

Telehealth Movement-to-Music with arm-based sprint-intensity interval training to improve cardiometabolic health and cardiorespiratory fitness in children with cerebral palsy: protocol for a pilot randomized controlled trial

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Telehealth Movement-to-Music with arm-based sprint-intensity interval training to improve cardiometabolic health and cardiorespiratory fitness in children with cerebral palsy: protocol for a pilot randomized controlled trial

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Abstract

Background: Children with mobility disabilities, including those with cerebral palsy, have limited options and limited time to exercise to manage their cardiometabolic health and cardiorespiratory fitness. Regular cardiovascular exercise during childhood is a critical health behavior for preventing health decline in adulthood. Thus, there is an urgent need for accessible, age-appropriate, convenient exercise modalities in this group. Sprint-intensity interval training (SIT), combined with telehealth procedures, may be ideal for children with disabilities. SIT includes repetitive bouts of maximal exercise effort combined with rest periods, which can be effective in eliciting comparable results to moderate-exercise training with very short training durations.

Objective: This phase I pilot feasibility randomized controlled trial aims to investigate the potential effects of a 12-week SIT program on indicators of cardiorespiratory fitness and cardiometabolic health among children with cerebral palsy. An ancillary aim is to evaluate the feasibility of the program via several process feasibility metrics.

Methods: This study includes a two-armed parallel group design. Fifty physically inactive children with cerebral palsy (aged 6-17 years) will be randomly allocated into one of two groups: a 12-week SIT or a waitlist control group that continues habitual activity for 12 weeks. The SIT prescription includes 3 tele-supervised sessions/week with 30 repeated sequences of 4-seconds of maximal arm exercise, with active recovery, warm-up, and cooldown periods (~20-min total session). SIT includes guided videos with child-themed arm routines and music. The exercise sessions will be remotely supervised through a web-based videoconference application and include safety monitoring equipment. Outcomes are measured at pre and post intervention (week 0, 13, respectively). Health outcome measures include peak oxygen consumption (VO₂ peak), measured by a graded exercise test; high sensitivity C-reactive protein and blood insulin, hemoglobin A1c, triglycerides, and cholesterol using a finger stick dried blood spot test; blood pressure, using a sphygmomanometer; and body composition (total mass, total lