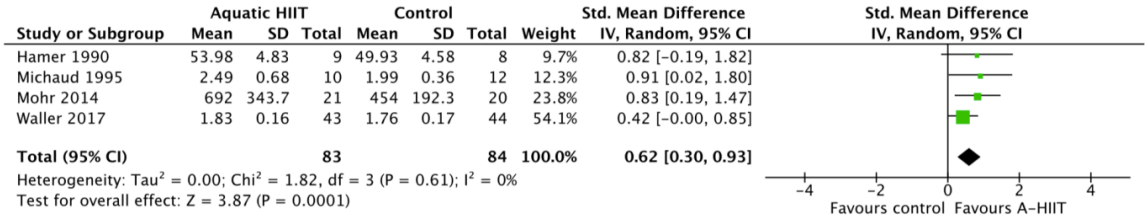


Study or Subgroup	Aquatic HIIT Mean	SD	Total	Control Mean	SD	Total	Weight	Std. Mean Difference IV, Random, 95% CI
Broman 2006	27.2	2.1	15	23.3	4	9	13.3%	1.28 [0.37, 2.20]
Hamer 1990	53.98	4.83	9	49.93	4.58	8	11.2%	0.82 [-0.19, 1.82]
Mohr 2014	692	343.7	21	454	192.3	20	25.5%	0.83 [0.19, 1.47]
Waller 2017	1.83	0.16	43	1.76	0.17	44	50.0%	0.42 [-0.00, 0.85]
Total (95% CI)			**88**			**81**	**100.0%**	**0.68 [0.34, 1.03]**

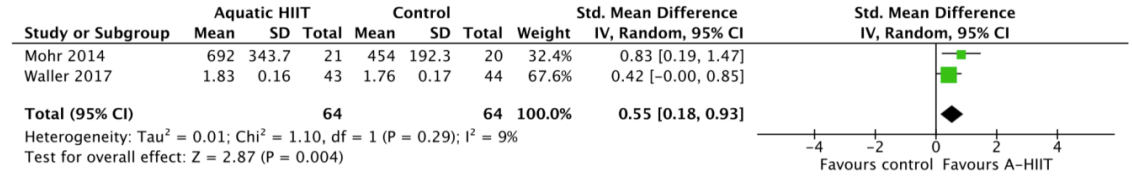
Heterogeneity: Tau² = 0.01; Chi² = 3.36, df = 3 (P = 0.34); I² = 11%
Test for overall effect: Z = 3.88 (P = 0.0001)

Std. Mean Difference IV, Random, 95% CI
−4 −2 0 2 4
Favours control Favours A-HIIT

3-3e: Sensitivity analysis for effect of A-HIIT versus control with removal of studies with training frequency of 2 or less per week



3-3f: Sensitivity analysis for effect of A-HIIT versus control with removal of studies conducted for 8 weeks or less



A-HIIT = aquatic high intensity interval training; CI = confidence interval; HIIT = high intensity interval training; L/min = litres per minute; mL/kg/min = millilitres per kilogram per minute; m = metres; m/s = metres per second; SD = standard deviation; Std = standardised; * = measurement unit in order of author listing

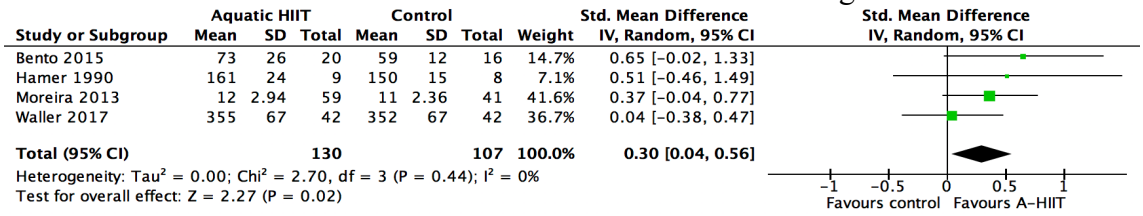
Figure 3-3: Effect of A-HIIT on aerobic performance, with sensitivity analyses

3.2.5.2 Lower limb muscle strength

Four studies reported on lower limb (knee extensor) strength, which was expressed as measures related to isometric contractions (newton or kilogram) or isokinetic contractions (maximum torque in newton metres). The effect of A-HIIT, over and above a control group, on lower limb strength was small, with an SMD of 0.30 (95% CI 0.04 to 0.56). This analysis demonstrated low heterogeneity ($I^2 = 0\%$) (Figure 3-4a).

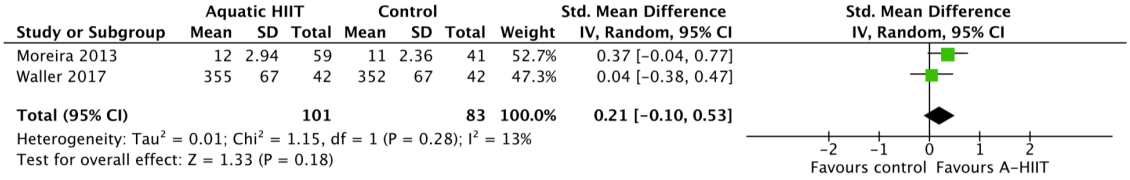
Sensitivity analysis that involved the exclusion of studies based on high attrition rates[98, 101] (Figure 3-4b) and studies that provided ≤8 weeks of training[101] resulted in the effect of A-HIIT on lower limb strength no longer meeting the threshold for statistical significance (Figure 3-4c). As all studies that contributed data to this outcome were RCTs, had a training frequency of >2 per week and had PEDro scores ≤3, additional sensitivity analyses were not possible.

3-4a: Effect of A-HIIT versus control on lower limb muscle strength

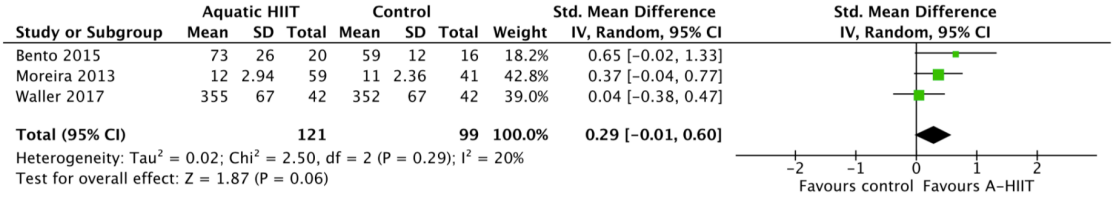


Strength measures reported as N.m or N; SD for Waller et al are pooled

3-4b: Sensitivity analysis for effect of A-HIIT versus control with removal of studies with high attrition bias



3-4c: Sensitivity analysis for effect of A-HIIT versus control with removal of studies conducted for 8 weeks or less



A-HIIT = aquatic high intensity interval training; CI = confidence interval; HIIT = high intensity interval training; SD = standard deviation; Std = standardised

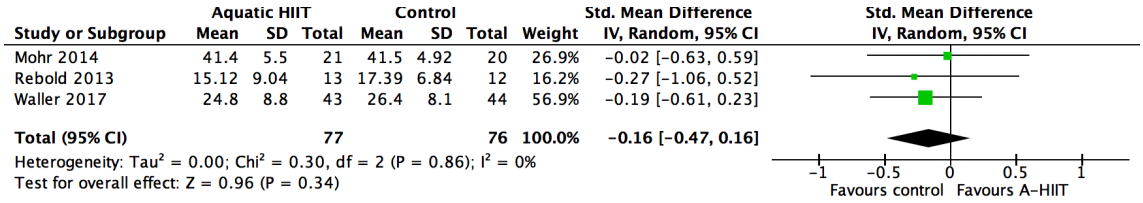
Figure 3-4: Effect of A-HIIT on lower limb muscle strength, with sensitivity analyses

3.2.5.3 Body composition

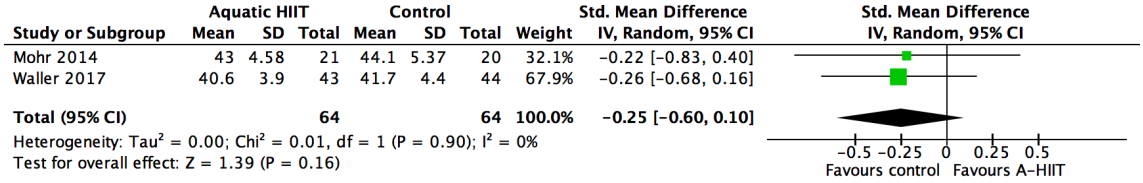
Three studies reported on fat mass (Figure 3-5a) and two studies reported on lean body mass (Figure 3-5b). Fat mass was reported as a body fat percentage[104, 109] or total fat mass in kilograms[110]. Lean body mass was reported as a body percentage[104] or in kilograms[110]. There was no effect of A-HIIT, over and above a control group, on body fat (SMD −0.16, 95% CI −0.47 to 0.16; $I^2 = 0\%$) or lean mass (SMD of −0.25, 95% CI −0.60 to 0.10; $I^2 = 0\%$).

Sensitivity analyses that involved the exclusion of studies based on low methodological quality (Figure 3-5c) or training frequency of ≤2 per week (Figure 3-5d) had little effect on the body fat mass results.[109] As all studies that contributed data to this outcome were RCTs, had attrition rates <15% or provided training for >8 weeks, additional sensitivity analyses were not possible.

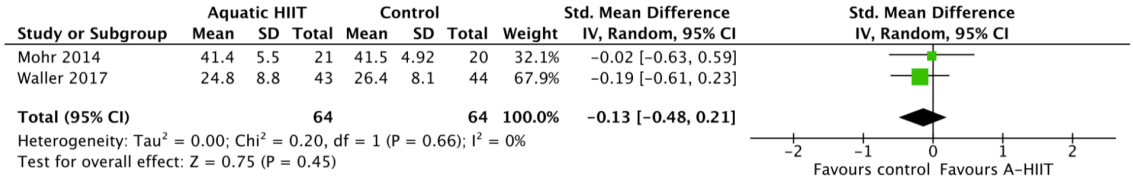
3-5a: Effect of A-HIIT versus control on body fat mass



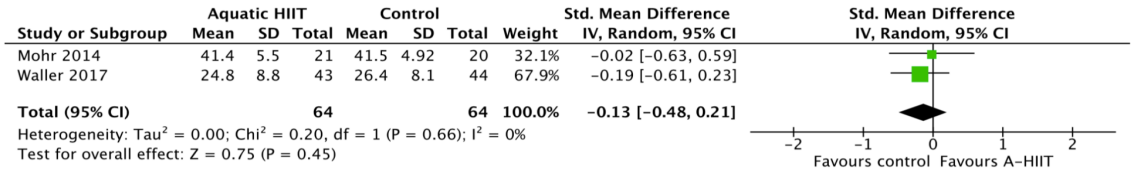
3-5b: Effect of A-HIIT versus control on lean body mass



3-5c: Sensitivity analysis for effect of A-HIIT versus control on body fat mass with removal of studies with low methodological quality



3-5d: Sensitivity analysis for effect of A-HIIT versus control on body fat mass with removal of studies with training frequency of two or less per week



A-HIIT = aquatic high intensity interval training; CI = confidence interval; SD = standard deviation; Std = standardised

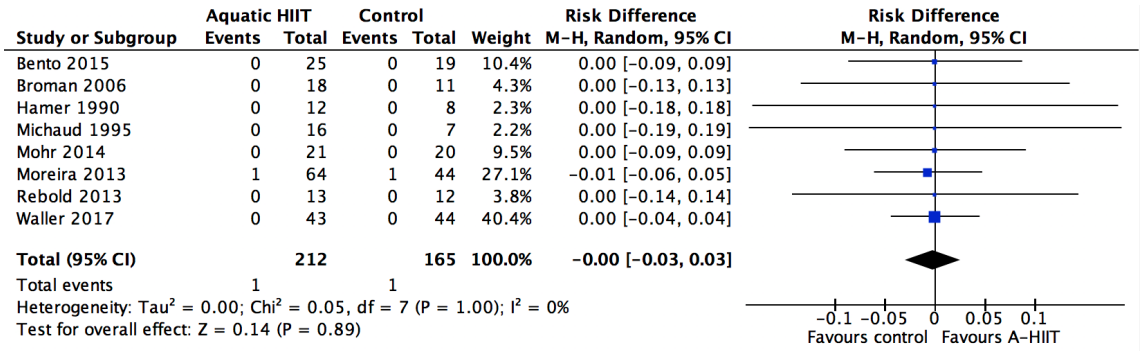
Figure 3-5: Effect of A-HIIT on body composition, with sensitivity analyses for body fat mass

3.2.5.4 Safety

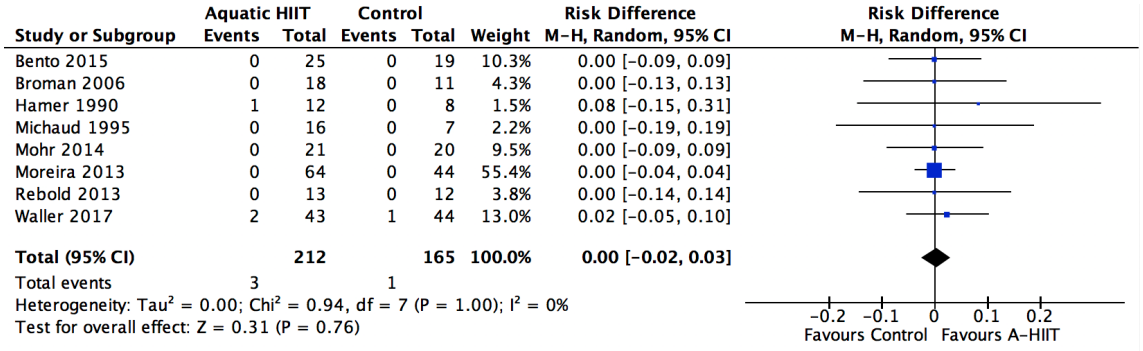
Moreira et al[106] reported two deaths during the intervention (one in the intervention group and one in the control group); neither was related to the study (vehicle accident, post-surgical intervention). The risk difference of a serious adverse event was –0.00 (95% CI –0.03 to 0.03) (Figure 3-5Figure 3-6).

Two studies reported minor adverse events (three events in the intervention group, one in the control group). Adverse events were reported as pain, dyspnoea, intolerability[110] and a 'flare' of a previous injury.[101] The risk difference of a minor adverse event was 0.00 (95% CI −0.02 to 0.03) (Figure 3-6).

13-6a: Effect of A-HIIT versus control for serious adverse events



3-6b: Effect of A-HIIT versus control for minor adverse events



A-HIIT = aquatic high intensity interval training; CI = confidence interval.

Figure 3-6: Safety of A-HIIT

3.2.5.5 Subgroup analysis

As no studies recruited participants with a mean age ≤18 years, the planned subgroup analysis based on mean age of participants could not be done.

3.3 Discussion

This systematic review with meta-analyses investigated the effects of HIIT in an aquatic environment in a non-athletic population. The main findings of this review were that A-HIIT resulted in a moderate improvement in aerobic performance and a small improvement in lower limb strength but had no effect on body composition. It was not associated with greater risk than non-exercising control groups.

Consistent with most exercise intervention studies, blinding of participants and personnel is difficult, if not impossible, and the risk of performance bias was high across all studies included in this meta-analysis. Nevertheless, the risk of selection, detection and attrition bias was generally low. Few studies had a registered protocol, which increased the risk of reporting bias. The other risk of bias related to the disproportionate number of female participants recruited to the included studies. While acknowledging this limitation and its possible impact for clinical interpretation, other studies have demonstrated no gender difference in relation to outcomes such as aerobic capacity following HIIT.[112, 113] Although quality of reporting the methodology was moderate, the description of exercise interventions scored highly. Our inclusion criteria meant that those studies without clear exercise intensity description or monitoring were not included in this meta-analysis and this accounts for the high CERT scores for included studies. Supervised and monitored interventions have been reported to elicit greater improvements in some measures than unsupervised training in older adults.[114] In

the studies included in this meta-analysis, exercise intensity was closely monitored and sessions were supervised, factors which may have contributed to better outcomes.

Data from the meta-analysis indicate that A-HIIT improved aerobic performance. This result was unaffected in the sensitivity analyses. This improvement in aerobic performance occurred after a relatively brief training duration, which is consistent with existing literature on HIIT.[115, 116] The effect on aerobic performance was similar to that reported in a recent meta-analysis of land HIIT in which training was undertaken for <12 weeks (SMD 0.74, 95% CI 0.36 to 1.12), although the author of that review recommends a training frequency of three times per week for more than 12 weeks for a larger effect (SMD 1.20, 95% CI 0.57 to 1.83).[115] In the current meta-analysis, more than 50% of participants described themselves as sedentary. It is possible that the effect of A-HIIT may be less in people who participate regularly in recreational exercise.[117]

Data from the meta-analysis indicate that A-HIIT had a small but significant effect on lower limb strength. Earlier research into the effect of training in an aquatic environment has been unable to demonstrate increases in knee strength.[118] The reasons for this disparity between earlier work and the current study may relate to the type of exercise training utilised and the supervised nature of the training sessions. Previous studies have suggested that inadequate application of resistance in water, or lack of fast-paced movement, is a significant contributor to the limited effectiveness of aquatic exercise interventions in improving peripheral muscle strength.[118, 119] In order to achieve high intensity exercise, all studies included in the current meta-analysis utilised fast-paced movement in water, which increased the resistance load applied to the muscles. This, in turn, may explain the gains in strength. Nevertheless, the effect of A-HIIT on lower limb muscle strength was lost during the sensitivity analyses in which studies

with high attrition bias and shorter duration training lengths were removed. This limits our confidence in this result.

Data from the meta-analysis suggest that A-HIIT does not have an effect on body composition. This contrasts with other studies that have demonstrated significant changes in both fat mass and lean muscle mass utilising either HIIT, the aquatic environment or both.[120-122] The disparity between these earlier studies and the current study does not appear to relate to differences in training dose, with earlier work showing differences with 8 weeks of training.[43, 120] It is likely that the lack of effect on measures of body composition in the current study reflect; (i) the small number of studies available to contribute data to these meta-analyses, and (ii) the limited sensitivity of some of the outcome measures to detect small change (e.g. skinfold measurements).

Regarding safety, data presented in this study indicate that A-HIIT is safe for a non-athletic population. This is consistent with other systematic reviews that show a trivial number of adverse events associated with both HIIT exercise and aquatic exercise.[22, 116]

3.4 Strengths and limitations

The main limitation of this study relates to the inclusion of a relatively small number of published studies. Also, most studies were at high risk of some bias. For the outcomes explored in this meta-analysis, only one study was rated as low risk for an outcome assessor blinded to group allocation.[106] Overcoming this methodological shortcoming should be considered when designing future studies in this area.

We anticipated, a priori, that there would be considerable disparity in the HIIT intervention, both in terms of prescription (e.g. work:rest ratios) and total dose (duration

and frequency of training). Therefore, we did not aim to compare training protocols in terms of effectiveness. Notwithstanding this limitation, the intervention used in the studies in this review were clearly described and monitored, as evidenced by high CERT scores. This makes replication feasible in clinical practice.

3.5 Conclusions

Based on the results of this systematic review with meta-analysis, in a non-athletic population, A-HIIT may produce a moderate improvement in aerobic performance and a small increase in lower limb strength, but there is no evidence that it can change body composition. Aquatic HIIT appears to be safe and feasible in elderly and clinical populations, such as those with minor arthritis and hypertension. More studies that ensure blinding of the outcome assessors and recruit larger participant numbers in diverse populations (e.g., children, clinical populations) are required to increase our confidence in the results of these meta-analyses.

This systematic review and meta-analysis demonstrate the effectiveness of an A-HIIT model using exercise stations in improving aerobic capacity. Specifically, it targets populations who are unused to vigorous exercise or who have impairments that limit the intensity that can be reached during land based exercise. A-HIIT programs in children and adolescents with cerebral palsy (CP) have not been explored, but some barriers to the training dose tolerated are similar to this study's elderly population such as pain during exercise, fatigue, poor balance and difficulties with co-ordination.

There are several trial components related to exercise prescription, the physical environment and factors that increased the risk of bias in this study, that were crucial in the design of an A-HIIT program in adolescents with CP. The thorough reporting of

exercise interventions analysed using the CERT in the systematic review, assisted in the design of the A-HIIT program for adolescents with CP. In particular the need for close monitoring of intensity of exercise using objective heart rate measurement, the use of exercise stations rather than swimming skills. and the benefits of a supervised group program. Highlighted in the inclusion criteria design of this study, was the difficulty in comparing groups that were not homogenous. The population group of adolescents with CP GMFCS level II was chosen to limit differences in functional and aerobic capacity and to maximise potential success in evaluating the A-HIIT exercises tolerability and feasibility.

The meta-analysis also demonstrates that low volume HIIT programs that focus on a slightly lower work or heart rate with longer bouts of intensive exercise and repetitions, undertaken in an aquatic environment, are still effective at inducing skeletal muscle adaption and improving functional performance. Further, it confirmed that low volume HIIT programs in water are tolerable and safe in populations that may not be used to frequent vigorous exercise. The A-HIIT program designed for adolescents with CP, was based on exercise prescriptions explored in this study. Utilising a low volume HIIT approach, the absolute intensity of the work bouts was lower than traditional HIIT programs in elite athletes, and the duration was slightly longer with shortened rest intervals. This practical HIIT model which considers potential movement limitations, consisted of 10 × 60 second work bouts at a load intensity that elicits greater or equal to 80% of maximal heart rate, interspersed with 60 seconds of recovery. To decrease the complexity of monitoring intensity in the water, rest was defined as minimal activity but not based on a recorded HR or RPE. This protocol was still time efficient in that only 10 min of exercise was performed over a 20 min training session.

An important difference when considering the design of the CP A-HIIT study, is the population group. There is disparity in age, potential difference in previous exposure to vigorous exercise and possible cognitive impairment in adolescent populations with CP. This further highlights the need to undertake a pilot study in determining feasibility of any A-HIIT program in this population, prior to undertaking a definitive trial.

Chapter 4:
Aquatic high intensity interval training study

4 Aquatic high intensity interval training study

Exercise training conducted in an aquatic environment using a high intensity interval training approach may be an effective and enjoyable method to increase exercise capacity for adolescents with cerebral palsy.[12, 22, 31, 43] The paucity of studies that have attempted aquatic high intensity interval training in adolescents with cerebral palsy means that a number of challenges need to be considered prior to conducting a definitive randomised controlled trial (RCT).[31] For example, the ability to measure the 'real time' heart rate responses of participants in water to titrate high intensity is unknown, as is the tolerability of this type of training for this population, and effect of potential physical barriers such as pain. Further, the feasibility to quantify improvements in aerobic capacity following aquatic interventions in this population using gold standard measures; namely peak rate of oxygen uptake measured during a laboratory based cardiopulmonary exercise test, is unclear, making prospective sample size calculations challenging. In these situations, there is strong support for a pilot study.[32]

Pilot RCTs are designed to test the feasibility and acceptability of the study protocol.[32] The aim of this pilot RCT was to report on outcomes, defined *a priori*, to determine if the study protocol was; (i) feasible to progress to an RCT in its current format, (ii) feasible to progress to an RCT but required modification, or (iii) not feasible to progress to an RCT without substantial modification.

This chapter outlines the methodology and results of a pilot randomised controlled trial (RCT) undertaken to:

1) Investigate the feasibility of a protocol for a future RCT, based on the following question: In adolescents with CP who are described as Gross Motor Function Classification System (GMFCS) level II (Figure 2-1: Description and illustration of the Gross Motor Function Classification System (GMFCS) – expanded and revised for adolescents between 12 and 18 years), does a 10-week water-based high intensity interval training (HIIT) program change aerobic capacity (primary outcome), lean muscle mass, pain and social interaction (secondary outcomes), over and above usual care?

2) Develop a standardised A-HIIT intervention for adolescents with CP in which intensity was measurable in real-time

3) Explore the way adolescents with CP participate in and respond to vigorous exercise and perceive the effects of aquatic therapy.

This study has been published as:

Depiazzi J, Smith N, Gibson N, Wilson A, Langdon K, Hill K. Aquatic high intensity interval training to improve aerobic capacity is feasible in adolescents with cerebral palsy: pilot randomised controlled trial. *Clinical Rehabilitation* Epub 2020 Sep 9 https://doi.org/10.1177/0269215520956499

This chapter presents this publication in a thesis format and includes additional details regarding methodology.

4.1 Study design

A pilot randomised controlled study design was undertaken.

4.2 Ethics and approvals

This study was prospectively registered with the Australian New Zealand Clinical Trials Registry (ANZCTR: 12618001926224). It was given ethical approval by the Human Research Ethics Committees of Child and Adolescent Health Service (HREC: 2016048EP) and Curtin University (HREC: 20170282)

4.3 Consumer engagement

Two adolescent consumers and their parents were employed to assist with study design. They were met on two occasions by the investigators (JD and NS). Structured and unstructured questions were asked around the procedures and processes of the assessments and interventions, and ways to facilitate participation, enjoyment and attendance. Examples of questions included 'Would you prefer the assessments to be all on one day?'; 'Do you think you could do this exercise fast in the pool?'; and 'Would you like to watch the exercises on videos before attending an exercise class?' Specific feedback was sought on visual aids for rate of perceived effort (the 'working hard' scale Appendix 3) and pain descriptors (Appendix 4 and Appendix 5) regarding usefulness and age-appropriate language and graphics. The effort scale was adjusted for colour and facial expression following feedback.[123] The consumers requested that participants have input into music choices for the intervention. Concerns were raised by the adolescent girls and their families on the impact of menstruation cycles on water exercise. This led to investigation of products available on the market (e.g., 'period proof' swimwear, https://www.modibodi.com/collections/swim) and subsequent uptake and use of products by female participants in the intervention. Feedback from participants in this cohort was that this directly influenced attendance in the intervention group.

4.4 Study protocol

Participation in the study involved completing a baseline assessment period, an intervention period and a re-assessment period. On completion of the baseline assessment period, participants were randomised to an intervention or control group. Participants randomised to the intervention group completed two A-HIIT sessions per week over 10 weeks. The control group continued with usual care which did not involve contact with the study team. On completion of this intervention period, the re-assessment period was completed over 2 weeks (Figure 4-1). Terms used to describe the intervention and baseline assessments are defined in the data dictionary (Appendix 6).

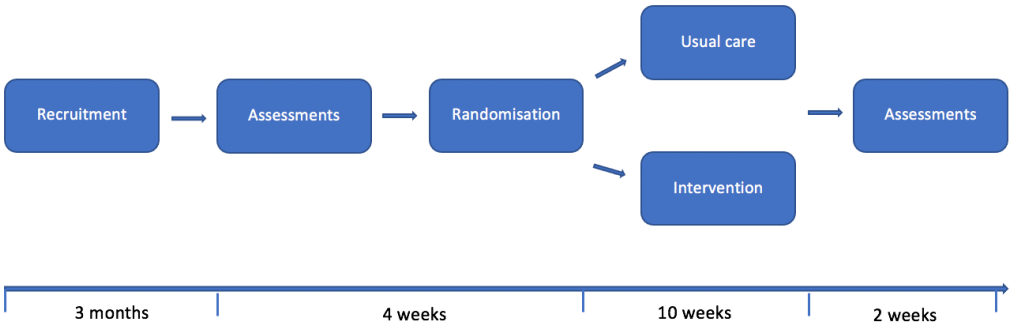


Figure 4-1: Study design timeline over 28 weeks

4.5 Eligibility criteria

Adolescents aged 12 to 17 years were eligible to participate in this study if they:

1) had a diagnosis of CP and classification as GMFCS level II (Figure 2-1: Description and illustration of the Gross Motor Function Classification System (GMFCS) – expanded and revised for adolescents between 12 and 18 years)

2) were born at not less than 28 weeks gestation and were discharged from hospital without an oxygen requirement, and

3) had no absolute or relative contraindications to cardiopulmonary exercise testing (CPET) as defined by the American Thoracic Society/American College of Chest Physicians.[124]

Potential participants were excluded if they:

1) had a concomitant medical condition limiting exercise performance or precluding participation in a high intensity exercise program (e.g. unstable seizures)

2) had any intervention that may decrease the capacity to exercise during the study period (e.g. orthopaedic surgery)

3) were unsuited to the hydrotherapy pool (e.g. open wounds, water fear)

4) were unable to follow instructions and participate in assessments and group exercise.

4.6 Screening and recruitment

Between December 2018 and April 2019, participants were identified by clinicians through the Perth Children's Hospital (PCH) paediatric rehabilitation clinics, the PCH CP clinical database, and advertisements through Western Australian community therapy agencies and social media. Presentations were made across medical teams at PCH and within community agencies to disseminate trial information and encourage enrolment. Written open invitations with study details were sent to metropolitan private practices known to treat this cohort. Written information and invitations to participate in the study were sent to pre-selected parents and adolescents who were given a 2-week period to actively opt out of further communication regarding the study. Those who did not make contact were telephoned for further screening and invitation to enrol by the principal investigators (JD, NS, NG). If deemed suitable, potential participants and their parents were invited to attend an initial appointment during which written consent (parent) and verbal assent (adolescent) were obtained. All participants enrolled in the study were deemed suitable to participate by a respiratory and/or paediatric rehabilitation medical consultant prior to baseline measures.

4.7 Randomisation and blinding

Participants were randomly allocated to intervention or control group after baseline assessments. The randomisation sequence was computer generated and concealed using opaque envelopes. Block randomisation was used for age groups of 12–15 and 16–18 years. Both randomisation and group allocation were performed by someone not involved in the study, with all other study personnel unaware of the sequence and randomisation process.

4.8 Intervention period

4.8.1 Control group

Participants allocated to the control group were asked to continue with their usual care and normal activities of daily living. There was no further interaction with the research team until the re-assessment period.

4.8.2 Intervention group

The intervention group were invited to participate in a water-based HIIT program at PCH hydrotherapy pool (mean temperature 33.6 degrees Celsius, standard deviation [SD] 0.2) during the school term April to July 2019. Parents were reimbursed for travel and parking expenses and refreshments were provided for participants and their families during and after each class. The intervention was conducted by two experienced clinicians in the water providing instructions, physical assistance for learning where required and encouragement. A third clinician provided poolside assistance with equipment, coaching, interval timing and troubleshooting technical difficulties with the heart rate system. When available, assistance was provided by a fourth staff member/therapy student in the pool. A research assistant was present at each class to assist with data collection (as described in section 4.8.6). At the beginning of each class, before entering the pool, participants were asked open-ended general health and activity questions about the days since their last visit, for example, ‘Have you felt well the past 3 days?’, and ‘Have you done any exercise since the last class?’. Data was recorded for review of pain frequency, illness and to identify any physical activity impact from the previous class attendance ().

4.8.3 Duration of the intervention and tracking adherence

The A-HIIT intervention was conducted over 10 weeks. Classes ran 3 days/week after school and on Saturday, with participants asked to attend 2 days/week. In the event a class was missed, participants were encouraged to attend an additional class the following week and attendance was tracked using class attendance sheets (Appendix 7). Parents were asked at the end of each class when participants were next likely to attend and were encouraged to contact the investigators if they were unable to. Contact was made with families if the child did not attend when expected and reasons for non-attendance were recorded. Participants were asked to make up missed classes within 1 week. Each exercise class was 40 minutes in duration and comprised 5 minutes of warm-up exercises, 20 minutes of HIIT, followed by 5 minutes of cool-down exercises and 10 minutes of stretches.

4.8.4 Exercise design

The design of the HIIT program utilised exercise stations rather than swimming, which allowed for accurate exercise dosage description and standardisation while accommodating the individual physical abilities of participants. Standardised exercises for the intervention were developed by the principal investigators (JD and NS). The exercises were based on exercise interventions described in the systematic review (Chapter 3), clinician experience, and equipment available at the facility and within reason for community agencies to purchase should the program be outsourced to local centres.[21, 31] Each exercise was described according to the Consensus on Exercise Reporting Template (CERT)[96] to assist repeatability of the intervention (Table 4-1).

Each class consisted of six to eight different exercises across 10 exercise stations. The same class design was repeated for three classes across the week. During each class, four stations were consistently utilised: two underwater bike stations, a 20 cm step-up station and a waist-deep running station. Thereafter, the remaining six stations differed between weeks, but were selected from a bank of 26 unique stations (Table 4-2). An exercise station plan was circulated at the beginning of each week and circulated among clinicians in the pool (Appendix 8). A visual pool map of stations was provided each week (Appendix 9) to assist equipment set-up by study investigators.

Where possible, exercises enabled a progression of individual workload by adding resistance or speed to assist individual participants to maintain/obtain targeted heart rates (HRs). A focus was on large muscle groups and gross motor movement rather than precision/quality of movement. All of the exercises were trialled in group format with healthy fellow colleagues and their children to ensure that target intensity could be achieved, and that exercises were easy to understand and follow without prior training and suitable for a small pool environment (Table 4-2). Videos utilising typically developing children to demonstrate the exercises to participants were developed and posted on a private web platform at the beginning of each week for participants to view as desired (Figure 4-2).

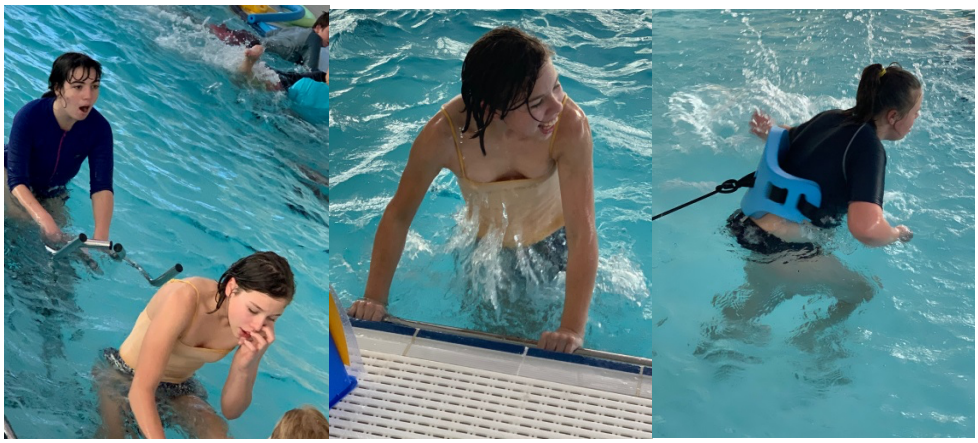


Figure 4-2: Examples of A-HIIT exercise stations

Table 4-1: Description of intervention based on Consensus on Exercise Reporting Template (CERT)

Item	**Details**
1. Exercise equipment	Warm-up: noodles, kick boards. Intervention: Noodles, foam dumbbells, kickboards, weighted step, goggles, diving rings, underwater bikes, underwater trampoline, water running vest, hand fins.
2. Qualifications and training	Three senior and one junior physiotherapist and one allied health assistant with experience in working with school-aged children with cerebral palsy delivered the program. All were certified with aquatic rescue.
3. Individual/group	Individualised target heart rates at 80% of peak heart rate (HRpeak) recorded during the baseline Cardiopulmonary Exercise Testing (CPET). The individualised high intensity interval training (HIIT) was delivered and performed in a group setting twice per week. Individual modifications to exercises were implemented by one of the senior physiotherapists as required, for example, hand strapping to assist with noodle hold.
4. Supervision	Participants were supervised in a ratio of 1:3 by physiotherapists experienced in exercise prescription. Exercises were posted on a Flickr® account so participants could see the exercises for the week ahead. The HIIT exercises were demonstrated at the end of warm-up and individuals were supported with one-on-one coaching during the program to make small adjustments to suit individuals and motivate them to reach at least 80% HRpeak.
5. Adherence to exercise	During every class, participants had their heart rate continuously monitored using chest strap telemetry (Polar® Australia, HR10) linked by Bluetooth and ANT+ connectivity to two apps: Polar Beat® (v:3.3.4, individual Samsung tablet) and Polar Club® (v:3.3.4.3, single group device iPad Pro). For each participant, the heart rate (HR) recorded during exercise, expressed as a percentage of their HRpeak, was digitally projected onto a wall using Polar Club® and colour coded to indicate at what intensity they were exercising (e.g. ≥80% HRpeak). Exercise intensity reached at each station was recorded by a research assistant and through software downloads linked to the telemetric HR monitoring device. Attendance was recorded for each participant and reported as a number of sessions out of 20 possible sessions.

Item	Details
6. Motivation strategies	Participants were encouraged with verbal feedback about their technique, both what was done well and what changes needed to be made. Technical mastery of the activities themselves was otherwise unimportant, barely visible to observers or other participants as occurring under water, and not measured. Therefore, participants were able to enjoy the program without worrying or comparing how they were moving to others. Participants were able to see their heart rate on a screen with colour coding to indicate what percentage of their maximal heart rate they had reached. For example – when a participant's HR was at high intensity, the zone would flash red; for moderate, it flashed orange, and for low intensity, green. Not only were the participants engaged and encouraged by these visual cues, but also the trainers, carers and other attendees spontaneously spurred on participants during sessions. Achievement of heart rate goals was evident and therefore celebrated enthusiastically during each activity by all present. Each participant was emailed at the end of the class their achievements of intensity for each exercise station and average class maximal heart rate for feedback and encouragement.
7a. Decision rule(s) for determining progression	If the participant was meeting their heart rate intensity, exercises were not changed or progressed. Changes to exercises if heart rate was not being met included encouraging faster movements and/or decreasing complexity of upper or lower limb co-ordination needed so the participant could exercise at higher intensities.
7b. How program was progressed	Once the participants were familiar with the program and were seen to easily achieve 80% of HRpeak, the colour codes were changed (e.g. red was now 85% HRpeak) to encourage motivation. This did not change the definition of an interval success. Four to six exercise stations were changed each week and were able to be progressed as participants became familiar with them (e.g. tramp jumping to tramp jogging).
8. Exercises	Please refer to Table 4-2 **Description of aquatic HIIT exercises** for the types of HIIT activities utilised. Each session followed the following structure: Warm-up – 5 minutes; movement of all joints in neck and spine, gentle walking, kicking and activation of core muscles/leg and arm muscles in preparation for HIIT activities. HIIT exercises – 20 minutes; 10 stations with 1 minute of HIIT and 1 minute of rest. Relaxation and stretches – 10–15 minutes; floating, trunk and body relaxation with therapists assisting, and active stretches of legs, spine and arms.
9. Home program	Participants were not asked to do a home program.

Item	Details
10. Non-exercise components	Not applicable.
11. Type and number of adverse events	Four minor adverse events that did not require medical attention were recorded throughout the program, including one stubbed toe, foot cut and stomach pain after intervention. Only one was related directly to exercise, which was lower back pain.
12. Setting	The program was delivered at the Perth Children's Hospital hydrotherapy pool, Western Australia, with a mean temperature of 33.6 ± 0.2 degrees Celsius.
13. Exercise intervention	Participants were asked to attend two 1-hour sessions per week for a total of 10 weeks. Classes were held on a Monday and Wednesday after school and on Saturday mornings.
14a. Generic/tailored	Each participant worked at an individually tailored intensity which was ≥80% of their peak heart rate recorded during their baseline CPET.
14b. How the exercises are tailored	The exercises were tailored for each participant according to the identified impairments impacting their ability by level of difficulty. The physiotherapists progressed the exercises according to the participant's response. Modifications to exercises were done by the physiotherapists if necessary, for example, not including bilateral co-ordination of arms and legs if this was impacting on the participant's ability to achieve intensity.
15. Decision rule for starting level	Each participant was asked to exercise at an individualised intensity of 80% HRpeak, pre-determined by their baseline CPET.
16a. Adherence/fidelity	All physiotherapists and the assistant undertook 4 hours training delivered by NS and JD covering the theoretical and practical underpinnings of the exercise program prior to the commencement of the program. The intervention was delivered by at least two physiotherapists who were the same from week to week. Each week JD and NS met to discuss exercise progressions and fidelity issues and troubleshoot motivational and exercise changes to ensure the exercises were consistently achieving high intensity for the participants.
16b. Intervention delivered as planned?	The intervention was delivered as planned. Participants in the intervention group attended a median of 99.5% of sessions.

Table 4-2: Description of aquatic high intensity interval training exercises

Exercise title	**Exercise number**	**Equipment required**	**Exercise instruction**	**Water depth for participant**	**Area in pool 1 = shallow 2 = middle 3 = deep 4 = all**
Step-ups	1	Underwater step	Hold the rail with one hand – step up and then back down as fast as you can.	On the step, hip height	1
Kickboard swish	2	Kickboard	Stand with legs wider than shoulder width apart. Hold kickboard vertically with half under water. Push the kickboard away from your chest as you squat down and then bring it back in as you stand up. Repeat as quickly as possible.	Waist depth	2
Kickboard rowing	3	Kickboard	Stand with legs wider than shoulder width apart. Hold kickboard vertically with half under water. Pull the kickboard to each side like you are paddle rowing. Twist in the middle to get both arms to each side. Repeat as quickly as possible.	Waist depth	2
Patter kick supine	4	Kickboard	Float on your back with the kickboard hugged to the chest with both arms. Kick fast and furiously from one end of the pool to the other.	Supine floating	4
Rail kicking	5	Edge of pool railing	Hold onto the side of the pool floating on stomach with neck and arms extended. Kick legs as fast as you can.	Prone floating	3
Sit to stand	6	Underwater chair or pool steps	Sit under water on a chair or step, move from sitting to standing and return to sitting. Movement should be fast and continuously repeated. Arms can be added to balance or increase resistance.	When sitting, waist deep	1
Dumbbell punches	7a	X 2 dumbbells	Stand with legs wider than shoulder width and knees slightly bent. Dumbbells should be vertical and held under water. Repeatedly punch left then right upper limb forwards; ensure dumbbell is under water.	Water just above elbows	2

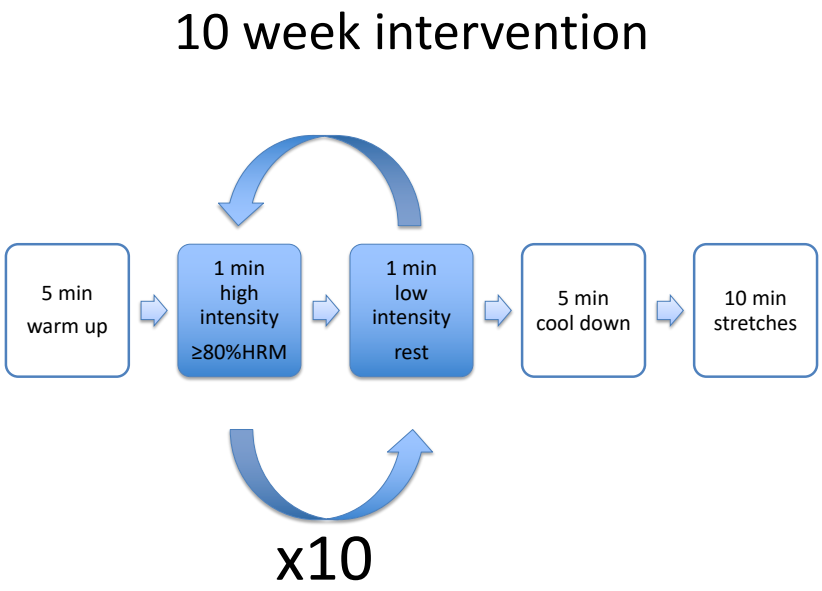
Exercise title	Exercise number	Equipment required	Exercise instruction	Water depth for participant	Area in pool 1 = shallow 2 = middle 3 = deep 4 = all
Dumbbell punches and cross leg jump	7b	× 2 dumbbells	Stand with legs wider than shoulder width and knees slightly bent. Dumbbells should be vertical and held under water. Repeatedly punch left then right upper limb forwards; ensure dumbbell is under water. At the same time, jump so that your right leg is in front, followed by the left leg in a scissor-like motion.	Water just above elbows	2
Pull buoy jump	8	Pull buoy	Hold onto a pull buoy above your head, bend legs down into squat position until your neck is submerged in water. Push off through legs and jump into the air, pushing the pull buoy above your head. On return, relax down into the crouch position again. Repeat continuously.	In crouch, neck level	2
Water running	9	Nil	Run as fast as you can from side to side of the pool– use your arms to assist.	Hip depth	1 or 2
Jump and swish	10	Nil	Swish both your arms to one side of your body while jumping and twisting your body to the opposite side. Do this as fast as you can.	Waist depth	2
Tramp jumping	11a	Trampoline	Hold onto the edge of the pool railing, jump quickly and repeatedly on the trampoline.	Waist depth	2
Tramp jogging	11b	Trampoline	Jog fast on the trampoline with your arms moving in a running action.	Mid chest	2
Tramp push-ups	11c	Trampoline	Holding onto the edge of the pool with bent arms, jump at the same time as straightening your arms to do a ‘push-up’ against the side of the pool. Repeat quickly.	Mid chest	2

Exercise title	Exercise number	Equipment required	Exercise instruction	Water depth for participant	Area in pool 1 = shallow 2 = middle 3 = deep 4 = all
Cycling	12	Bike	Wearing bike shoes, cycle as hard and as fast as you can.	Sitting on bike, waist deep	2
Noodle skiing	13	Noodle	Hold the noodle in each hand with the noodle arched above you. Punch arms forward alternately while scissoring legs forwards and back in a skiing motion. Repeat quickly.	Elbow depth	2
Noodle wave/wiggle	14	Noodle	Stand with legs wide apart and noodle stretched horizontally in front of you and just under the water, wave each end of the noodle forwards and then backwards as fast as you can.	Hip height	1
Noodle wide leg jog	15	Noodle	Tuck the noodle behind you under your arms and relax into a semi-seated position with legs wide apart. Jog with your legs as fast as you can.	Not able to touch the ground	3
Noddle pony rides	16	Noodle	Sit over the noodle, cycle with your legs while breaststroking with your arms. Move from side to side of the pool or stay in one spot.	Not able to touch the ground	3
Noodle scissors	17	Noodle	Stand in the water with the noodle around your waist and held in each hand. Cross the two ends of the noodle and then pull apart. Repeat quickly.	Mid chest	2
Noodle star jumps	18	Noodle	Hold the noodle so it makes an arch above your head. Start with your elbows bent by your side. As you ‘jump’ out with your legs, straighten the noodle. As you ‘jump’ in, bring the noodle ends back to your side. Repeat quickly.	Waist depth	1 to 2
Hand paddle scissors	19	Hand fins	Stand with wide legs, hand fins slightly under water and vertical. Push arms out to sides as far as you can then pull them back to the middle. Repeat as fast as you can.	Mid chest	2

Exercise title	**Exercise number**	**Equipment required**	**Exercise instruction**	**Water depth for participant**	**Area in pool 1 = shallow 2 = middle 3 = deep 4 = all**
Basketball jog	20	Balls, basketball hoop	Water jog/swim with the ball above your head. Place in a hoop by the side of the pool. Swim fast to the other side to collect the next ball. Repeat.	Not able to touch the ground	3
Noodle tug of war	21	Noodle	Push and pull the noodle with resistance from your partner.	Waist depth	1 to 2
Duck dive fetch	22	Weighted rings/toys	Dive down to pick up weights from bottom of the pool and swim to place them in a basket. Pick up as many objects from the bottom as you can.	Not able to touch the ground	3
Bungy run	23	Running vest and elastic	Put vest on which is attached to the railing of the pool. Walk away from the side of the pool so rubber tubing is taught, run against the resistance of the elastic as fast as you can on the spot.	Waist to mid chest	1 to 2

4.8.5 Exercise intensity

HIIT is often based on maximal or supramaximal work rates. This study sought to design a practical model of HIIT that was time efficient while also having wider application to populations other than just healthy athletes. To accomplish this goal, the absolute intensity of the work bouts was decreased, but their duration was increased. This practical HIIT model involved participants exercising at two different intensities: high, defined as exercise that elicited an HR response ≥80% of their peak heart rate (HRpeak) measured using CPET; and low, defined as rest with minimal movement and progression to the next exercise station. The ratio of high to low intensity exercise was 60 seconds high to 60 seconds low and this 2-minute cycle was repeated 10 times (Figure 4-3). The protocol was time efficient in that only 10 minutes of exercise was performed over a 20-minute training session. Importantly, this practical, time-efficient HIIT model is effective at inducing rapid skeletal muscle remodelling towards a more oxidative phenotype, as demonstrated in previous Wingate-based HIIT studies of low-volume HIIT protocols in athletes and the systematic review outlined in Chapter 3.[9, 125]



HRM= heart rate maximum

Figure 4-3: HIIT design: 10-week intervention with two training sessions per week

4.8.6 Exercise monitoring

To titrate and record training intensity during A-HIIT, HR was continuously monitored during each exercise station using HR chest strap telemetry (Polar® Australia, HR10). This device samples HR data in real-time, at 60Hz. This was linked via ANT+ and Bluetooth technology to an app (Polar Beat®, v:3.3.4) on a tablet device (Samsung Galaxy, South Korea). Participants' HRs were also bluetoothed to an exercise group display app (Polar Club®, v:3.3.4.3) located on a separate tablet device (iPad Pro, Apple, United States) and projected on the wall behind the pool. In this way, each participant was able to visualise their HR in real time. The projection of the HRs were colour coded to reflect intensity, for example, red equalled ≥80% HRpeak. During each exercise station, participants were strongly encouraged to achieve ≥80% HRpeak (Figure 4-4). Immediately after an interval of HIIT, participants were asked to ensure their chest strap was above the water line if possible, to assist with bluetooth transmission. Participants' HRs were continuously monitored during the class by a research assistant. Using data shown on the group app display, the research assistant was responsible for recording whether or not each participant, during each work interval, met the criteria for interval success (defined as achieving ≥80% HRpeak) (Appendix 7). On class completion, data from the Polar monitor were downloaded using the Polar Beat® software (Polar, Finland). These data were used to verify whether or not a participant met the criteria for interval success if the Bluetooth technology failed to project during the class. (Appendix 10). Further, for each participant, the peak heart rate was recorded during each work interval where available. HRpeak was defined as the average of the highest three HRs in a 60-second interval of exercise. The highest

HR data from available data using either the Polar Beat® group app data or Polar Beat® download was recorded as the interval success HR (Appendix 11).

Figure 4-4: Picture of the participants and projected heart rate during class

4.9 Outcome measures related to assessment of feasibility

4.9.1 Primary outcome measures of feasibility

Primary outcomes used for the evaluation of protocol feasibility were determined a priori and defined in Table 4-3. They were based on measures of: 1) recruitment, 2) completion of baseline assessments, 3) adherence to the intervention, 4) intervention fidelity, and 5) completion for all components of the study protocol.

Table 4-3: Primary outcome measures to evaluate feasibility of using this study protocol to progress to a definitive randomised controlled trial

Feasibility outcome	**Method used to calculate**	**Target**
Recruitment fraction	The number who provided consent and assent, expressed as a percentage of those who met study criteria and were approached to participate.	≥75%
Completion fraction for baseline assessments	Of those who agreed to participate, the percentage of participants who completed all baseline assessments.	≥70%
Adherence fraction to intervention	Of those randomised to intervention, the number of classes attended and participated in, expressed as a percentage of total classes expected to attend.	≥80%
Intervention fidelity fraction	The percentage of exercise intervals where target heart rate was reached, expressed as a percentage of the total number of exercise intervals completed during the classes.	≥85%
Completion fraction for all components of the study protocol	Of those randomised, the percentage of participants who completed all aspects of the study.	≥70%

These primary study outcomes were used to decide if the study protocol was: (1) feasible to progress to an RCT in its current format; (2) feasible to progress to an RCT but requiring some modification; or (3) not feasible to progress to an RCT without substantial modification (Figure 4-5).

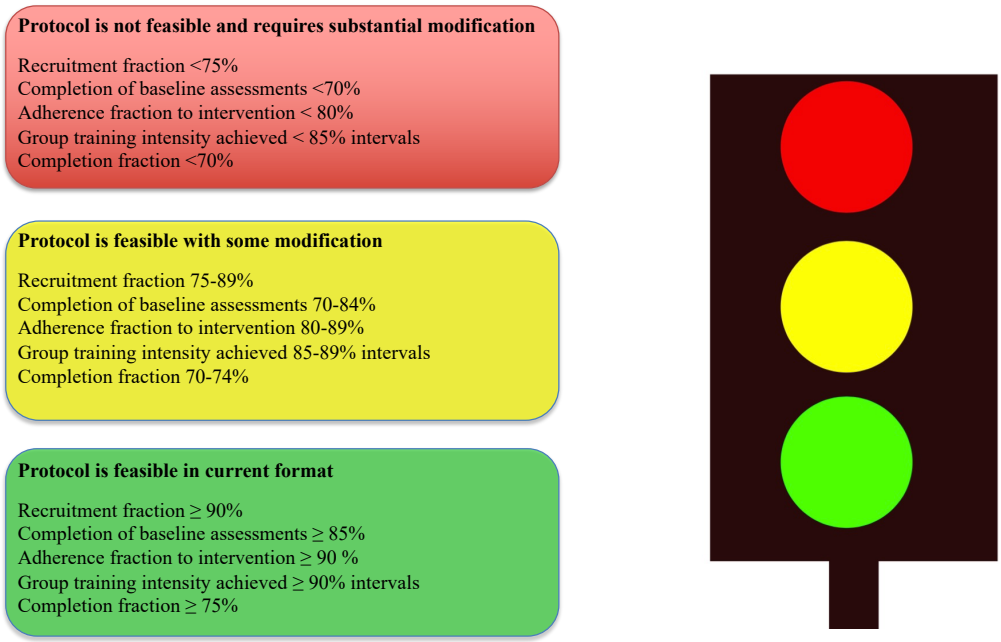


Figure 4-5: Traffic light feasibility to progress to a randomised controlled trial

In addition to these primary outcomes, for each participant, three binary outcomes were also generated for each class. These were; (1) completion (or not) of an exercise class, defined as participation in all 10 intervals of HIIT in an exercise class, (2) achievement (or not) of HIIT throughout the class defined as achieving interval success (i.e. ≥80% HRpeak) for at least 9 out of 10 intervals and (iii) adequate (or not) attendance defined as participation in at least 80% of all scheduled classes (i.e. at least 16 classes over 10 weeks). Finally (3), participants were defined as achieving 'overall' intervention fidelity if ≥85% of all intervals completed by the participant over 10 weeks met the criteria for interval success. Where HR data were missing, it was recorded as an unsuccessful HIIT interval. Success of HR telemetry was evaluated through the number of missing HR data and therefore the ability to determine interval training intensity.

4.9.2 Intervention group interviews

For those participants in the intervention group, health-related quality of life (HRQoL) interview questions were administered immediately before each exercise class and recorded (Appendix 12). Tolerance of the intervention was monitored at the end of each class through considering participant responses to pre-determined questions regarding their general health, activity levels and pain since the previous class. Specifically, participants were asked if they had pain, to describe their pain and to comment on its intensity. Visual cues using Wong-Baker pain faces[126] and word descriptors with analogue scales of 0–10 were used to assist participants to quantify the intensity of any pain both before and immediately after the intervention (Appendix 4 and Appendix 5). Participants' comments related to the intervention that the principal investigators felt were of value were recorded verbatim.

On completion of the intervention period, families of those randomised to the intervention group were interviewed by telephone in a structured format by a clinician. (Appendix 13). To minimise social desirability bias, the clinician who conducted these interviews was not the principal investigators (JD and NS). Questions were directed to assist in A-HIIT exercise design changes and gather informal feedback that participants made to their parents. Comments were recorded verbatim.

4.9.3 Secondary outcome measures of feasibility

Secondary outcomes related to screening, recruitment, randomisation, retention, assessment completeness and intervention adherence and fidelity were also collected (Table 4-4). These are defined in the data dictionary (Appendix 6).

Table 4-4: Secondary outcome measures

Study component	Measure of feasibility	Examples of data collected to explore issues related to measures of feasibility
Screening	Number of participants screened	Number of participants screened who met eligibility criteria Number of participants who opted out after receiving letter of invitation
Recruitment	Number of participants invited who enrol in the study	Reasons given for declining participation e.g. travel distance, sibling commitments Demographic data of participants who decline and participate e.g. postcode, age, gender
Randomisation	Tolerance of randomisation	Number of participants who withdraw from the study after randomisation Description of unbalanced groups e.g. gender
Retention	Participant retention	Attendance vs non-attendance at assessments (both pre-randomisation and following the intervention period): reasons related to non-attendance at assessments e.g. acute illness Attendance vs non-attendance at each intervention exercise session: reasons related to non-attendance at intervention e.g. session day, session time, conflicting family schedules
Assessment completeness	Proportion of planned assessments that are completed Adherence to assessment protocol e.g. assessment fidelity	Completeness vs non-completeness for each assessment measure: reasons for non-completeness e.g. demographic data that may be related to non-completeness such as postcode address Number and type of adverse events during the assessments Time requirement (participant and investigator) for assessment completion For each CPET, the proportion who meet the criteria for a maximal test (as determined a priori to PCH CPET guidelines)
Intervention adherence	Adherence to intervention protocol e.g. intervention fidelity	Adherence to training intensity: number of high intensity intervals per class where a participant reaches their target heart rate Equipment and software malfunction e.g. unusable heart rate data Number and type of adverse events during intervention Time requirement (participant and investigator) for each intervention session

CPET = cardiopulmonary exercise testing; PCH = Perth Children's Hospital

4.10 Outcome measures related to estimating the effect of the intervention

Measures used to estimate the effect of the intervention were collected during baseline assessments (up to 4 weeks before the intervention) and post-intervention assessments (2 weeks after the intervention period) in all participants. All data collection took place at PCH.

4.10.1 Aerobic capacity

VO_{2peak} was measured by CPET performed on an electronically braked cycle ergometer. CPET is the gold standard assessment for determining aerobic capacity and has been used in interventional studies for cardiovascular aquatic physiotherapy programs in adults[31] and has been validated as an outcome measure in adolescents with GMFCS II CP.[27] However, the CPET has not been used to evaluate the effect of A-HIIT in children and young people with CP and has not been used as a clinical or research method in children with CP in Western Australia. The CPET protocol in this study was based on a maximal incremental cycle ergometer protocol validated in ambulant adolescents with CP[76] and PCH respiratory laboratory protocols for CPET testing in adolescents. Breath-by-breath analysis was undertaken using Vmax 229 and SensorMedics Vmax encore (Software Version 21-1A, United States), which was calibrated and maintained according to manufacturer's instructions. The test was overseen by a senior respiratory scientist with experience working with children and undertaking CPETs, and an experienced clinician who assisted with adaption to the participants' requirements such as foot taping and balance support to prevent falls and assist with cycling. A family member was also present.

Standardised instructions and strong verbal encouragement were used by the respiratory scientist to facilitate maximal effort by each participant. The test was considered successful for determining a VO_{2peak} if two out of three of the following criteria were achieved:

1) HR ≥180 beats/min
2) respiratory exchange ratio (RER) >1, and/or
3) subjective signs of exhaustion.

On completion of the test, the respiratory scientist and respiratory consultant (AW) reviewed and provided analysis of data to the investigators. Predicted VO_{2peak} was calculated using previously reported equations.[127] Both the scientist and clinician responsible for overseeing the test were blinded to group allocation during post-intervention data collection.

4.10.2 Body composition

Measures of body composition including fat mass, lean mass and bone density were collected using dual energy X-ray absorptiometry (DXA) scanner (Lunar Prodigy Advance, Belgium). The scan was administered and conducted as per the normal clinical protocol for children undertaking DXA at PCH. Scans were conducted by two radiographers familiar with PCH DXA scanning and protocols and reported as per normal clinical practice at PCH (see example in Appendix 14). The two radiographers were blinded to group allocation. Predicted equations for measures of body composition were calculated using standard PCH software.[128]

4.10.3 Health-related quality of life

Pain, activity participation and social interaction were measured using the brief Bath Adolescent Pain Questionnaire-5 (BAPQ-5),[129] the Modified Brief Pain Inventory – short form (ModBPI-sf),[129] and Pediatric Quality of Life Inventory (PedsQL™v4.0)[130] including fatigue module – adolescent version, and individualised goals were set using the Canadian Occupational Performance Measure (COPM)[131] (Appendix 15 and Appendix 16). These tools have been shown to have acceptable reliability and validity in adolescents and are responsive to change following treatment and over time.[132-137] They have been utilised with children and adolescents with CP to determine the prevalence of pain and its effect on activities of daily living.[69, 129] All questionnaires were administered as per standardised instructions by a clinician at PCH experienced with children and adolescents with CP, familiar with the assessment tools and blinded to group allocation.

4.11 Methods used to report and evaluate the pilot study

This pilot study was reported in accordance with published guidelines for reporting randomised pilot and feasibility trials.[89] Specifically, the Consolidated Standards of Reporting Trials (CONSORT) recommends that pilot RCTs are reported with pre-specified criteria used to judge whether or how to proceed to a definitive RCT.[89]As this was a pilot study, no inferential statistical analyses were conducted. The results of the study were reported using descriptive statistics such as means and standard deviations (parametric data), or medians and interquartile ranges (non-parametric or ordinal data). Categorical data were reported as frequencies and proportions. Justification of the sample size for pilot studies is a contentious issue as the sample size needs to balance issues of imprecision (small samples) against cost (large samples).[81] Although it is

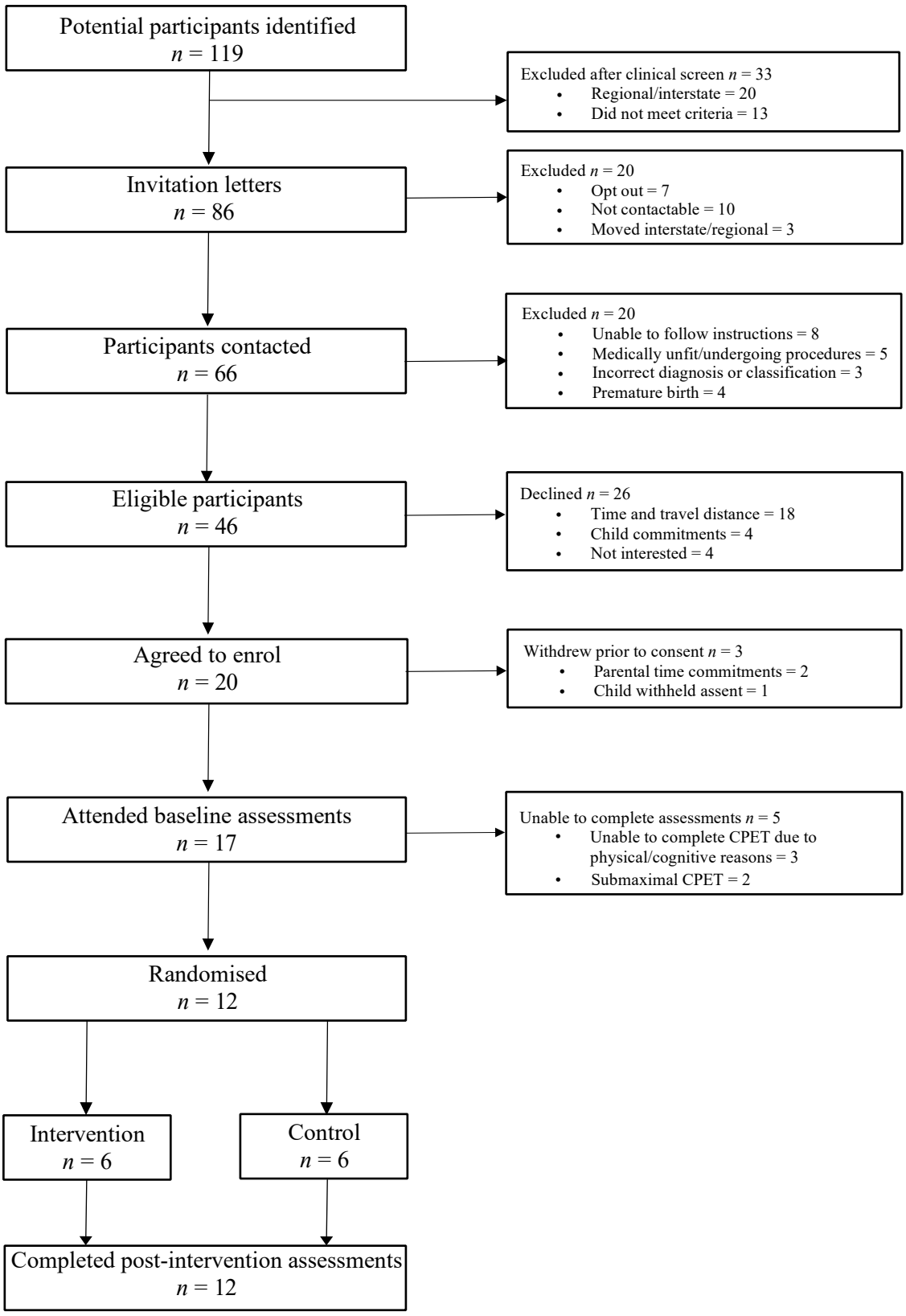
inappropriate to calculate the sample size required to have adequate power to detect a given treatment effect (between-group difference) using inferential statistics, experts agree that some justification of sample size is required.[81, 89] For this study, an approach that has been described by experts in the field to justify a sample size for a pilot study was utilised.[89, 138] This approach utilised confidence intervals (CI) to provide a level of precision around an estimate of a given feasibility outcome (e.g. recruitment fraction). Rather than the usual 95% CI required for hypothesis testing, experts suggest that an 80% CI will satisfy the need for reasonable certainty for decision-making based on pilot data.[138] This approach showed that a total sample of $n = 20$ was appropriate for this pilot study.

4.12 Results

4.12.1 Participants

Participant screening commenced in December 2018 with the first participant enrolled in April 2019. The study was completed in August 2019. Flow of participants through the study is presented in Figure 4-6. Of the 119 potential participants screened, 33 (28%) adolescents did not meet the eligibility criteria. Reasons for exclusion are given in Figure 4-7. Cognitive impairment was the most common reason (n = 21). Of 46 eligible participants, 29 (63%) declined to participate. Reasons are given in Figure 4-8. The most common barrier to participation was travel distance (n = 11). Seventeen (7 [41%] males) consented and attended baseline assessments. The recruitment fraction was 37% (95% CI 23 to 52).

Characteristics of participants are reported in Table 4-5. For participants who consented, the mean (SD) for age was 14 years 6 months (2 years), mean lean muscle mass was 31.5 (8.1) kg and mean body fat percentage was 33.9 (12.0)%.



CPET = cardiopulmonary exercise testing

Figure 4-6: CONSORT diagram

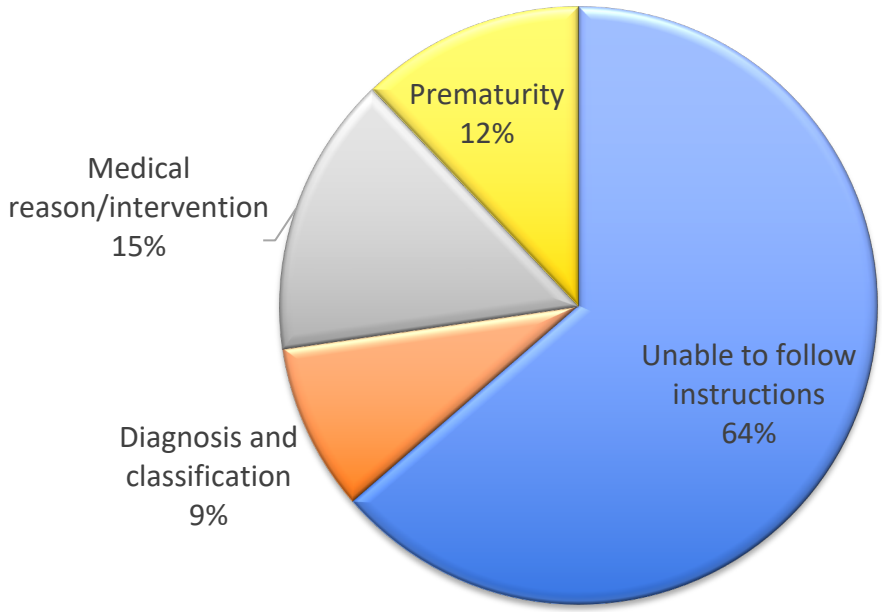


Figure 4-7: Reasons that 33 potentially suitable participants were not recruited to the study

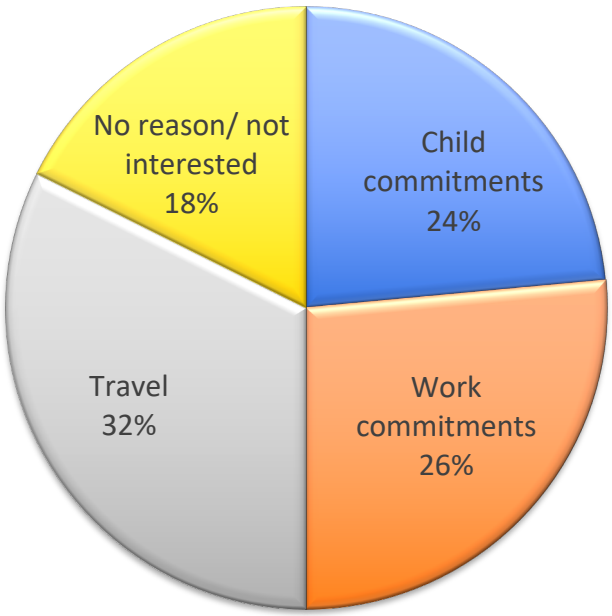


Figure 4-8: Reasons that 29 eligible participants decline to enrol in the study

Table 4-5: Participant characteristics

Parameter	**Intervention group (*n* = 6)**	**Control group (*n* = 6)**	**Non-randomised group (*n* = 5)**
Age (years. months)	14.1 (1.6)	14.7 (2.5)	13.1 (1.9)
Females: males	3:3	4:2	3:2
Weight (kg)	60.7 (13.6)	43.2 (10.4)	62.5 (19.1)
Height (cm)	161.4 (6.7)	155.2 (16.2)	162.3 (7.2)
Lean muscle mass (kg)	35.5 (6.2)	27.2 (9.2)	31.8 (7.6)
Total body fat (% body weight)	33.2 (11.6)	28.7 (10.5)	41.0 (12.7)
Involved with school educational support programs (*n*)	3	4	5

Mean (standard deviation). Anthropometric measures obtained from DXA.

4.12.2 Outcomes related to feasibility of assessments and intervention

The completion fraction for all baseline assessments was 71% (95% CI 44 to 90) (Table 4-6). All participants attempted baseline CPET; 11 (65%) required foot taping onto the cycle and seven (41%) required physical assistance (e.g. balance support). Twelve (71%) participants achieved a maximal performance during the CPET with a mean RER of 1.18 (range 1.03 to 1.24). All participants completed the baseline DXA scan. All participants attempted the ModBPI and PedsQL™. Four (24%) required coaching from a parent and one (6%) was unable to complete one element of the Physical Functioning component of the PedsQL™. Sixteen (94%) participants completed the COPM as one declined to do so. No adverse events occurred during the baseline assessments and assessors remained blinded to group allocation.

Travel time for families to PCH ranged from 18 to 62 minutes one way. Data were collected across 119 classes and 1190 exercise intervals. Five of six participants allocated to the intervention group attended all classes. One participant missed a single session due to illness. The adherence fraction to the intervention was 99% (95% CI 95 to 100) (Table 4-6). Four minor adverse events were recorded during the intervention which did not require medical attention (e.g. stubbed toe, stomach pain). Prior to commencing a class, musculoskeletal pain was reported by participants on 22 occasions resolving upon class completion on 14 occasions. Exacerbation of back pain following a class was reported on one occasion.

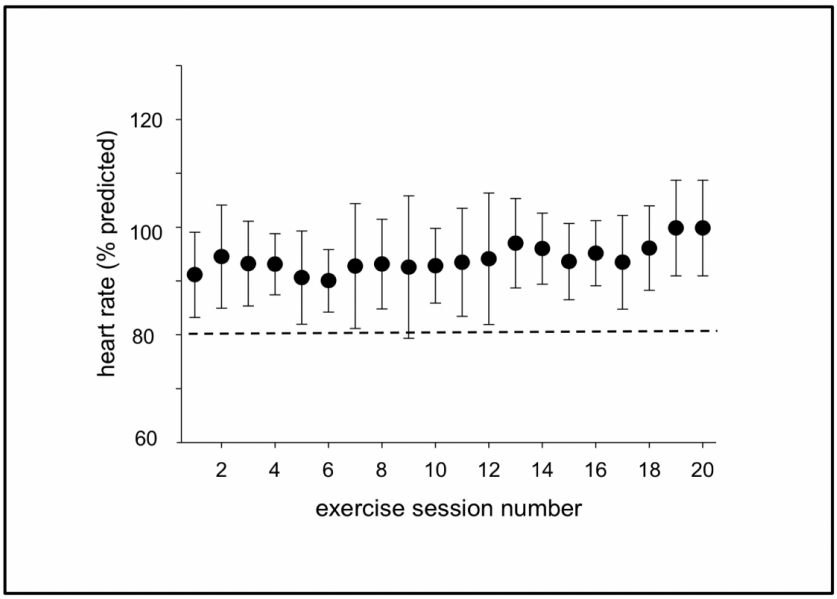
Regarding training intensity, HR data were available across 1180 intervals; data were lost on 10 (1%) intervals due to monitor failure. The intervention fidelity fraction (considering all intervals) was 93% (95% CI 92 to 95). The average exercise HR achieved during the exercise intervals, expressed as a percentage of the HRpeak, was 94

(0.1) % (Figure 4-9). A HRpeak ≥80% was achieved by at least one participant across all 26 different types of exercises selected for the exercise stations. The completion fraction for all components of the study protocol was 100%.

Table 4-6: Outcome measures to evaluate feasibility of using this study protocol to progress to a definitive randomised controlled trial

Feasibility Outcome	**Definition**	**Result**	**95% Confidence interval (CI)**	**Protocol feasibility**
Recruitment fraction	Of those invited and eligible, ≥75% agree to participate	37%	23 to 53	Not feasible
Completion fraction of baseline assessments	Of those who agree to participate, ≥70% complete all baseline assessments	71%	44 to 90	Requires modification
Adherence fraction to intervention	Of those randomised to intervention, participants attend and participate in ≥80% of classes	99%	95 to 100	No modification required
Intervention fidelity fraction	For those randomised to the intervention group, individual target heart rate is reached (or exceeded) in ≥85% of the exercise intervals	93%	92 to 95	No modification required
Completion fraction for all components of the study protocol	Of those randomised, ≥70% complete all aspects of the study	100%	74* to 100	No modification required

CI calculated for a proportion. * One-sided 97.5% CI



Data are presented as mean and SD. X axis represents each class. Y axis represents peak heart rate expressed as a percentage of the heart rate peak determined during baseline cardiopulmonary exercise testing, across all exercise intervals, for all participants in that class. Dash line = pre-determined definition of high intensity (i.e. ≥80% of heart rate peak)

Figure 4-9: Participant peak heart rate (expressed as % predicted) across each class

4.12.3 Interview data

During exit interviews, participants and families reported the intervention was age appropriate and fun, and felt it improved function, was family centred and encouraged friendships. All families and participants expressed a desire to continue with a similar program despite the substantial time commitment and travel. Organising appointments in school holidays, flexible days, the novel assessments, and providing parking and travel costs were all cited as advantageous to retention in the study. Nevertheless, access to the intervention at local venues was reported to be preferable. No changes to the intervention were suggested.

4.13 Outcomes related to estimating of the effect of the intervention

Measures collected during baseline and follow-up assessments are presented in Table 4-7. Of the 17 participants who undertook baseline assessments, serendipitous findings were 13 (76%) had an area identified as low or at risk of low bone density on DXA scan with a Z-score of −1.0 or below,[128] and 11 (65%) participants described pain in one or more regions.

Table 4-7: Measures collected before and after the intervention period

Parameter	Intervention group (n = 6)		Control group (n = 6)		Group unstandardised coefficients	
	Baseline	Follow-up	Baseline	Follow-up	B	Standard error
VO_{2peak} (mL/kg/min)	32.4 (9.4)	34.4 (13.3)	31.4 (4.5)	32.3 (5.2)	0.92	2.39
VO_{2peak} (% predicted)	71 (17)	76 (25)	72 (11)	74 (10)	–	–
Heart rate peak (bpm)	175 (11.2)	185 (14.5)	176 (16.8)	181 (11.4)	–	–
Lean muscle mass (kg)	35.5 (6.2)	36.3 (5.9)	27.2 (9.2)	27.6 (9.4)	0.76	0.81
Fat mass (kg)	19.5 (11.0)	19.2 (10.8)	10.9 (5.2)	11.8 (6.3)	–1.94	0.94
Bone density (TBLH Z-score)	–0.83 (1.08)	–1.03 (1.00)	–1.55 (0.71)	–1.53 (0.71)	–	–
PedsQL™: Total	70.5 (6.4)	80.8 (4.8)	71.9 (14.9)	79.4 (10.1)	2.29	2.78
Physical health	70.4 (10.5)	81.8 (9.6)	68.2 (22.1)	72.4 (23.7)	7.69	7.23
Psychosocial health	69.7 (7.1)	80.3 (3.8)	74.4 (14.2)	86.4 (7.3)	–5.39	3.51
Fatigue total score	72.7 (17.4)	79.2 (10.9)	66.3 (10.8)	76.0 (14.1)	–0.36	6.14
BAPQ (score/28)	2.8 (3.1)	3.3 (3.4)	1.7 (2.1)	2.0 (2.0)	0.39	1.13
COPM: Performance	4.1 (1.0)	5.3 (2.0)	4.3 (1.6)*	5.4 (2.6)*	–	–
Satisfaction	4.7 (2.0)	5.5 (2.7)	4.1 (1.3)	6.5 (2.2)	–	–
Pain present (n, %)	5 (83%)	4 (67%)	3 (50%)	4 (67%)	–	–

Data are presented as mean (standard deviation); * n = 5; BAPQ = Bath Adolescent Pain Questionnaire; COPM = Canadian Occupational Performance Measure; heart rate peak = peak heart rate; PedsQL™ = Pediatric Quality of Life Inventory; TBLH = total body less head

4.14 Discussion

Recruitment to this study was more challenging than expected with less than half of those screened meeting eligibility criteria. Cognitive impairment limited participation because the protocol involved reasonably complex data collection procedures (e.g. CPET) and reliable communication was essential to optimise for safety during the group aquatic exercise classes. Additional medical conditions (e.g. anaemia) and upcoming procedures (e.g. orthopaedic surgery) further excluded participants. Smaller recruitment pools than anticipated have been described in another aquatic study in young people with disability for similar reasons.[139]

Adolescents in our study who met eligibility criteria but declined to participate cited similar barriers to enrolment reported in previous studies involving young people with disability such as time, travel and other trial involvement.[139, 140] Our low recruitment fraction indicates a larger screening population would be required for recruitment of appropriate sample sizes for a definitive efficacy trial in this area. This requires multi-site collaboration and possible broadening of our target population to include GMFCS level I, posing challenges (including financial) associated with ensuring intervention fidelity.

The completion fraction for individual baseline assessments ranged from 100% (DXA scans) to 70% (CPET). This range likely reflects the physical and cognitive requirements for the different assessments. The CPET was novel to all study participants and some (n = 5; 29%) were unable to achieve the criteria needed for the test to be considered ‘maximal’. This was largely due to underestimation by families at enrolment of the physical and procedural learning challenges required.[141] It is unlikely performance in CPET would improve with the introduction of a familiarisation test as

re-test repeatability of VO_{2peak} in this population is reported as excellent.[142] Although optimising the number of participants who can achieve a 'maximal' CPET performance is an important consideration when exploring the effect of this intervention using gold standard measures, it is unlikely to be an issue in clinical practice as changes in exercise capacity can be assessed using less complicated field-tests, such as the modified shuttle run, which have been validated in CP.[37]

Despite good HRQoL questionnaire completion data, some participants required coaching, which may be explained by cognitive fatigue described in this population,[67] induced by the number and repetitive nature of these questionnaires. Nevertheless, as the effect of aerobic exercise on HRQoL in adolescents with CP is unknown, such assessments are likely to be important in these studies.[143] Future studies may need to consider careful rationalisation of the type and number of HRQoL assessments to minimise cognitive fatigue and maximise data quality.

This study comprehensively tested the feasibility of an A-HIIT program and successfully monitored and collected HR in the water using telemetry. It was anticipated that, for adolescents and populations with a physical disability, conventional HIIT at maximal or supramaximal work rates may not be tolerable and therefore, the HIIT used in the current study opted for lower work rates and longer intervals.[144] The excellent adherence with our intervention suggests that it was not only tolerable but enjoyable. The only comparable HIIT study in adolescents with CP[78] prescribed land based exercise in which 30% (n=6) of the sample did not complete the intervention.[78] The reasons for this are unclear and enjoyment of participation was not reported. Participant and family comments from this study's exit interviews were overwhelmingly positive regarding the intervention, highlighting that participants desire

to continue this therapy approach, with statements encompassing all six of the 'F-words' of disability: function, family, fitness, fun, friends and future.[39]

Lack of reporting of exercise intensity has been cited as a concern in CP exercise interventions.[143] A novel component of this study was the collection of HR data over 1190 intervals, which provided precise information related to exercise intensity. Previous studies have reported that achieving HRs consistent with HIIT may be difficult during water immersion as exercising in water increases stroke volume and diminishes the contribution increased HR makes to meeting the cardiac output required during exercise.[145] Further, recording of HR during exercise in water is challenging as the immersion interferes with Bluetooth performance. In our study, ≥90% of exercise intervals were at high intensity. A number of factors are likely to have assisted in attainment of high exercise HR's. First, the majority of exercise stations did not involve full body immersion.[21] Second, participants had their exercise HRs projected onto a wall in bright colours to provide real-time feedback, which the participants found very motivating. Third, exercise technique was not the focus, allowing participants to focus on speed. Finally, of the 10 separate exercise stations, four were kept consistent in all sessions, with familiarisation of new exercises offered via video demonstrations. This approach reduced the time required for procedural learning,[141] allowing rapid progression to a focus on intensity. Use of a research assistant for live HR recording and monitor backup storage optimised data available.

4.15 Strengths and limitations

A strength of this study is that participant retention after randomisation was high. Other studies involving people with disability have reported poor tolerance to control group

allocation.[139, 140] Enjoyment and interest in both the novel assessments and interventions for this cohort possibly contributed to retention. Technical prowess in the intervention was also neither measured nor a focus of participation, which may have been a pleasurable and novel experience for children with CP in a therapeutic environment. This high retention rate (100%) and tolerance to randomisation suggests minimal inflation of the sample size is required to account for attrition in any future definitive trial. This study did not investigate the ability of community agencies to replicate A-HIIT for a therapy program and this may be a limitation.

As this was designed a priori as a pilot RCT, inferential statistics exploring within- or between-group differences were not conducted. Nevertheless, the coefficients presented in Table 4-7 represent the best estimate of the effect of this intervention and are likely to be of interest to clinicians. These results may also assist with the selection of responsive outcome measures to assess efficacy of this intervention in any future trials. Caution should be taken with interpretation of these data, as they were collected in a small sample with large variation between participants.

4.16 Conclusion

This A-HIIT protocol was delivered in a standardised, reproducible way that was feasible to measure and tolerated well in adolescents with GMFCS II CP. This study provides comprehensive information regarding the feasibility of undertaking a novel intervention for people with disability. Our data suggest modification of the protocol used for recruitment is required, but overall feasibility and tolerability of assessment and intervention were acceptable.

The aim of this study was to determine if the protocol in its current format was feasible to progress to an RCT without modification. This study did determine that A-HIIT in adolescents with CP with GMFCS level II is feasible using the current protocol with minor modification of the recruitment process. The uncertainty that remains on the efficacy of the intervention, highlights the need for a large definitive trial.

Chapter 5:
Summary, clinical implications and directions for future research

5 Summary, clinical implications and directions for future research

The studies in this thesis explored the practicality and feasibility of a novel exercise approach in water, primarily to improve aerobic capacity. Efficacy and design of this type of exercise training was first established in a systematic review of aquatic high intensity interval training (A-HIIT) studies in non-athletic populations. The results from this study were translated into an A-HIIT study in adolescents with cerebral palsy (CP). The studies were underpinned by the rationale that while the ability of some adolescents with CP to participate in a land-based high intensity interval training (HIIT) program had been described,[78] the feasibility of conducting HIIT in water with this population was unknown.

A pilot randomised controlled trial (RCT) was considered to be needed prior to undertaking a definitive RCT of this intervention due to the number of unknown factors likely to impact study fidelity.

5.1 Synthesis of findings

Prior to these studies, it was unknown whether exercise intensity could be successfully quantified and reported during an A-HIIT intervention in children with CP. Data synthesised during the systematic review and meta-analysis provided valuable insights into the effect of A-HIIT on aerobic performance, strength and body composition in a non-athletic population. In this review, data were reported on eight studies across 13 papers. Data from 377 participants (A-HIIT n = 212, control n = 165) were included. Participants were predominantly female (89%), with the mean age ranging from 21.7 to

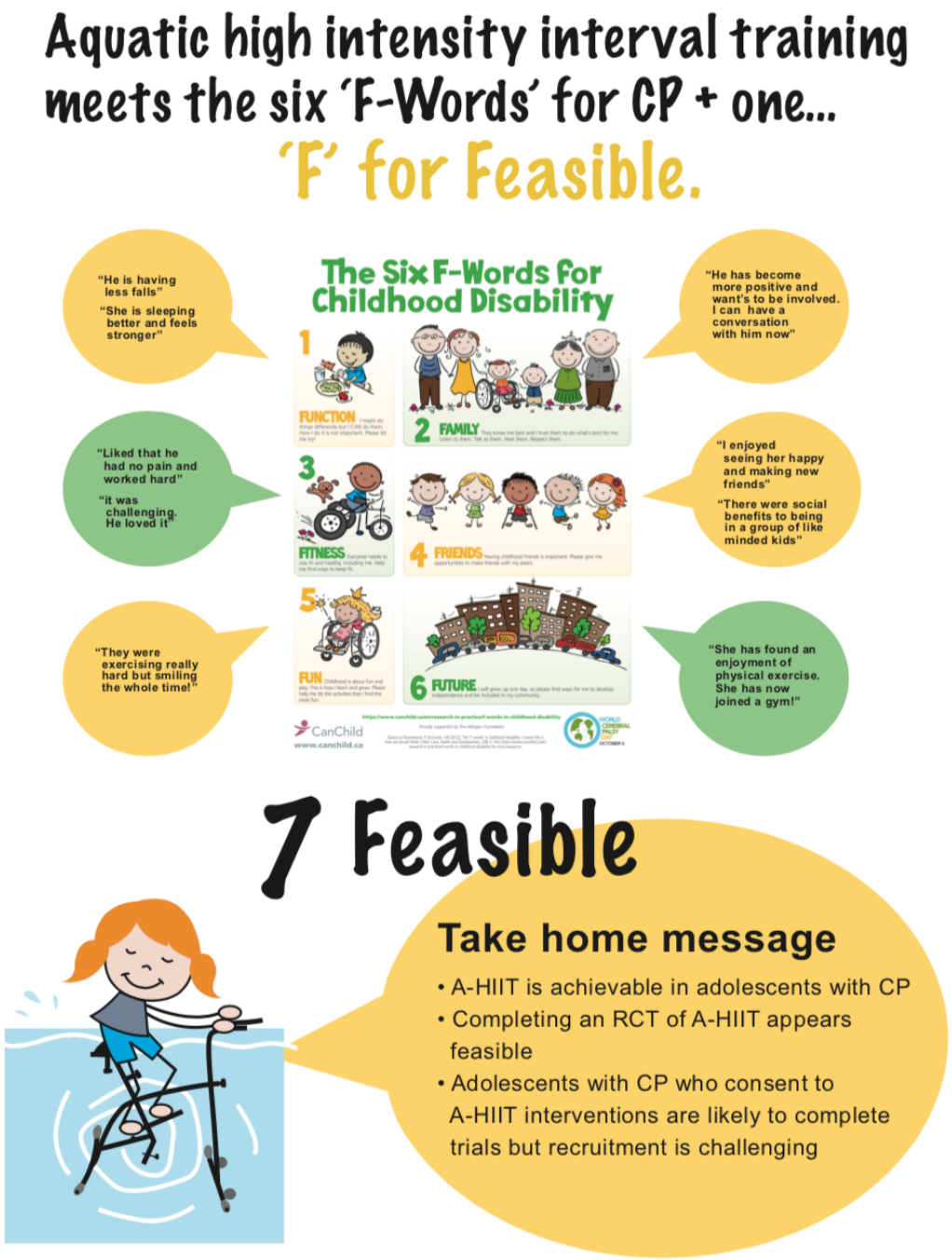
69.0 years for A-HIIT and 21.7 to 69.8 years for controls. Those who completed A-HIIT programs demonstrated greater aerobic performance (standardised mean difference [SMD] 0.69, 95% confidence interval [CI] 0.39 to 0.98; $I^2 = 0\%$; $n = 191$) and improved lower limb muscle strength (SMD 0.30, 95% CI 0.04 to 0.56; $I^2 = 0\%$; $n = 237$). No differences were seen in measures of body composition or adverse events. In a non-athletic population, A-HIIT was safe and improved aerobic performance and lower limb strength. The confidence intervals for these estimates were wide and therefore there is little precision around the estimate of effect size. The exercise intervention was well described, and monitoring and reporting of exercise intensity in water was feasible.

Trial design components related to exercise prescription, reporting of heart rate intensity, and factors that increased the risk of bias were identified using the Physiotherapy Evidence Database (PEDro) tool, the Cochrane risk of bias tool and the Consensus on Exercise Reporting Template (CERT). The (mean ± standard deviation) PEDro and CERT scores were 4.9 ± 1.5 and 15.1 ± 2.1, respectively. Although quality of reporting the methodology was moderate, the description of exercise interventions scored highly. Exercise intensity was closely monitored and sessions were supervised, factors which may have contributed to better outcomes.

Several elements described in these earlier studies were considered in the design of the second A-HIIT study in adolescents with CP: the use of known tools such as the CERT to standardise exercise prescription; the use of video links, consumer engagement, visual heart rate projection and group exercise to engage participants; the blinding of outcome assessors; the use of exercise stations rather than swimming skill to enable accurate heart rate assessment; and the extensive use and testing of monitoring methods

to ensure intensity of work rate could be collected. These factors likely contributed to the success in achieving high intensity exercise (i.e. high intensity exercise was achieved in 93% of exercise intervals; 95% CI 92 to 95), near perfect collection of heart rate data (i.e. heart rate data were available for 1180 of the 1190 stations completed) and high retention rates (100%; n = 12) in the pilot RCT.

The monitoring of heart rate rather than use of self-description of work rate was an important methodological strength of the A-HIIT program. It is reported that one in two children with CP have an intellectual impairment and that self-description of work rate may be unreliable and unknown tasks difficult to complete.[141] Intellectual impairment in this clinical population was noted in the current study and influenced screening, enrolment and ability to randomise participants. However, those who were enrolled and capable of completing assessments were able to engage fully in the intervention. Moreover, feedback and high retention rates indicated enjoyment, with participant-expressed themes around normalisation of exercise and building of friendships rather than usual goals of therapy interventions. For therapeutic activity to be feasible in young people, it must not only be technically achievable but also be age appropriate, encouraging of friendships and fun. We highlight this concept across the six 'F-words' for childhood disability,[39] with a playful interpretation including 'F' for feasibility of A-HIIT programs in adolescents with CP (Figure 5-1).



A-HIIT = aquatic high intensity interval training; CP = cerebral palsy; RCT = randomised controlled trial

Figure 5-1: Modification of the F-words as applied to aquatic high intensity interval training exercise for adolescents with cerebral palsy

5.2 Significance of the study findings

Using the study protocol described for the pilot RCT, a definitive RCT to answer whether this exercise model is effective in improving aerobic capacity in adolescents with CP appears to be feasible. The well-developed and standardised exercise structure and reporting will enable the intervention it to be replicated across different locations by different investigators. This is likely to be necessary, together with expansion of eligibility criteria to include those with Gross Motor Function Classification System (GMFCS) level I CP (who can ambulate independently including negotiating uneven terrain, curbs and stairs), to meet required participant numbers required to have adequate power to detect a small change on VO_{2peak}. Although higher than reported values for GMFCS II, adolescents with GMFCS level I still have reduced exercise capacity compared their typically developing peers.[27] Their functional abilities would allow a smooth integration into the current program whilst maintaining the dynamic of the group exercise environment, and significantly increase a recruitment pool. Positives of this study are the high rate of retention, which may offset the need to inflate recruitment targets, and the extensive and clear reporting which will allow meta-analyses of data.

The findings from the studies in this thesis highlight a number of areas for future research that could be explored using the design of this A-HIIT program:

1) *Ventilatory response during exercise*. Exercise may not be limited by disability factors alone and maximisation of lung health in these children should be explored as a means of overcoming barriers to exercise participation.[146]

2) *Bone health.* Adolescents with CP are known to have lower bone density and be at risk of fractures compared to their typically developing peers. However, to date this has only been a focus in non-ambulant children. The children in our sample walked without aids but some individuals were demonstrated to be at risk for low bone density. In typically developing young people, interventions have demonstrated bone growth in children following targeted exercise programs.[147] As any improvements in bone health are best gained in the pre- and peri-pubertal stages of development, risk factors for bone health screening and the benefit of exercise programs in ambulant children with CP could be further explored.[148]

3) *Health-related quality of life.* Previous data have suggested quantitative values of the Pediatric Quality of Life Inventory (PedsQL™) could be used to determine children at risk of reduced health-related quality of life. Independent clinical populations have established data cut off points associated with this risk. Quantitative values of risk in each domain of the PedsQL™ have not been established for ambulant adolescents with CP. The children who participated in the A-HIIT study demonstrated positive trend changes in their PedsQL™ scoring that may be interesting to explore in future studies. In particular, the impact of exercise interventions on lessening risk of reduced health-related quality of life in this population.[149]

4) *Enjoyment and participation methods.* Adolescents with CP have reduced participation in physical activity compared to their peers. Further qualitative assessment of the value participants placed on the intervention may assist future

design of aerobic exercise programs in this group, with potentially better translation to ongoing physical activity.[26]

5) *Pain and the influence of the aquatic environment.* In this group of adolescents with CP, pain was not further exacerbated by participation in the A-HIIT program. Given pain is a significant barrier to ongoing physical activity, further exploration the effects this intervention has on pain and the long-term adherence to such a program would be valuable.[69]

5.3 Conclusion

There is increasing interest in describing aerobic capacity in young people with CP due to the emergence of evidence that aerobic fitness is poorer in this group than in their typically developing peers which may have life-long consequences for health-related quality of life.[76] This thesis provides clear evidence of the feasibility of an A-HIIT intervention aimed at improving cardiorespiratory health for young people with CP (GMFCS level II). The findings support the application for funding to conduct a multi-centre RCT which is powered to detect change in target outcomes. Further, the design of this A-HIIT program may raise potential opportunity for other health related domains across the World Health Organisation's International Classification of Functioning, Disability and Health[38] to be explored.

References

6 References

1. Lee I-M, Shiroma EJ, Lobelo F, et al. Effect of physical inactivity on major non-communicable diseases worldwide: an analysis of burden of disease and life expectancy. *The Lancet* 2012; 380: 219-229.
2. Ruiz JR, Cavero-Redondo I, Ortega FB, et al. Cardiorespiratory fitness cut points to avoid cardiovascular disease risk in children and adolescents; what level of fitness should raise a red flag? A systematic review and meta-analysis. *British Journal of Sports Medicine* 2016; 50: 1451-1458.
3. World Health Organization. *Global status report on noncommunicable diseases 2014. Geneva:*. Geneva: World Health Organization, 2014.
4. Ekelund U, Ward HA, Norat T, et al. Physical activity and all-cause mortality across levels of overall and abdominal adiposity in European men and women: the European Prospective Investigation into Cancer and Nutrition Study (EPIC). *The American Journal of Clinical Nutrition* 2015; 101: 613-621.
5. Thorpe O, Johnston K and Kumar S. Barriers and enablers to physical activity participation in patients with COPD: a systematic review. *Journal of Cardiopulmonary Rehabilitation and Prevention* 2012; 32: 359-369.
6. Wilcox S, Der Ananian C, Abbott J, et al. Perceived exercise barriers, enablers, and benefits among exercising and nonexercising adults with arthritis: results from a qualitative study. *Arthritis care & research* 2006; 55: 616-627.
7. Sawyer A, Ucci M, Jones R, et al. Supportive environments for physical activity in deprived communities in the United Kingdom: a qualitative study using photo elicitation. *Social Science & Medicine* 2018; 197: 49-58.
8. Tucker P and Gilliland J. The effect of season and weather on physical activity: a systematic review. *Public Health* 2007; 121: 909-922.
9. Burgomaster KA, Hughes SC, Heigenhauser GJ, et al. Six sessions of sprint interval training increases muscle oxidative

potential and cycle endurance capacity in humans. *Journal of Applied Physiology* 2005; 98: 1985-1990.
10. Gibala MJ, Little JP, van Essen M, et al. Short-term sprint interval versus traditional endurance training: similar initial adaptations in human skeletal muscle and exercise performance. *The Journal of Physiology* 2006; 575: 901-911.
11. Vogiatzis I, Nanas S and Roussos C. Interval training as an alternative modality to continuous exercise in patients with COPD. *European Respiratory Journal* 2002; 20: 12-19.
12. Bartlett JD, Close GL, MacLaren DP, et al. High-intensity interval running is perceived to be more enjoyable than moderate-intensity continuous exercise: implications for exercise adherence. *Journal of Sports Sciences* 2011; 29: 547-553.
13. Martinez N, Kilpatrick MW, Salomon K, et al. Affective and enjoyment responses to high-intensity interval training in overweight-to-obese and insufficiently active adults. *Journal of Sport and Exercise Psychology* 2015; 37: 138-149.
14. Thum JS, Parsons G, Whittle T, et al. High-intensity interval training elicits higher enjoyment than moderate intensity continuous exercise. *PLoS One* 2017; 12: e0166299.
15. Güeita-Rodríguez J, García-Muro F, Cano-Díez B, et al. Identification of intervention categories for aquatic physical therapy in pediatrics using the International Classification of Functioning, Disability and Health - Children and Youth: a global expert survey. *Brazilian Journal of Physical Therapy* 2017; 21: 287-295.
16. McNamara RJ, McKeough ZJ, McKenzie DK, et al. Water-based exercise training for chronic obstructive pulmonary disease. *Cochrane Database of Systematic Reviews* 2013: CD008290.
17. Strike K, Mulder K and Michael R. Exercise for haemophilia. *Cochrane Database of Systematic Reviews* 2016; 12: Cd011180. 2016/12/20.
18. Barker AL, Talevski J, Morello RT, et al. Effectiveness of aquatic exercise for musculoskeletal conditions: a meta-analysis. *Archives of Physical Medicine and Rehabilitation* 2014; 95: 1776-1786.

19. McNamara RJ, McKeough ZJ, McKenzie DK, et al. Water-based exercise in COPD with physical comorbidities: a randomised controlled trial. *European Respiratory Journal* 2013; 41: 1284-1291. 2012/09/22.
20. Adsett JA, Mudge AM, Morris N, et al. Aquatic exercise training and stable heart failure: a systematic review and meta-analysis. *International Journal of Cardiology* 2015; 186: 22-28.
21. Nagle EF, Sanders ME and Franklin BA. Aquatic high intensity interval training for cardiometabolic health: benefits and training design. *American Journal of Lifestyle Medicine* 2017; 11: 64-76.
22. Kelly M and Darrah J. Aquatic exercise for children with cerebral palsy. *Developmental Medicine & Child Neurology* 2005; 47: 838-842.
23. Nagle EF, Robertson RJ, Jakicic JJ, et al. Effects of a combined aquatic exercise and walking in sedentary obese females undergoing a behavioral weight-loss intervention. *International Journal of Aquatic Research and Education* 2007; 1: Article 5.
24. Blair E and Cans C. The definition of cerebral palsy. *Cerebral Palsy*. Springer, 2018, pp.13-17.
25. Novak I. Evidence-based diagnosis, health care, and rehabilitation for children with cerebral palsy. *Journal of Child Neurology* 2014; 29: 1141-1156.
26. Verschuren O, Wiart L, Hermans D, et al. Identification of facilitators and barriers to physical activity in children and adolescents with cerebral palsy. *The Journal of Pediatrics* 2012; 161: 488-494.
27. Balemans AC, van Wely L, De Heer SJ, et al. Maximal aerobic and anaerobic exercise responses in children with cerebral palsy. *Medicine and Science in Sports and Exercise* 2013; 45: 561-568.
28. Ryan JM, Hensey O, McLoughlin B, et al. Associations of sedentary behaviour, physical activity, blood pressure and anthropometric measures with cardiorespiratory fitness in children with cerebral palsy. *PLoS One* 2015; 10: e0123267.

29. Peterson MD, Ryan JM, Hurvitz EA, et al. Chronic conditions in adults with cerebral palsy. *JAMA* 2015; 314: 2303-2305.
30. Parkinson KN, Dickinson HO, Arnaud C, et al. Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study. *Archives of Disease in Childhood* 2013; 98: 434-440.
31. Depiazzi JE, Forbes RA, Gibson N, et al. The effect of aquatic high-intensity interval training on aerobic performance, strength and body composition in a non-athletic population: systematic review and meta-analysis. *Clinical Rehabilitation* 2019; 33: 157-170.
32. Eldridge SM, Lancaster GA, Campbell MJ, et al. Defining feasibility and pilot studies in preparation for randomised controlled trials: development of a conceptual framework. *PLoS One* 2016; 11: e0150205.
33. Leon AC, Davis LL and Kraemer HC. The role and interpretation of pilot studies in clinical research. *Journal of Psychiatric Research* 2011; 45: 626-629.
34. Blair E, Langdon K, McIntyre S, et al. Survival and mortality in cerebral palsy: observations to the sixth decade from a data linkage study of a total population register and National Death Index. *BMC Neurology* 2019; 19: 111.
35. Blackmore AM, Bear N, Blair E, et al. Prevalence of symptoms associated with respiratory illness in children and young people with cerebral palsy. *Developmental Medicine & Child Neurology* 2016; 58: 780-781.
36. Strauss D, Cable W and Shavelle R. Causes of excess mortality in cerebral palsy. *Developmental Medicine & Child Neurology* 1999; 41: 580-585.
37. Verschuren O, Peterson MD, Balemans AC, et al. Exercise and physical activity recommendations for people with cerebral palsy. *Developmental Medicine and Child Neurology* 2016; 58: 798-808.
38. Stucki G. International Classification of Functioning, Disability, and Health (ICF): a promising framework and

classification for rehabilitation medicine. *American Journal of Physical Medicine & Rehabilitation* 2005; 84: 733-740.
39. Rosenbaum P and Gorter J. The 'F-words' in childhood disability: I swear this is how we should think! *Child: Care, Health and Development* 2012; 38: 457-463.
40. Xie B, Yan X, Cai X, et al. Effects of high-intensity interval training on aerobic capacity in cardiac patients: a systematic review with meta-analysis. *BioMed Research International* 2017: 5420840.
41. Saquetto MB, da Silva CM, Martinez BP, et al. Water-based exercise on functioning and quality of life in poststroke persons: a systematic review and meta-analysis. *Journal of Stroke and Cerebrovascular Diseases* 2019; 28: 104341.
42. Kamioka H, Nobuoka S and Iiyama J. Overview of systematic reviews with meta-analysis based on randomized controlled trials of balneotherapy and spa therapy from 2000 to 2019. *International Journal of General Medicine* 2020; 13: 429-442.
43. Costigan SA, Eather N, Plotnikoff RC, et al. High-intensity interval training for improving health-related fitness in adolescents: a systematic review and meta-analysis. *British Journal of Sports Medicine* 2015; 49: 1253-1261.
44. Bressel E, Wing JE, Miller AI, et al. High-intensity interval training on an aquatic treadmill in adults with osteoarthritis: effect on pain, balance, function, and mobility. *Journal of Strength & Conditioning Research* 2014; 28: 2088-2096.
45. Samadi Z, Bambaeichi E, Valiani M, et al. Evaluation of changes in levels of hyperandrogenism, hirsutism and menstrual regulation after a period of aquatic high intensity interval training in women with polycystic ovary syndrome. *International Journal of Preventive Medicine* 2019; 10: 187.
46. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. *Developmental Medicine and Child Neurology. Supplement* 2007; 109: 8-14.
47. Blair E and Watson L. Epidemiology of cerebral palsy. *Seminars in Fetal and Neonatal Medicine* 2006; 11: 117-125.

48. Palisano R, Rosenbaum P, Bartlett D, et al. *Gross Motor Function Classification System Expanded and Revised (GMFCS-E & R)*. CanChild Center for Childhood Disability Research, McMaster University, 2007.
49. Deloitte. *The cost of cerebral palsy in Australia in 2018. Report prepared for Cerebral Palsy Australia, Cerebral Palsy Alliance, and The Australasian Academy of Cerebral palsy and Developmental Medicine.* 2019. Canberra.
50. Centres for Disease Control and Prevention. Data and statstics for cerebral palsy, https://www.cdc.gov/ncbddd/cp/data.html (2019, accessed 10 November 2020).
51. Zwier JN, van Schie PE, Becher JG, et al. Physical activity in young children with cerebral palsy. *Disability and Rehabilitation* 2010; 32: 1501-1508.
52. Balemans AC, van Wely L, Becher JG, et al. Longitudinal relationship among physical fitness, walking-related physical activity, and fatigue in children with cerebral palsy. *Physical Therapy* 2015; 95: 996-1005.
53. Hoofwijk M, Unnithan V and Bar-Or O. Maximal treadmill performance of children with cerebral palsy. *Pediatric Exercise Science* 1995; 7: 305-313.
54. Maltais D, Pierrynowski M, Galea V, et al. Physical activity level is associated with the O2 cost of walking in cerebral palsy. *Medicine and Science in Sports and Exercise* 2005; 37: 347-353.
55. Rieckert H, Bruhm U and Schwalm U. Endurance training within a program of physical education in children predominantly with cerebral palsy. *Die Medizinische Welt* 1977; 28: 1694-1701.
56. Unnithan VB, Clifford C and Bar-Or O. Evaluation by exercise testing of the child with cerebral palsy. *Sports Medicine* 1998; 26: 239-251.
57. Verschuren O and Takken T. Aerobic capacity in children and adolescents with cerebral palsy. *Research in Developmental Disabilities* 2010; 31: 1352-1357.
58. Verschuren O, Ketelaar M, Gorter JW, et al. Exercise training program in children and adolescents with cerebral palsy:

a randomized controlled trial. *Archives of Pediatrics and Adolescent Medicine* 2007; 161: 1075-1081.
59. Unnithan VB, Katsimanis G, Evangelinou C, et al. Effect of strength and aerobic training in children with cerebral palsy. *Medicine and Science in Sports and Exercise* 2007; 39: 1902-1909.
60. Williams SA, McFadden LM, Blackmore A, et al. Do adolescents with cerebral palsy meet recommendations for healthy weight and physical activity behaviours? *Disability and Rehabilitation* 2020; 42: 1227-1232.
61. Buffart LM, Westendorp T, Van Den Berg-Emons RJ, et al. Perceived barriers to and facilitators of physical activity in young adults with childhood-onset physical disabilities. *Journal of Rehabilitation Medicine* 2009; 41: 881-885.
62. Rimmer JH and Marques AC. Physical activity for people with disabilities. *The Lancet* 2012; 380: 193-195.
63. Lauruschkus K, Nordmark E and Hallström I. Parents' experiences of participation in physical activities for children with cerebral palsy–protecting and pushing towards independence. *Disability and Rehabilitation* 2017; 39: 771-778.
64. Beckung E and Hagberg G. Neuroimpairments, activity limitations, and participation restrictions in children with cerebral palsy. *Developmental Medicine and Child Neurology* 2002; 44: 309-316.
65. Schenk-Rootlieb A, Van Nieuwenhuizen O, Schiemanck N, et al. Impact of cerebral visual impairment on the everyday life of cerebral palsied children. *Child: Care, Health and Development* 1993; 19: 411-423.
66. Alriksson-Schmidt A and Hägglund G. Pain in children and adolescents with cerebral palsy: a population-based registry study. *Acta Paediatrica* 2016; 105: 665-670.
67. Novak I, Hines M, Goldsmith S, et al. Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics* 2012; 130: e1285-e1312.
68. Eriksson E, Hägglund G and Alriksson-Schmidt AI. Pain in children and adolescents with cerebral palsy–a cross-sectional register study of 3545 individuals. *BMC Neurology* 2020; 20: 15.

69. Ramstad K. *Recurrent musculoskeletal pain in paediatric cerebral palsy: relations to mental health, health-related quality of life and participation*. Doctoral thesis, University of Oslo, Norway, 2012.
70. Verschuren O, Ketelaar M, Keefer D, et al. Identification of a core set of exercise tests for children and adolescents with cerebral palsy: a Delphi survey of researchers and clinicians. *Developmental Medicine and Child Neurology* 2011; 53: 449-456.
71. Slaman J, Roebroeck M, van der Slot W, et al. Can a lifestyle intervention improve physical fitness in adolescents and young adults with spastic cerebral palsy? A randomized controlled trial. *Archives of Physical Medicine and Rehabilitation* 2014; 95: 1646-1655.
72. Nsenga A, Shephard R and Ahmaidi S. Aerobic training in children with cerebral palsy. *International Journal of Sports Medicine* 2013; 34: 533-537.
73. Garber CE, Blissmer B, Deschenes MR, et al. American College of Sports Medicine position stand. Quantity and quality of exercise for developing and maintaining cardiorespiratory, musculoskeletal, and neuromotor fitness in apparently healthy adults: guidance for prescribing exercise. *Medicine and Science in Sports and Exercise* 2011; 43: 1334-1359.
74. Forbregd TR, Aloyseus MA, Berg A, et al. Cardiopulmonary capacity in children during exercise testing; the differences between treadmill, and upright and supine cycle ergometry. *Frontiers in Physiology* 2019; 10: 1440.
75. Mitropoulos A, Gumber A, Crank H, et al. Validation of an arm crank ergometer test for use in sedentary adults. *Journal of Sports Science and Medicine* 2017; 16: 558-564.
76. Balemans AC and Bolster EA. *Aerobic and anaerobic fitness in children and youth with cerebral palsy*. Cham, Switzerland: Springer, 2019.
77. van Wely L, Balemans AC, Becher JG, et al. Physical activity stimulation program for children with cerebral palsy did not improve physical activity: a randomised trial. *Journal of Physiotherapy* 2014; 60: 40-49.

78. Lauglo R, Vik T, Lamvik T, et al. High-intensity interval training to improve fitness in children with cerebral palsy. *BMJ Open Sport and Exercise Medicine* 2016; 2: e000111.
79. Engel FA, Ackermann A, Chtourou H, et al. High-intensity interval training performed by young athletes: a systematic review and meta-analysis. *Frontiers in Physiology* 2018; 9: 1012.
80. Cao M, Quan M and Zhuang J. Effect of high-intensity interval training versus moderate-intensity continuous training on cardiorespiratory fitness in children and adolescents: a meta-analysis. *International Journal of Environmental Research and Public Health* 2019; 16: 1533.
81. Thabane L, Ma J, Chu R, et al. A tutorial on pilot studies: the what, why and how. *BMC Medical Research Methodology* 2010; 10: 1.
82. Lancaster GA, Dodd S and Williamson PR. Design and analysis of pilot studies: recommendations for good practice. *Journal of Evaluation in Clinical Practice* 2004; 10: 307-312.
83. Arain M, Campbell MJ, Cooper CL, et al. What is a pilot or feasibility study? A review of current practice and editorial policy. *BMC Medical Research Methodology* 2010; 10: 67.
84. Kaur N, Figueiredo S, Bouchard V, et al. Where have all the pilot studies gone? A follow-up on 30 years of pilot studies in *Clinical Rehabilitation*. *Clinical Rehabilitation* 2017; 31: 1238-1248.
85. Kannan S and Gowri S. Pilot studies: are they appropriately reported? *Perspectives in Clinical Research* 2015; 6: 207-210.
86. Shanyinde M, Pickering RM and Weatherall M. Questions asked and answered in pilot and feasibility randomized controlled trials. *BMC Medical Research Methodology* 2011; 11: 117.
87. Malmqvist J, Hellberg K, Möllås G, et al. Conducting the pilot study: a neglected part of the research process? Methodological findings supporting the importance of piloting in qualitative research studies. *International Journal of Qualitative Methods* 2019; 18: 1609406919878341.
88. Lancaster G. Pilot and feasibility studies come of age! *Pilot and Feasibility Studies* 2015; 1: 1-4.

89. Eldridge SM, Chan CL, Campbell MJ, et al. CONSORT 2010 statement: extension to randomised pilot and feasibility trials. *BMJ* 2016; 355: i5239.
90. Turner L, Moher D, Shamseer L, et al. The influence of CONSORT on the quality of reporting of randomised controlled trials: an updated review. *Trials* 2011; 12: A47.
91. Liberati A, Altman DG, Tetzlaff J, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. *Journal of Clinical Epidemiology* 2009; 62: e1-e34.
92. Borg GA. Psychophysical bases of perceived exertion. *Medicine and Science in Sports and Exercise* 1982; 14: 377-381.
93. Higgins JPT, Altman DG, Gøtzsche PC, et al. The Cochrane Collaboration's tool for assessing risk of bias in randomised trials. *BMJ* 2011; 343. DOI: 10.1136/bmj.d5928.
94. Sterne JA, Sutton AJ, Ioannidis JP, et al. Recommendations for examining and interpreting funnel plot asymmetry in meta-analyses of randomised controlled trials. *BMJ* 2011; 343: d4002.
95. de Morton NA. The PEDro scale is a valid measure of the methodological quality of clinical trials: a demographic study. *Australian Journal of Physiotherapy* 2009; 55: 129-133.
96. Slade SC, Dionne CE, Underwood M, et al. Consensus on exercise reporting template (CERT): explanation and elaboration statement. *British Journal of Sports Medicine* 2016; 50: 1428-1437.
97. Cohen J. *Statistical power analysis for the behavioral sciences*. 2nd ed. Mahwah, NJ: Lawrance Erlbaum, 2013.
98. Bento PC and Rodacki AL. Muscle function in aged women in response to a water-based exercises program and progressive resistance training. *Geriatrics & Gerontology International* 2015; 15: 1193-1200. 2014/12/17.
99. Broman G, Quintana M, Lindberg T, et al. High intensity deep water training can improve aerobic power in elderly women. *European Journal of Applied Physiology* 2006; 98: 117-123.
100. Connolly LJ, Nordsborg NB, Nyberg M, et al. Low-volume high-intensity swim training is superior to high-volume low-intensity training in relation to insulin sensitivity and glucose

control in inactive middle-aged women. *European Journal of Applied Physiology* 2016; 116: 1889-1897.
101. Hamer PW and Morton AR. Water-running : training effects and specificity of aerobic anaerobic and muscular parameters following an eight-week interval training programme. *Australian Journal of Science & Medicine in Sport* 1990; 21: 13-22.
102. Munukka M, Waller B, Rantalainen T, et al. Efficacy of progressive aquatic resistance training for tibiofemoral cartilage in postmenopausal women with mild knee osteoarthritis: a randomised controlled trial. *Osteoarthritis and Cartilage* 2016; 24: 1708-1717.
103. Michaud TJ, Brennan DK, Wilder RP, et al. Aquarunning and gains in cardiorespiratory fitness. *The Journal of Strength & Conditioning Research* 1995; 9: 78-84.
104. Mohr M, Nordsborg NB, Lindenskov A, et al. High-intensity intermittent swimming improves cardiovascular health status for women with mild hypertension. *BioMed Research International* 2014; 2014
105. Mohr M, Helge EW, Petersen LF, et al. Effects of soccer vs swim training on bone formation in sedentary middle-aged women. *European Journal of Applied Physiology* 2015; 115: 2671-2679.
106. Moreira L, Fronza FC, dos Santos RN, et al. High-intensity aquatic exercises (HydrOS) improve physical function and reduce falls among postmenopausal women. *Menopause* 2013; 20: 1012-1019.
107. Moreira LD, Fronza FC, dos Santos RN, et al. The benefits of a high-intensity aquatic exercise program (HydrOS) for bone metabolism and bone mass of postmenopausal women. *Journal of Bone and Mineral Metabolism* 2014; 32: 411-419.
108. Nordsborg NB, Connolly L, Weihe P, et al. Oxidative capacity and glycogen content increase more in arm than leg muscle in sedentary women after intense training. *Journal of Applied Physiology* 2015; 119: 116-123.
109. Rebold M, Kobak M and Otterstetter R. The influence of a Tabata interval training program using an aquatic underwater

treadmill on various performance variables. *Journal of Strength & Conditioning Research* 2013; 27: 3419-3425.
110. Waller B, Munukka M, Rantalainen T, et al. Effects of high intensity resistance aquatic training on body composition and walking speed in women with mild knee osteoarthritis: a 4-month RCT with 12-month follow-up. *Osteoarthritis Cartilage* 2017; 25: 1238-1246.
111. Borg GA. Psychophysical bases of perceived exertion. *Med Sci Sports Exerc* 1982; 14: 377-381.
112. Astorino TA, Allen RP, Roberson DW, et al. Adaptations to high-intensity training are independent of gender. *European Journal of Applied Physiology* 2011; 111: 1279-1286.
113. Metcalfe RS, Tardif N, Thompson D, et al. Changes in aerobic capacity and glycaemic control in response to reduced-exertion high-intensity interval training (REHIT) are not different between sedentary men and women. *Applied Physiology, Nutrition, and Metabolism* 2016; 41: 1117-1123.
114. Lacroix A, Hortobagyi T, Beurskens R, et al. Effects of supervised vs unsupervised training programs on balance and muscle strength in older adults: a systematic review and meta-analysis. *Sports Medicine* 2017; 47: 2341-2361.
115. Batacan RB, Duncan MJ, Dalbo VJ, et al. Effects of high-intensity interval training on cardiometabolic health: a systematic review and meta-analysis of intervention studies. *British Journal of Sports Medicine* 2017; 51: 494-503.
116. Weston KS, Wisloff U and Coombes JS. High-intensity interval training in patients with lifestyle-induced cardiometabolic disease: a systematic review and meta-analysis. *British Journal of Sports Medicine* 2014; 48: 1227-1234.
117. Milanović Z, Sporiš G and Weston M. Effectiveness of high-intensity interval training (HIT) and continuous endurance training for VO 2max improvements: a systematic review and meta-analysis of controlled trials. *Sports Medicine* 2015; 45: 1469-1481.
118. Heywood S, McClelland J, Mentiplay B, et al. Effectiveness of aquatic exercise in improving lower limb strength in musculoskeletal conditions: a systematic review and meta-

analysis. *Archives of Physical Medicine and Rehabilitation* 2017; 98: 173-186.
119. Prado AK, Reichert T, Conceição MO, et al. Effects of aquatic exercise on muscle strength in young and elderly adults: a systematic review and meta-analysis of randomized trials. *Journal of Strength and Conditioning Research* 2016; August.
120. Boutcher SH. High-intensity intermittent exercise and fat loss. *Journal of Obesity* 2011; 2011: 868305.
121. Maillard F, Pereira B and Boisseau N. Effect of high-intensity interval training on total, abdominal and visceral fat mass: a meta-analysis. *Sports Medicine* 2018; 48: 269-288.
122. Irandoust K and Taheri M. The effects of aquatic exercise on body composition and nonspecific low back pain in elderly males. *Journal of Physical Therapy Science* 2015; 27: 433-435.
123. Robertson RJ, Goss FL, ANDREACCI JL, et al. Validation of the Children's OMNI-Resistance Exercise Scale of perceived exertion. *Medicine & Science in Sports & Exercise* 2005; 37: 819-826.
124. American Thoracic Society. ATS/ACCP statement on cardiopulmonary exercise testing. *American Journal of Respiratory and Critical Care Medicine* 2003; 167: 211-277.
125. Beauchamp MK, Nonoyama M, Goldstein RS, et al. Interval versus continuous training in individuals with chronic obstructive pulmonary disease - a systematic review. *Thorax* 2010; 65: 157-164.
126. Garra G, Singer AJ, Taira BR, et al. Validation of the Wong-Baker FACES pain rating scale in pediatric emergency department patients. *Academic Emergency Medicine* 2010; 17: 50-54.
127. Bongers BC, Hulzebos EH, Van Brussel M, et al. Pediatric norms for cardiopulmonary exercise testing. *Uitgeverij BOXPress, s'-Hertogenbosch* 2012; 2: 30-34.
128. Pasco JA, Nicholson GC and Kotowicz MA. Cohort profile: Geelong osteoporosis study. *International Journal of Epidemiology* 2012; 41: 1565-1575.

129. Kingsnorth S, Orava T, Provvidenza C, et al. Chronic pain assessment tools for cerebral palsy: a systematic review. *Pediatrics* 2015; 136: e947-e960.
130. Varni JW, Seid M and Rode CA. The PedsQL™: measurement model for the Pediatric Quality of Life Inventory. *Medical Care* 1999: 126-139.
131. Law M, Baptiste S, Carswell A, et al. *Canadian Occupational Performance Measure (COPM)*. 5th ed. Ottawa: Canadian Association of Occupational Therapists (CAOT), 2014.
132. Varni JW, Burwinkle TM, Berrin SJ, et al. The PedsQL in pediatric cerebral palsy: reliability, validity, and sensitivity of the Generic Core Scales and Cerebral Palsy Module. *Developmental Medicine & Child Neurology* 2006; 48: 442-449.
133. Varni JW, Seid M and Kurtin PS. PedsQL™ 4.0: reliability and validity of the Pediatric Quality of Life Inventory™ Version 4.0 Generic Core Scales in healthy and patient populations. *Medical Care* 2001; 39: 800-812.
134. Atkinson TM, Rosenfeld BD, Sit L, et al. Using confirmatory factor analysis to evaluate construct validity of the Brief Pain Inventory (BPI). *Journal of Pain and Symptom Management* 2011; 41: 558-565.
135. Jelsness-Jorgensen LP, Moum B, Grimstad T, et al. Validity, reliability, and responsiveness of the Brief Pain Inventory in inflammatory bowel disease. *Canadian Journal of Gastroenterology and Hepatology* 2016; 2016: 5624261.
136. Eyssen I, Steultjens M, Oud T, et al. Responsiveness of the Canadian Occupational Performance Measure. *Journal of Rehabilitation Research and Development* 2011; 48: 517-528.
137. McColl MA, Paterson M, Davies D, et al. Validity and community utility of the Canadian Occupational Performance Measure. *Canadian Journal of Occupational Therapy* 2000; 67: 22-30.
138. Cocks K and Torgerson DJ. Sample size calculations for pilot randomized trials: a confidence interval approach. *Journal of Clinical Epidemiology* 2013; 66: 197-201.
139. Hind D, Parkin J, Whitworth V, et al. Aquatic therapy for boys with Duchenne muscular dystrophy (DMD): an external

pilot randomised controlled trial. *Pilot and Feasibility Studies* 2017; 3: 16.
140. Beckers L, Rameckers E, Smeets R, et al. Barriers to recruitment of children with cerebral palsy in a trial of home-based training. *Contemporary Clinical Trials Communications* 2019; 15: 100371.
141. Gofer-Levi M, Silberg T, Brezner A, et al. Cognitive procedural learning among children and adolescents with or without spastic cerebral palsy: the differential effect of age. *Research in Developmental Disabilities* 2014; 35: 1952-1962.
142. Brehm M-A, Balemans AC, Becher JG, et al. Reliability of a progressive maximal cycle ergometer test to assess peak oxygen uptake in children with mild to moderate cerebral palsy. *Physical Therapy* 2014; 94: 121-128.
143. Ryan JM, Cassidy EE, Noorduyn SG, et al. Exercise interventions for cerebral palsy. *Cochrane Database of Systematic Reviews* 2017; (6): CD011660.
144. Little JP, Safdar A, Wilkin GP, et al. A practical model of low-volume high-intensity interval training induces mitochondrial biogenesis in human skeletal muscle: potential mechanisms. *Journal of Physiology* 2010; 588: 1011-1022.
145. Svedenhag J and Seger J. Running on land and in water: comparative exercise physiology. *Medicine and Science in Sports and Exercise* 1992; 24: 1155-1160.
146. Marpole R, Blackmore AM, Gibson N, et al. Evaluation and management of respiratory illness in children with cerebral palsy. *Frontiers in Pediatrics* 2020; 8.
147. Vicente-Rodríguez G. How does exercise affect bone development during growth? *Sports Medicine* 2006; 36: 561-569.
148. Houlihan CM. Bone health in cerebral palsy: who's at risk and what to do about it? *Journal of Pediatric Rehabilitation Medicine* 2014; 7: 143-153.
149. Huang I-C, Thompson LA, Chi Y-Y, et al. The linkage between pediatric quality of life and health conditions: establishing clinically meaningful cutoff scores for the PedsQL. *Value in Health* 2009; 12: 773-781.

Appendices

7 Appendices

Appendix 1: Example of search strategy

Search Details

Query Translation:

```
(("high-intensity interval training"[MeSH Terms] OR ("high-
intensity"[All Fields] AND "interval"[All Fields] AND
"training"[All Fields]) OR "high-intensity interval
training"[All Fields] OR ("high"[All Fields] AND
"intensity"[All Fields] AND "interval"[All Fields] AND
"training"[All Fields]) OR "high intensity interval
training"[All Fields]) OR (interval[All Fields] AND
("education"[Subheading] OR "education"[All Fields] OR
"training"[All Fields] OR "education"[MeSH Terms] OR
"training"[All Fields])) OR (high[All Fields] AND
```

Search URL

Result:

1801

Translations:

high intensity interval training	"high-intensity interval training"[MeSH Terms] OR ("high-intensity"[All Fields] AND "interval"[All Fields] AND "training"[All Fields]) OR "high-intensity interval training"[All Fields] OR ("high"[All Fields] AND "intensity"[All Fields] AND "interval"[All Fields] AND "training"[All Fields]) OR "high intensity interval training"[All Fields]
training	"education"[Subheading] OR "education"[All Fields] OR "training"[All Fields] OR "education"[MeSH Terms] OR "training"[All Fields]
exercise	"exercise"[MeSH Terms] OR "exercise"[All Fields]
water	"water"[MeSH Terms] OR "water"[All Fields] OR "drinking water"[MeSH Terms] OR ("drinking"[All Fields] AND "water"[All Fields]) OR "drinking water"[All Fields]
hydrotherapy	"hydrotherapy"[MeSH Terms] OR "hydrotherapy"[All Fields]
immersion	"immersion"[MeSH Terms] OR "immersion"[All Fields]
head	"head"[MeSH Terms] OR "head"[All Fields]
swim	"swimming"[MeSH Terms] OR "swimming"[All Fields] OR "swim"[All Fields]

Database:

PubMed

User query:

((high intensity interval training OR interval training OR high intensity OR intermittent exercise OR plyometric)) AND (aquatic exercise OR water exercise OR hydrotherapy OR immersion OR head out aquatic OR water run OR swim)

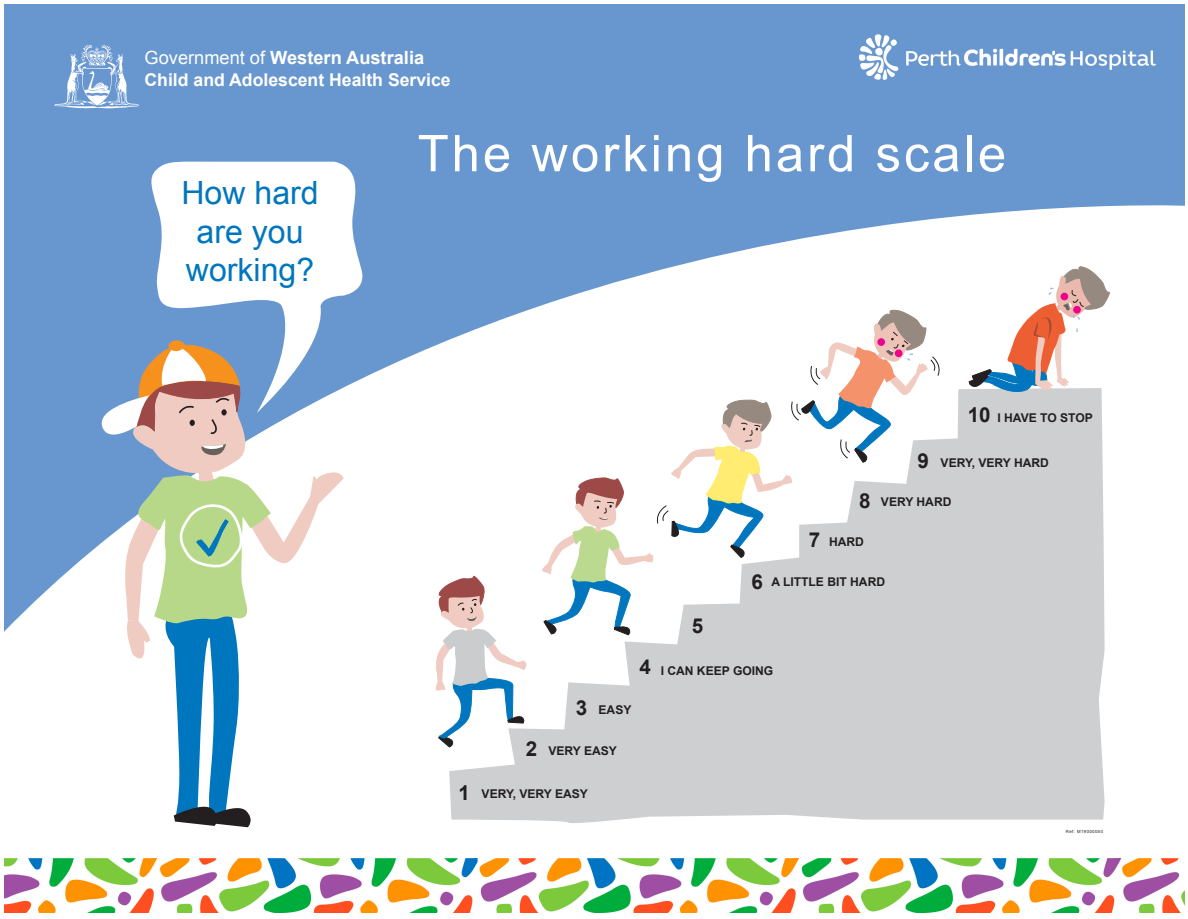
Appendix 2: Proforma Consensus on Exercise Reporting Template (CERT) assessment form[96]

Author and year
Title
Journal
Study location
Reviewer and date

Item	Description	Data extraction—details	Location (pg, URL etc)		YES: 1 NO: 0	'Reasons for rating': eg, 'not reported or not clearly described'
			Primary paper	Other*		
1	Detailed description of the type of exercise equipment		Eg, page, column, paragraph			
2	Detailed description of the qualifications, expertise and/or training					
3	Describe whether exercises are performed individually or in a group					
4	Describe whether exercises are supervised or unsupervised; how they are delivered					
5	Detailed description of how adherence to exercise is measured and reported					
6	Detailed description of motivation strategies					
7a	Detailed description of the decision rule(s) for determining exercise progression					
7b	Detailed description of how the exercise program was progressed					
8	Detailed description of each exercise to enable replication					
9	Detailed description of any home programme component					
10	Describe whether there are any non-exercise components					
11	Describe the type and number of adverse events that occur during exercise					
12	Describe the setting in which the exercises are performed					
13	Detailed description of the exercise intervention					
14a	Describe whether the exercises are generic (one size fits all) or tailored					
14b	Detailed description of how exercises are tailored to the individual					
15	Describe the decision rule for determining the starting level					
16a	Describe how adherence or fidelity is assessed/measured					
16b	Describe the extent to which the intervention was delivered as planned					
Total score						

* eg, protocol paper, published reference papers, supplementary data, online appendices, websites.

Appendix 3: The 'working hard' scale



Appendix 4: Wong-Baker FACES Pain Rating Scale

Appendix 5: Pain description and severity score

Pain description pre and post intervention

The descriptive words used by a participant to describe pain during the intervention class or assessments – prompts to assist description include aching, cramping, fearful, gnawing, heavy, hot or burning, sharp and shooting.	Designated by number allocation	0 = no pain
		1 = aching
		2 = cramping
		3 = fearful
		4 = gnawing
		5 = heavy
		6 = hot or burning
		7 = sharp or shooting
		8 = other

Appendix 6: Data dictionary

Variable name	Description	Data type	Values/rules
Activity participation	The verbal description given by participants of their physical activity e.g. 1-hour bike ride.		
Adverse event	Any undesirable clinical occurrence in a participant whether it is considered to be study related or not, that includes a clinical sign, symptom or condition and/or an observation of an unintended outcome of the study.		
Aerobic capacity	A measure of the ability of the heart and lungs to get oxygen to the muscles which influences a child's ability to sustain a certain level of aerobic activity for a certain length of time.		
Age	Age in years and months of participant at time of baseline assessment.	Number	12 to 17 only
Assessment/ measure A	A measurement or data collection taken at pre-intervention assessment.		
Assessment/ measure B	A measurement or data collection taken at post-intervention assessments.		
Assessments	Refers to the collective group of measures a participant undertakes pre and post the intervention period.		
Cardiometabolic risk	Cardiometabolic risk refers to a high lifetime risk for cardiovascular disease (CVD).		
Completion fraction	Of those who agree to participate, the percentage of participants who complete all aspects of the study.	Percentage	0 to 100
Completion of baseline assessments	The individual participated in all assessments where an outcome measure was achieved.	Number	0 = withdrawn; 1 = completed; 2 = unable to complete

Variable name	Description	Data type	Values/rules
Completion of baseline assessments fraction	Of those who agree to participate, the percentage of participants who complete all pre-intervention baseline assessments. Completion is defined as participated in all assessments where an outcome measure was achieved.	Percentage	0 to 100
Completion of post-intervention assessments	The individual participated in all assessments within 2 weeks of completion of the intervention period where an outcome measure was achieved.	Number	0 = withdrawn; 1 = completed; 2 = unable to complete
Completion of post-intervention assessments fraction	Baseline assessments performed in the 2 weeks post intervention or control period. Completion is defined as the percentage of participants who complete all post-intervention assessments where an outcome measure was achieved.	Percentage	0 to 100
CERT	Consensus of Exercise Reporting Template.		
Continuous training	Any type of physical training that involves activity without rest intervals. Continuous training can be performed at low, moderate or high exercise intensities.		
Control group	Participants who complete baseline assessments and who are randomised to usual care and will not attend the 10-week water-based training.	Number	0 to 10
Cool-down	Supervised and standardised exercises in the water for 5 minutes to relax and reduce participant heart rate following interval training.		
COPM	Canadian Occupational Performance Measure – an evidence-based outcome measure designed to capture a client's self-perception of performance in everyday living, over time.	Number	
COPM completion	Where a participant has entered and scored at least one task in each of the three domains.	Number	0 = no; 1 = yes

Variable name	Description	Data type	Values/rules
CP	Cerebral palsy – describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non- progressive disturbances that occurred in the developing fetal or infant brain.		
CP GMFCS II	Gross Motor Functional Classification System of II (12–18 years) – youth walk in most settings but environmental factors and personal choice influence mobility choices. At school or work they may require a handheld mobility device for safety and climb stairs holding onto a railing. Outdoors and in the community youth may use wheeled mobility when travelling long distance.	Expanded and revised edition	
CPET	Cardiopulmonary exercise testing – tests performed using a maximal incremental cycle ergometer standardised instruction protocol, validated in ambulant adolescents with CP, using breath-by-breath analysis (Vmax 229 and SensorMedics Vmax encore – Software Version 21-1A).		
Demographic data	Statistical data collected about the characteristics of the population, including age, gender, educational year, geographical household location, number of siblings and sibling commitments.		
DXA	Dual-energy X-ray absorptiometry – where two X-ray beams, with different energy levels, are aimed at the patient's bones to measure bone mineral density (BMD) and body mass characteristics.		
Entered pool on time	Ready to participate in first high intensity interval (HII).	Number	0 = no; 1 = yes; 3 = entered during HII
Exclusion criteria	Characteristics that disqualify prospective subjects from inclusion in the study.		

Variable name	Description	Data type	Values/rules
Exercise class	The water-based training intervention classes.	Number	Classes numbered in chronological order offered to participants from 01 to 30, followed by day and month, e.g. 011504
Exercise classes attended	Number of aquatic high intensity interval classes attended in total.	Number	0 to 20
Gender	Male or female.	Number	0 = male; 1 = female
General activity question	A question/questions asked of participants to ascertain any recent or unusual activity, e.g. “have you played sport this week?”, “What did you do on the weekend?”		
General health question	A question/questions asked of participants to ascertain any recent or current illness or health concern, e.g. “Have you been well this week?”, “Do you have a cold at the moment?”		
Goal HR	Goal heart rate – the heart rate of a participant required to meet with guideline for high intensity exercise of ≥80% HRM. Absolute number, i.e. not rounded up.	Beats per minute	
Heart rate intensity colours	The colours used to describe heart rate zone achievement within an exercise class – maximum, hard, moderate, light and very light.	RED	80% and above
		YELLOW	75% to 79%
		GREEN	70% to 74%
		BLUE	60% to 69%

Variable name	Description	Data type	Values/rules
		GREY	30% to 59%
Heart rate maximum intensity	Heart rate achieved was 80% heart rate maximum or above, i.e. RED.	Percentage	80% and above
Heart rate moderate intensity	HR achieved was within 5% of a successful epoc, i.e. YELLOW 75–79% pHRM.	Percentage	75% to 79%
HR 80% iPad/10	Number of exercise stations out of 10 where heart rate as reflected on the iPad was recorded above 80%.	Number	0 to 10
HR 80% sensor graphic/10	Number of exercise stations out of 10 where heart rate was recorded in the 'red' zone (80% or above) in the Polar Club H10 heart rate sensor graphic.	Number	0 to 10
HR 80% beat/10	Number of exercise stations out of 10 where heart rate was recorded at 80% heart rate maximum or greater as reflected in beat-by-beat battery data in Polar Beat.	Number	0 to 10
HR max (beat)	The highest heart rate recorded in Polar Beat for an individual participant's class.	Beats per minute	0 to 240
HR average (club)	The average heart rate recorded in Polar Club for an individual participant's class.	Beats per minute	0 to 240
HR min (club)	The minimum HR recorded in Polar Club for an individual participant's class.	Beats per minute	0 to 240
HR data fidelity – participant	The number of intervals where heart rate data is available for a participant and success able to be determined within a class.	Percentage	0 to 100
HR data fidelity – class	The number of intervals where heart rate data is available compared to total number of intervals completed by all participants in a class.	Percentage	0 to 100

Variable name	Description	Data type	Values/rules
HRQoL	Health-related quality of life.		
HR telemetry	The technology method of establishing a participant's heart rate using a chest strap device, wrist watch and/or receiver.		
HR training	Exercise intensity determined by the goal of achieving a targeted heart rate.		
HRM	Heart rate maximum – the maximum heart rate in beats per minute achieved by a participant during a CPET that was deemed maximal by the exercise scientist.	Number	
HRM percentage	A heart rate expressed as a percentage of the maximum heart rate in beats per minute achieved by a participant during a CPET that was deemed maximal by the exercise scientist.		
Height	The height of the participant taken at baseline measurement.	Number	centimetres 0 decimals
HII	High intensity interval – a 60-second interval of exercise training aimed at achieving a peak heart rate ≥80% of participant's peak heart rate (measured during the CPET).		
HII success	The participant reaches their target heart rate during the 60-second interval or in the immediate rest period following.	Number	0 = participated but did not achieve target 1 = participated and achieved target 2 = did not participate 3 = heart rate data not available

Variable name	Description	Data type	Values/rules
HII success fraction	The combined intervention group total interval success compared to total number of intervals participated in by the intervention group over the study period.	Percentage	0 to 100
HII total (attended)	The total number of intervals the participant attended.	Number	0 to 200
HII total (attended)	The total number of intervals where the participant attended expressed as a percentage of the total number of HII the participant participated in.	Percentage	0 to 100
HIIT	High intensity interval training.		
HIIT group session success	The number of participants in a class who reach their target heart rate in 9 of the 10 intervals within a class.	Number	0 to 10
HIIT group session success percentage	The number of participants in a class who reach their target heart rate in 9 of the 10 intervals within a class expressed as a percentage.	Percentage	0 to 100
HIIT participant session success	The participant reaches their target heart rate in 9 of the 10 intervals within a class.	Number	0 = no; 1 = yes; 2 = data not available
HIIT participant session success percentage	The percentage of classes (0–20) attended by a participant where in 9 out of 10 intervals data is available to state targeted heart rate was achieved.	Percentage	0 to 100
Hydrostatic pressure	The pressure exerted by a fluid at equilibrium at a given point within the fluid, due to the force of gravity.		
Hydrotherapy pool temperature	The water temperature at Perth Children's Hospital hydrotherapy pool in degrees Celsius.	Number	28 to 38
Inclusion criteria	Characteristics that the prospective subjects must have if they are to be included in the study.		

Variable name	Description	Data type	Values/rules
Informed consent	Consent from both parent/guardian and assent from adolescent agreeing to participate in the study.		
Interval based training	A type of training that involves a series of low to high intensity workouts interspersed with rest or relief periods.		
Intervention adherence (participant)	Of those randomised to the intervention group, the percentage of classes that the participant attended and participated in compared to the number of classes required (i.e. 20).	Percentage	0 to 100
Intervention adherence fraction (group)	Of those randomised to intervention, the average percentage of classes that the collective intervention group attended and participated in compared to the total number of classes required.	Percentage	0 to 100
Intervention group	Participants who complete baseline assessments and who are randomised to the 10-week water based training group.	Number	0 to 10
Intervention retention	The participant in the intervention group remained in the study until completion of all post-intervention assessments.	Number	0 = withdrawn; 1 = completed
Intervention retention fraction	The percentage of participants in the intervention group who remained in the study until completion compared to the number who formally withdrew from the intervention group.	Percentage	0 to 100
Land-based exercise training	Exercise conducted where no body parts are immersed in water.		
LBW	Lean body weight – the amount of body weight (mass) minus fat tissue mass. It includes the weight of organs, skin, bones, body water and muscles.	Number	grams
Lean muscle mass	Lean muscle mass in grams as determined through DXA.	Number	grams 0.00 decimal places

Variable name	Description	Data type	Values/rules
Letter of introduction	A letter to families to discuss the study and provide an opportunity for families to 'opt out' of being contacted by study researchers.		
Low intensity interval	A 60-second interval of rest or minimal movement.		
Mod BPI	Modified Brief Pain Inventory – a self-administered questionnaire for evaluating pain and which addresses the relevant aspects of pain – history, intensity, timing, location and quality – and the pain's ability to interfere with the participant's activities. It involves recall of the past 24 hours.	Number	
Mod BPI completion	Where a participant has completed more than 50% of items to be completed to be a valid measure. Measure expressed as a mean score.		
Pain between classes score and description	The number attributed in a visual analogue scale (0–10) of the maximal musculoskeletal pain felt since the last pool class with any comment related to that pain, e.g. pain felt after doctor's visit.	Number	0 to 10
Pain with exercise	The descriptive words used by a participant to describe pain during the intervention class or CPET assessment – prompts to assist description include aching, cramping, fearful, gnawing, heavy, hot or burning, sharp and shooting.	Number	0 = no pain 1 = aching 2 = cramping 3 = fearful 4 = gnawing 5 = heavy 6 = hot or burning 7 = sharp or shooting 8 = other
Participant study ID	Study identification number issued to a participant after recruitment.	Number	1001 to 1025

Variable name	Description	Data type	Values/rules
Peak VO_2	The peak oxygen uptake or rate of oxygen consumption determined during CPET where at least 2 of the following are recorded: The respiratory exchange ratio is greater than 1.0 Physical evidence of fatigue and maximal effort Maximal predicted heart rate is achieved or ≥180 beats per minute.	Number	mL/kg/min to 0.00 decimal places
PedsQL™	Pediatric Quality of Life Inventory™ Measurement Model – a modular approach to measuring health-related quality of life (HRQoL) in healthy children and adolescents and those with acute and chronic health conditions.	Number	0 to 100
PedsQL™ completion	Where a participant has completed more than 50% of items to be a valid measure. Measure expressed as a mean score. Scores can be calculated for 4 separate domains.	Number	0 = no; 1 = yes
pHRM	Predicted heart rate maximum – the estimated maximum heart rate allocated to a participant who is unable to perform a maximum CPET.	Number	>180
Physical activity	Any bodily movement produced by skeletal muscles that requires energy expenditure.		
Pictorial CERT	Pictorial Children's Effort Rating Table (CERT). A validated and illustrated 1–10 perceived exertion scale for children.	Number	1 to 10
Pilot RCT	Pilot randomised controlled trial.		
Post Ix P score	Post-intervention pain score – the number attributed in a visual analogue scale (0–10) of the maximal musculoskeletal pain felt after exiting the pool in the intervention group.	Number	0 to 10

Variable name	Description	Data type	Values/rules
Pre Ix P score	Pre-intervention pain score – the number attributed in a visual analogue scale (0–10) of the maximal musculoskeletal pain felt prior to entry into the pool in the intervention group.	Number	0 to 10
Randomisation	The blinded allocation of participants post baseline assessments to the control or intervention group.	Number	0 = intervention; 1 = control
Randomisation tolerance	The number of participants who remain in the study after randomisation.	Number	0 to 20
Recruitment fraction	Of those invited to participate in the study, the percentage who agree to participate.	Percentage	0 to 100
Relative or absolute contraindication to CPET	As defined by the American Thoracic Society/American College of Chest Physicians.		
Research assistant	Anyone who is a paid assistant to the master's student and completes an aspect of the study.		
Retention fraction	The number of participants who remain in the study at the completion of all assessments versus the number of participants enrolled.	Percentage	0 to 100
RPE	Rate of perceived exertion – in this study, RPE refers to use of a pictorial Children's Effort Rating Table.	Number	0 to 10
School year	The year of school the participant is currently attending, e.g. Year 8. 13 is not attending school.	Number	7 to 13
Screening number	Total number of participants screened who met eligibility criteria.	Number	
Screening opt out fraction	The number of potential participants who opted out after receiving letter of invitation compared to all letters mailed.	Percentage	0 to 100
Sequence allocation	The process of determining the sequence generation for randomisation in two blocks – 12–15 years and 16–17 years – by computer random number generator (performed by KH).		

Variable name	Description	Data type	Values/rules
Serious adverse event	Any untoward medical occurrence that results in death, is life threatening, requires inpatient hospitalisation, persistent or significant disability/incapacity, or is a medically important event or reaction.		
Stretches	Supervised and standardised stretches in the water for 5–10 minutes at the conclusion of interval training.		
Study completion	Participant completes all aspects of the study.	Number	0 = withdrawn; 1 = completed
Study duration	The period of time between the study's first baseline assessment and the last participant baseline assessment post intervention or usual care period.		
Supervised exercise	Exercise performed and facilitated under the direct supervision of the master's student or research assistant.		
Supramaximal exercise	Exercise at a work rate greater than 100% VO_2 peak.		
Tanner score	Developmental age of sexual characteristics.	Number	1 to 5
TDP	Typically developing peers – adolescents considered to have development typical of the age progression of growth and physical, mental, emotional and social changes.		
Traffic light feasibility	A description of the method used in analysis of the data to assess the pilot study feasibility to progress to a randomised controlled trial(RCT) in its current format; progress to an RCT, with modification; or to determine that the study is not feasible to progress to an RCT without substantial modification.		
Usual care	No exercise or rest advice given to a participant. The participant is encouraged to continue their normal routine and activity level. Minimal contact with the research team.		

Variable name	Description	Data type	Values/rules
Vigorous intensity physical activity	Vigorous intensity activities are defined as activities ≥6 metabolic equivalents (METS). Vigorous activities require a high amount of oxygen consumption to complete the activity.		
Warm-up	Supervised and standardised exercises in the water for 5 minutes to prepare participants for physical exertion. It may include short and gentle practices of the exercises to be performed during high intensity intervals.		
Water-based exercise training	Exercise undertaken where at least part of the body is submerged in water.		
Water buoyancy	The upward force exerted by water that opposes the weight of an immersed object.		
Weight	The weight of the participant taken at baseline measurement.	Number	grams 0.00 decimal places
	Data missing.	Number	
	Query/check data.	Number	
	Data value entered is checked and correct/intentionally blank.		

Appendix 7: Participant attendance and heart rate data collection for Polar Club® app

CP HYDRO CLASS ATTENDANCE

Class number (1-30) _____ Date _____ Clinicians present _____

Participants Name	Device number	Attended	Entered pool on time	Interval success									
Pool Temp: _____		✓= yes X = no	✓= yes X = no, before HIIT XX = no, entered during HII	HII score of R = red; Y = yellow; G = green; B = Blue; G = grey XX = HRM did not work entire HII; X = HR capture too inconsistant to determine ✓= team opinion met criteria but no HR evidence dash(-) = did not participate/ complete									
				HII 1	HII 2	HII 3	HII 4	HII 5	HII 6	HII 7	HII 8	HII 9	HII 10
Participant 1													
Participant 2													
Participant 3													
Participant 4													
Participant 5													
Participant 6													

Appendix 8: Example of A-HIIT weekly exercise explanation for clinicians

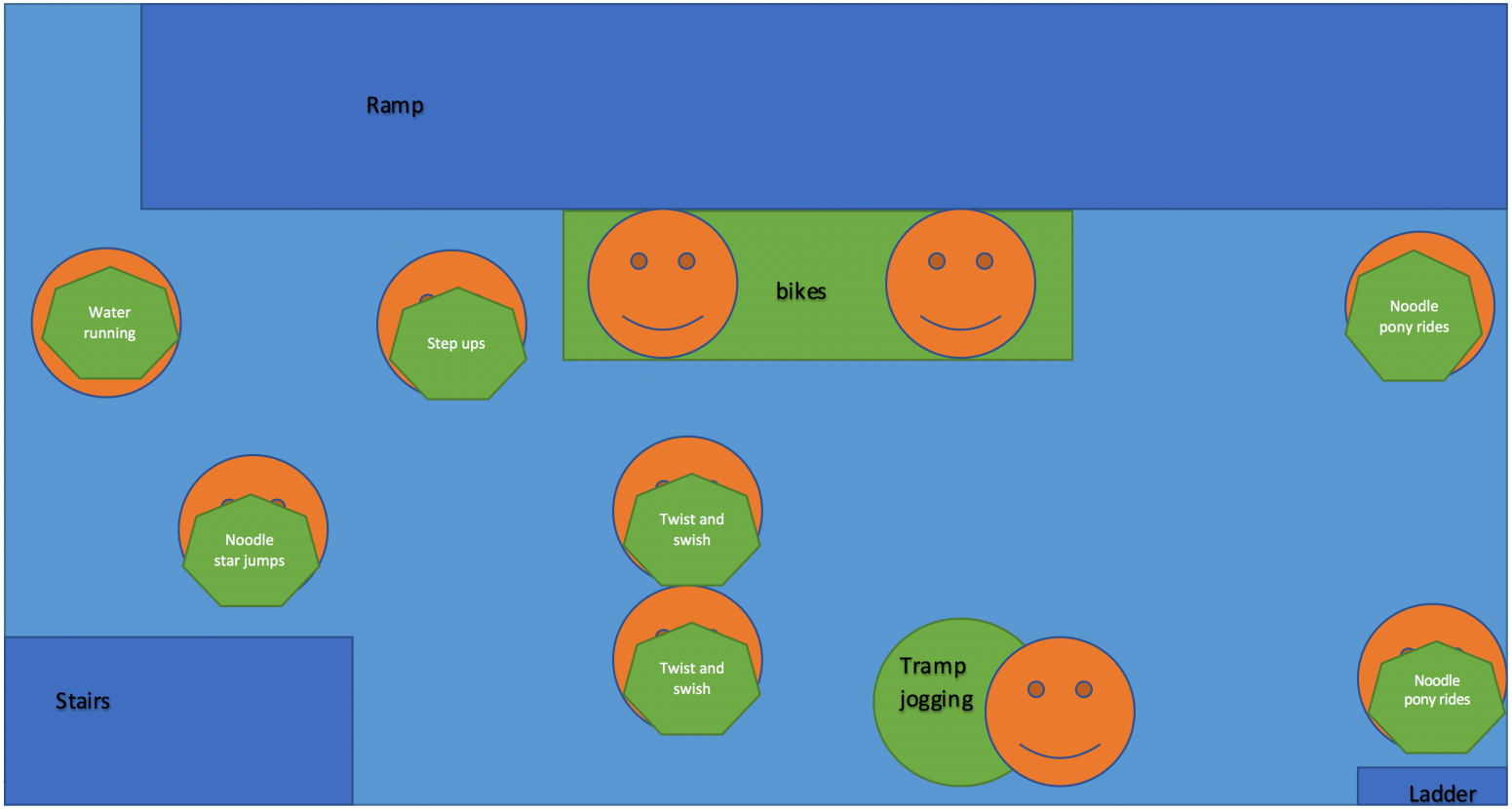
Hydro Exercises for HIIT – Week Four

Exercise Title	Exercise number	Progression	Polar compatible	Equipment required	Exercise description	Water depth for participant	Area in pool 1=shallow 2= middle 3 = deep 4 = all
Twist and swish	11.0	Yes	Yes	Nil	Allow your arms to sway side to side just under the surface of the water, twisting your trunk.	Waist depth	2
Tramp jogging	12.1	Yes	Yes	Trampoline	Jog fast on the trampoline with your arms moving in a running action	Mid chest	2
Cycling	13.0	No	Yes	Bike	Wearing bike shoes, cycle as hard and as fast as you can	Waist deep when sitting on bike	2
Noodle skiing	14.0	No	Yes	Noodle	Hold the noodle in each hand with the noodle arched above you. Punch arms forward alternatively whilst scissoring legs forwards and back in a skiing motion. Repeat quickly	Elbow depth	2
Noddle pony rides	17.0	No	No	Noodle	With the noodle between your legs, cycle with your legs whilst breast stroking with your arms. Move from side to side of the pool or stay in one spot	Deep	3
Noodle scissors	18.0	No	Yes	Noodle	Standing in the water with the noodle around your waist and held in each hand. Cross the two ends of the noodle and then pull apart. Repeat quickly	Mid chest	2
Noodle star jumps	20.0	No	Yes	Noodle	Hold the noodle in an arch above you with arms at 90 degrees by your side. As you 'jump' out with your legs, straighten the noodle. As you 'jump' in, bring the noodle ends back to your side. Repeat quickly	Waist	1 - 2

Modifications for Individual participants

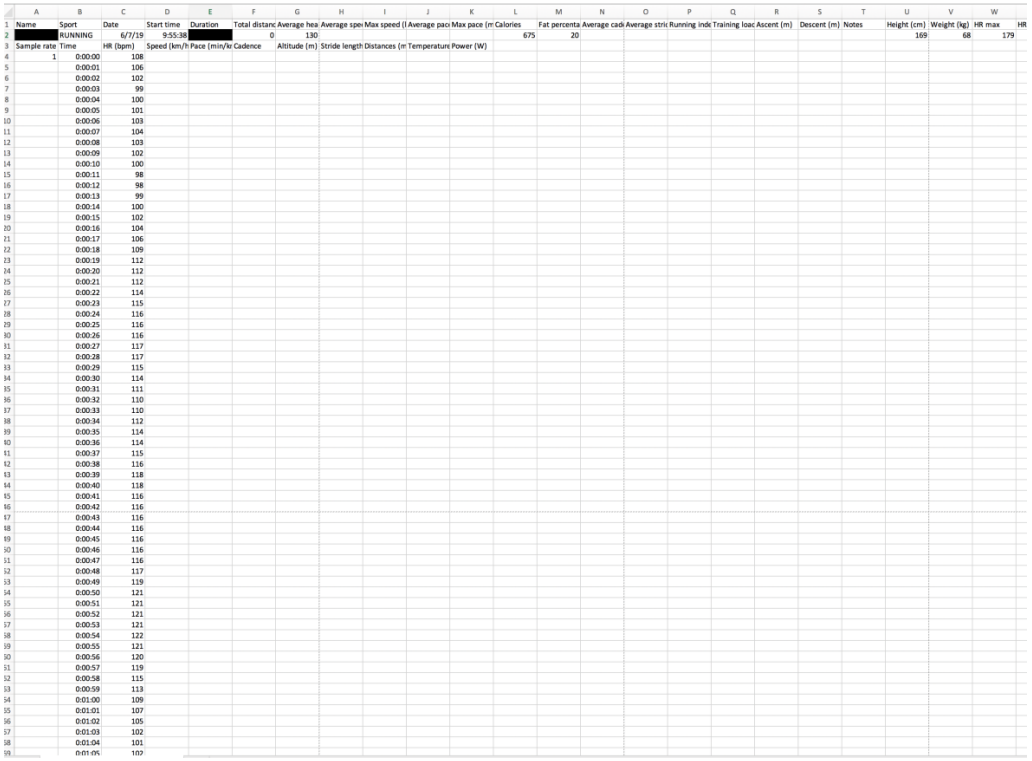
- Increase distance between legs with standing to increase BOS
- Change to one arm activity
- Use an aid to assist with bilateral arms eg pull boy
- Alter location so that support arm is the least affected side
- Increase water depth for stability
- Reduce the amount of equipment under water to decrease resistance
- Single limb activity with increased resistance

Appendix 9: Example of A-HIIT class pool layout for setting up equipment



Appendix 10: Example of participant's HR over time as represented in Polar Beat® app display

Appendix 11: Example of participant manual heart rate calculation from Polar Beat® downloads



Name	Sport	Date	Start time	Duration	Total distanc	Average hea	Average spee	Max speed (I	Average pac	Max pace (m	Calories	Fat percenta	Average cad	Average stri	Running inde	Training loac	Ascent (m)	Descent (m)	Notes	Height (cm)	Weight (kg)	HR max	HR
[redacted]	RUNNING	6/7/19	9:55:38	[redacted]	0	130					675	20								169	68	179	

Sample rate	Time	HR (bpm)	Speed (km/h	Pace (min/k	Cadence	Altitude (m)	Stride length	Distances (m	Temperatur	Power (W)
1	0:00:00	108								
	0:00:01	106								
	0:00:02	102								
	0:00:03	99								
	0:00:04	100								
	0:00:05	101								
	0:00:06	103								
	0:00:07	104								
	0:00:08	103								
	0:00:09	102								
	0:00:10	100								
	0:00:11	98								
	0:00:12	98								
	0:00:13	99								
	0:00:14	100								
	0:00:15	102								
	0:00:16	104								
	0:00:17	106								
	0:00:18	109								
	0:00:19	112								
	0:00:20	112								
	0:00:21	112								
	0:00:22	114								
	0:00:23	115								
	0:00:24	116								
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	0:00:26	116								
	0:00:27	117								
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	0:00:30	114								
	0:00:31	111								
	0:00:32	110								
	0:00:33	110								
	0:00:34	112								
	0:00:35	114								
	0:00:36	114								
	0:00:37	115								
	0:00:38	116								
	0:00:39	118								
	0:00:40	118								
	0:00:41	116								
	0:00:42	116								
	0:00:43	116								
	0:00:44	116								
	0:00:45	116								
	0:00:46	116								
	0:00:47	116								
	0:00:48	117								
	0:00:49	119								
	0:00:50	121								
	0:00:51	121								
	0:00:52	121								
	0:00:53	121								
	0:00:54	122								
	0:00:55	121								
	0:00:56	120								
	0:00:57	119								
	0:00:58	115								
	0:00:59	113								
	0:01:00	109								
	0:01:01	107								
	0:01:02	105								
	0:01:03	102								
	0:01:04	101								
	0:01:05	102								

0:26:01	164
0:26:02	164
0:26:03	163
0:26:04	162
0:26:05	162
0:26:06	162
0:26:07	162
0:26:08	163
0:26:09	163
0:26:10	163
0:26:11	163
0:26:12	163
0:26:13	165
0:26:14	165
0:26:15	165
0:26:16	166
0:26:17	168
0:26:18	168
0:26:19	169
0:26:20	169
0:26:21	169
0:26:22	170
0:26:23	171
0:26:24	171
0:26:25	172
0:26:26	172
0:26:27	172
0:26:28	172
0:26:29	173
0:26:30	173
0:26:31	173
0:26:32	174
0:26:33	174
0:26:34	174
0:26:35	174
0:26:36	174
0:26:37	174
0:26:38	174
0:26:39	173
0:26:40	171
0:26:41	169
0:26:42	165
0:26:43	163
0:26:44	162
0:26:45	160
0:26:46	158

174

Appendix 12: General health and pain collection data for the intervention group

CP HYDRO CLASS Pain and general health **Class number** _____ **Day** _____ **Date** _____

Participants Name	General health Qu	General activity Qu	Pain since last visit	Fatigue		Pre class pain and descriptor	Post class pain score	Post class pain descriptor	General comments
	✓= "well/ fine/Ok" otherwise comment	comment on exercise/ holidays/ school activity	Score severity 0-10 + comment eg post footy	After last class 1-10	Now 1-10	Score 0 to 10	Score 0 to 10	Score 0 to 7	e.g. late to class due to traffic/ siblings. Not looking forward to class after last week/ poor sleep
Participant 1									
Participant 2									
Participant 3									
Participant 4									
Participant 5									
Participant 6									

Appendix 13: Post-intervention family interview questions

1. What did you and your child like most about this research program?

2. Have you noticed any benefits for your child throughout the past 10 weeks?

3. As a parent how did you find the time commitment, how did your child find the extra time commitment?

4. What could we have done to improve our program?

5. Would you recommend this intervention to other children with cerebral palsy?

Appendix 14: Example of a DXA scan report at Perth Children's Hospital

[redacted]

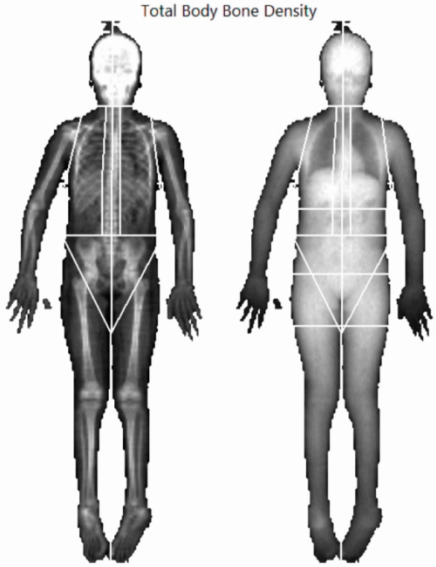
ACC#:P10735039
ST:12/04/2019 15:46:16
SE:
SE:1 IM:1

PCH

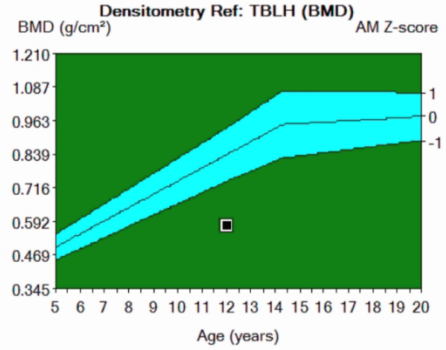
Perth Children's Hospital

Patient:	[redacted]	**Facility ID:**	
Birth Date:		**Referring Physician:**	ANDREW CHARLES WILSON
Height / Weight:	128.6 cm 28.0 kg	**Measured:**	12/04/2019 3:46:16 PM (14.10)
Sex / Ethnic:	Female White	**Analyzed:**	12/04/2019 3:48:55 PM (14.10)

Total Body Bone Density



Densitometry Ref: TBLH (BMD)



Densitometry (Enhanced Analysis)

Region	BMD[1] (g/cm²)	Age-Matched[2,3] Z-score
TBLH	0.575	-2.7

COMMENTS: school sport

Image not for diagnosis
Printed: 12/04/2019 3:51:12 PM (14.10)76:0.15:153.04:31.4 0.00:-1.00
4.81x6.50 8.4:%Fat=36.8%
0.00:0.00 0.00:0.00
Filename: I18upp6il0.dfb
Scan Mode: Thin 0.4 µGy

1 - Statistically 68% of repeat scans fall within 1SD (± 0.010 g/cm² for Total Body TBLH)
2 - Australia (Combined Geelong/Lunar) Total Body Reference Population (v113)
3 - Matched for Age, Ethnic

GE Healthcare
Lunar Prodigy Advance
PA+38[illegible]
DXA Reports
Bone Densitometry
LgM: EI:

PCH
Dr:WILSON,ANDREW,CHARLES

Appendix 15: Outcome measures of health-related quality of life

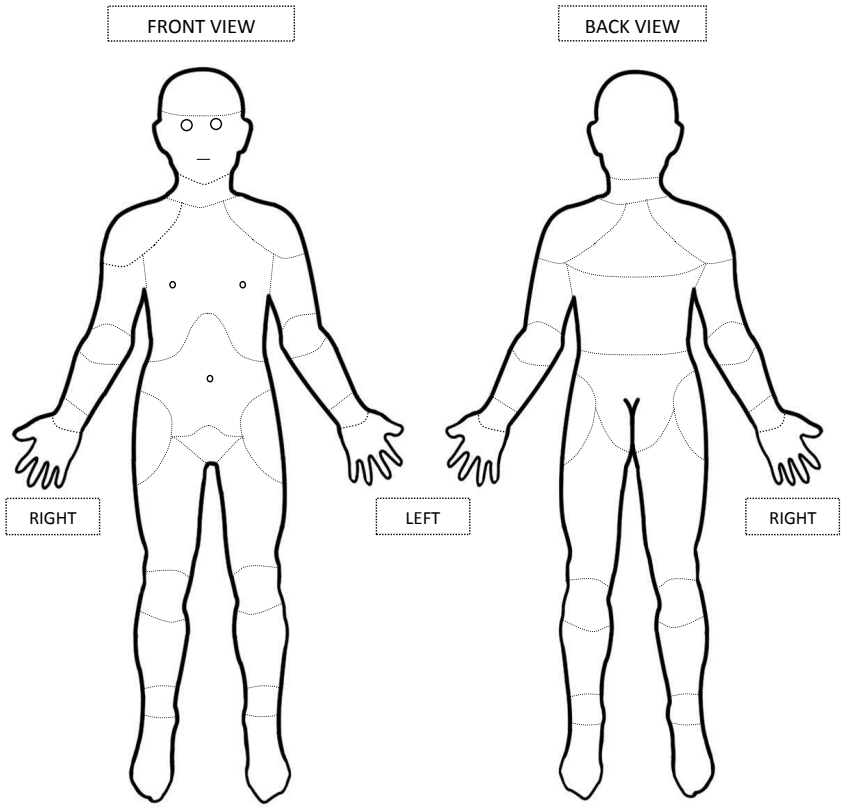
V1.0

For patients aged 13 years and over	Name: …….. Today's date: ……..

Adolescent Questionnaire

Your pain

On the diagram below, shade in the areas where you feel pain. Put an X on the area that hurts most.



Office use only

Main pain	☐ Head (exc face)	☐ Forearm	☐ Knee	**Other pain**	☐ Head (exc face)	☐ Forearm	☐ Knee
	☐ Face/jaw/temple	☐ Wrist	☐ Calf		☐ Face/jaw/temple	☐ Wrist	☐ Calf
	☐ Throat/neck	☐ Hand	☐ Ankle		☐ Throat/neck	☐ Hand	☐ Ankle
	☐ Shoulder	☐ Abdomen	☐ Foot		☐ Shoulder	☐ Abdomen	☐ Foot
	☐ Chest	☐ Hip	☐ Upper back		☐ Chest	☐ Hip	☐ Upper back
	☐ Upper arm	☐ Groin/pubic area	☐ Mid back		☐ Upper arm	☐ Groin/pubic area	☐ Mid back
	☐ Elbow	☐ Thigh	☐ Low back		☐ Elbow	☐ Thigh	☐ Low back

Pain Chart Source: Childhood Arthritis and Rheumatology Research Alliance, www.carragroup.org
von Baeyer CL et al, *Pain Management*, 2011;1(1):61-68.

Which statement best describes your pain? (tick one box only)

☐ Always present (always the same intensity)

☐ Always present (intensity varies)

☐ Often present (pain free periods last less than 6 hours)

☐ Occasionally present (pain occurs once to several times per day, lasting up to an hour)

☐ Rarely present (pain occurs every few days or weeks)

Rate your pain by circling the one number that best describes the following:	
a) Your **worst** pain in the last week?	0 1 2 3 4 5 6 7 8 9 10 No pain — Pain as bad as you can imagine
b) Your **least** pain in the last week?	0 1 2 3 4 5 6 7 8 9 10 No pain — Pain as bad as you can imagine
c) Your **usual** pain in the last week?	0 1 2 3 4 5 6 7 8 9 10 No pain — Pain as bad as you can imagine
d) How much pain do you have **right now**?	0 1 2 3 4 5 6 7 8 9 10 No pain — Pain as bad as you can imagine

BAPQ 5

There are many possible ways that pain can affect the lives of young people. Below are some statements that may or may not apply to you. Please read each statement and put a cross in the box (x) under the word that describes how often you have experienced each of these things in the **LAST TWO WEEKS**. Please make sure that you answer all questions

Please tell us about any specific worries or concerns you have about your pain

		Never	Hardly ever	Some times	Often	Always
1	I worry about my pain problem	☐	☐	☐	☐	☐
2	I avoid activities that cause pain	☐	☐	☐	☐	☐
3	When I think about my pain, it makes me upset	☐	☐	☐	☐	☐
4	Pain scares me	☐	☐	☐	☐	☐
5	I worry that I will do something to make my pain worse	☐	☐	☐	☐	☐
6	When I have pain, I think something harmful is happening	☐	☐	☐	☐	☐
7	I am afraid to move due to pain	☐	☐	☐	☐	☐

PedsQL™

Paediatric Quality of Life Inventory

Version 4.0 – Australian English

TEENAGER REPORT (ages **13-18**)

DIRECTIONS

On the following page is a list of things that might be a problem for you.
Please tell us **how much of a problem** each one has been for you
in the **LAST MONTH** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the **LAST MONTH**, how much of a **problem** has this been for you …*

ABOUT MY HEALTH AND ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is difficult for me to walk more than 100 metres	0	1	2	3	4
2. It is difficult for me to run	0	1	2	3	4
3. It is difficult for me to play sport or do exercise	0	1	2	3	4
4. It is difficult for me to lift something heavy	0	1	2	3	4
5. It is difficult for me to have a bath or shower by myself	0	1	2	3	4
6. It is difficult for me to help around the house	0	1	2	3	4
7. I get aches and pains	0	1	2	3	4
8. I have low energy	0	1	2	3	4

ABOUT MY FEELINGS *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel afraid or scared	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I have trouble sleeping	0	1	2	3	4
5. I worry about what will happen to me	0	1	2	3	4

HOW I GET ALONG WITH OTHERS *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I have trouble getting along with other teenagers	0	1	2	3	4
2. Other teenagers do not want to be my friend	0	1	2	3	4
3. Other teenagers tease me	0	1	2	3	4
4. I cannot do things that other people my age can do	0	1	2	3	4
5. It is hard to keep up with other teenagers	0	1	2	3	4

ABOUT SCHOOL *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard to pay attention in class	0	1	2	3	4
2. I forget things	0	1	2	3	4
3. I have trouble keeping up with my school work	0	1	2	3	4
4. I am away from school because I feel sick	0	1	2	3	4
5. I am away from school to go to the doctor or hospital	0	1	2	3	4

ID# _____

Date:_____

PedsQL™

Multidimensional Fatigue Scale

Standard Version – Australian English

TEEN REPORT (ages **13-18**)

DIRECTIONS

On the following page is a list of things that might be a problem for you. Please tell us **how much of a problem** each one has been for you during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

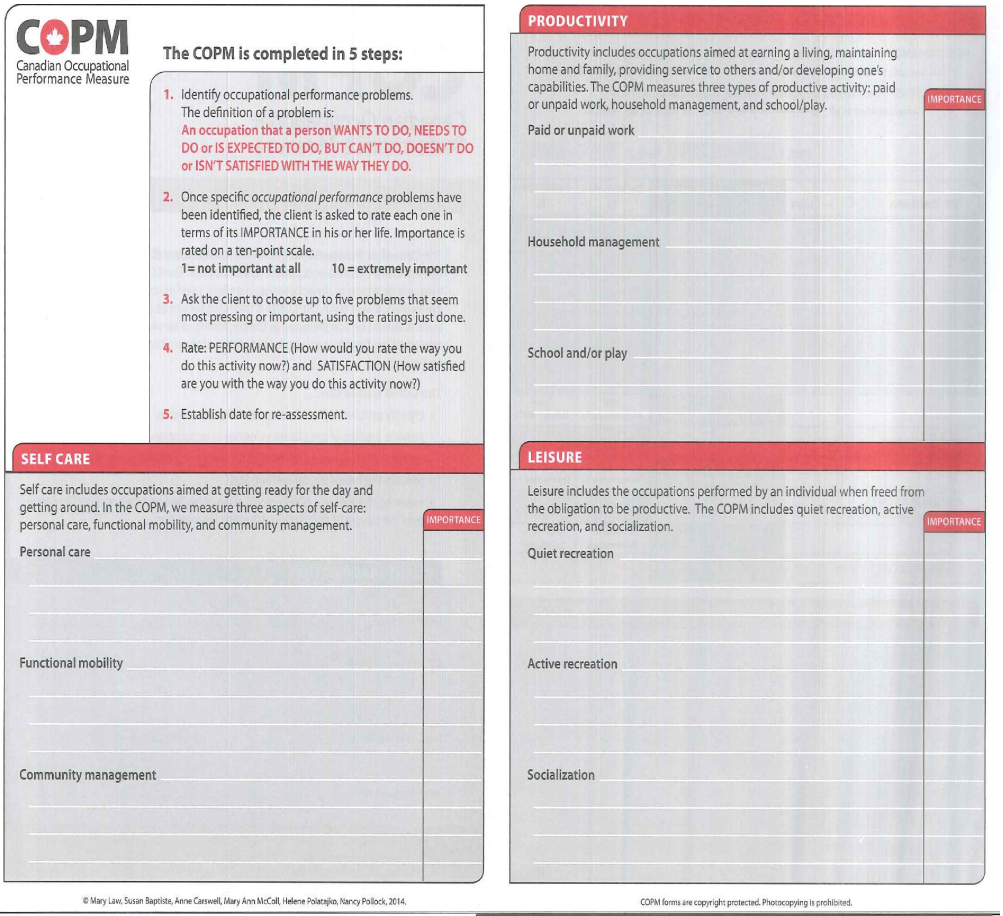
*In the past **ONE month,** how much of a **problem** has this been for you?*

GENERAL FATIGUE ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I feel tired	0	1	2	3	4
2. I feel physically weak (not strong)	0	1	2	3	4
3. I feel too tired to do things that I like to do	0	1	2	3	4
4. I feel too tired to spend time with my friends	0	1	2	3	4
5. I have trouble finishing things	0	1	2	3	4
6. I have trouble starting things	0	1	2	3	4

SLEEP/REST FATIGUE ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. I sleep a lot	0	1	2	3	4
2. It is hard for me to sleep through the night	0	1	2	3	4
3. I feel tired when I wake up in the morning	0	1	2	3	4
4. I rest a lot	0	1	2	3	4
5. I take a lot of naps	0	1	2	3	4
6. I spend a lot of time in bed	0	1	2	3	4

COGNITIVE FATIGUE ***(problems with…)***	**Never**	**Almost Never**	**Some-times**	**Often**	**Almost Always**
1. It is hard for me to keep my attention on things	0	1	2	3	4
2. It is hard for me to remember what people tell me	0	1	2	3	4
3. It is hard for me to remember what I just heard	0	1	2	3	4
4. It is hard for me to think quickly	0	1	2	3	4
5. I have trouble remembering what I was just thinking	0	1	2	3	4
6. I have trouble remembering more than one thing at a time	0	1	2	3	4

Appendix 16: Canadian Occupational Performance Measure



COPM
Canadian Occupational Performance Measure

The COPM is completed in 5 steps:

1. Identify occupational performance problems.
   The definition of a problem is:
   An occupation that a person WANTS TO DO, NEEDS TO DO or IS EXPECTED TO DO, BUT CAN'T DO, DOESN'T DO or ISN'T SATISFIED WITH THE WAY THEY DO.
2. Once specific *occupational performance* problems have been identified, the client is asked to rate each one in terms of its IMPORTANCE in his or her life. Importance is rated on a ten-point scale.
   1= not important at all 10 = extremely important
3. Ask the client to choose up to five problems that seem most pressing or important, using the ratings just done.
4. Rate: PERFORMANCE (How would you rate the way you do this activity now?) and SATISFACTION (How satisfied are you with the way you do this activity now?)
5. Establish date for re-assessment.

SELF CARE

Self care includes occupations aimed at getting ready for the day and getting around. In the COPM, we measure three aspects of self-care: personal care, functional mobility, and community management.

IMPORTANCE

Personal care

Functional mobility

Community management

PRODUCTIVITY

Productivity includes occupations aimed at earning a living, maintaining home and family, providing service to others and/or developing one's capabilities. The COPM measures three types of productive activity: paid or unpaid work, household management, and school/play.

IMPORTANCE

Paid or unpaid work

Household management

School and/or play

LEISURE

Leisure includes the occupations performed by an individual when freed from the obligation to be productive. The COPM includes quiet recreation, active recreation, and socialization.

IMPORTANCE

Quiet recreation

Active recreation

Socialization

SCORING

PERFORMANCE (How would you rate the way you do this activity now?)
1 = not able to do it at all ⟷ 10 = able to do it extremely well

SATISFACTION (How satisfied are you with the way you do this activity now?)
1 = not satisfied at all ⟷ 10 = extremely satisfied

TIME 1: ____/____/____ TIME 2: ____/____/____

OP Problems	Impt	Performance T_1	Satisfaction T_1	Performance T_2	Satisfaction T_2
1.					
2.					
3.					
4.					
5.					
		Total Performance T_1	Total Satisfaction T_1	Total Performance T_2	Total Satisfaction T_2
TOTAL SCORES					
		Average Performance T_1	Average Satisfaction T_1	Average Performance T_2	Average Satisfaction T_2
AVERAGE SCORES (Total score / number of problems)					
				Change in Performance	Change in Satisfaction
CHANGE SCORES (T2 - T1)					

NOTES AND OBSERVATIONS

Initial Assessment

Re-assessment

COPM
Canadian Occupational Performance Measure

The **Canadian Occupational Performance Measure (COPM)** supports high-quality, client-centred, occupation-based practice. The COPM is an individualized measure designed to detect change in a client's self-perception of occupational performance over time. The COPM is intended for use as an outcome measure. As such, it should be administered at the beginning of service to establish intervention goals, and again at an appropriate interval thereafter to determine progress and outcome.

The COPM is used to:

- identify problem areas in occupational performance;
- provide a rating of the client's priorities in occupational performance;
- evaluate performance and satisfaction relative to those problem areas;
- provide the basis for goal-setting; and,
- measure changes in a client's perception of his/her occupational performance over the course of occupational therapy intervention.

CLIENT INFORMATION

Client name: _____

Client date of birth: ____/____/____

Initial assessment: ____/____/____ Re-assessment: ____/____/____

Therapist name: _____

Appendix 17: Minor amendments made to protocol during the study period

Assessments

For the first eight cardiopulmonary exercise tests conducted, participants were asked to grade their perceived effort during the test using a pictorial children's exertion rating table.[123] This table was abandoned after the first eight exercise tests as scores created stress for some participants and the clinicians conducting the tests did not feel that these scores were helpful in determining maximal exercise effort.

Intervention

For the first few HIIT classes, of the 10 stations, five or more were changed each week to maximise variety. After 2 weeks, this was modified so that no more than two stations were changed per week. This modification was in response to participant feedback about experiencing anxiety around learning new routines/exercises. Stability of some exercises every week (e.g. cycling and step-ups) were helpful to participants and the protocol was adjusted accordingly.

Participants were initially asked to rate their perceived effort after each exercise station during a class with analogue scales of 0–10. Reporting of the rate of perceived effort was simplified after the first week to report on the easiest and most difficult exercise station after the class had finished rather than after each interval. This was to simplify the procedural learning and attention required while exercising.

For each participant, the HR recorded during exercise, expressed as a percentage of their HRpeak, was digitally projected onto a wall using Polar Club® to provide HR feedback to participants (e.g. ≥80% HRpeak). The HR was colour coded to reflect intensity and assist in goal setting. By the fifth week of the intervention period, it was clear to the investigators that participants were able to exercise at HRs ≥80% HRpeak in water. To maintain motivation, the colour coding criteria was changed to encourage participants to exercise at even higher intensity. That is 'red' (highest HR goal) was changed from 80% HRpeak to 85% HRpeak. Goal setting was further increased by 5% 2 weeks later. This did not change the study definition of success of an interval (i.e., ≥80% HRpeak) but did assist with participant motivation and enjoyment.

Appendix 18: Attribution statement: the effect of aquatic high intensity interval training on aerobic performance and body composition in a non-athletic population

Conception and Design	Acquisition of Data and Method	Data Conditioning/ Manipulation	Analysis and Statistical Method	Interpretation and Discussion	Final Approval	Total % contribution
Co-Author 1: Julie Depiazzi	✓	✓	✓	✓	✓	40%
Co-Author 1 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 2: Kylie Hill	✓		✓	✓	✓	20%
Co-Author 2 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 3: Rachel Forbes	✓	✓	✓		✓	15%
Co-Author 3 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 4: Noula Gibson	✓		✓	✓	✓	10%
Co-Author 4 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 5: Nadine Smith	✓				✓	5%
Co-Author 5 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 6: Andrew Wilson	✓			✓	✓	5%
Co-Author 6 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 7: Roslyn Boyd	✓				✓	5%

Co-Author 7 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:		
Total %		**100%**

Appendix 19: Attribution statement – aquatic high intensity interval training to improve aerobic capacity is feasible in adolescents with cerebral palsy – pilot randomised controlled trial

Conception and Design	Acquisition of Data and Method	Data Conditioning/ Manipulation	Analysis and Statistical Method	Interpretation and Discussion	Final Approval	Total % contribution
Co-Author 1: Julie Depiazzi	✓	✓	✓	✓	✓	40%
Co-Author 1 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 2: Nadine Smith	✓	✓		✓	✓	20%
Co-Author 2 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 3: Kylie Hill	✓		✓	✓	✓	20%
Co-Author 3 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 4: Noula Gibson	✓		✓	✓	✓	10%
Co-Author 4 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 5: Andrew Wilson	✓			✓	✓	5%
Co-Author 5 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Co-Author 6: Kate Langdon				✓	✓	5%
Co-Author 6 Acknowledgment: I acknowledge that these represent my contribution to the above research output Signed:						
Total %						**100%**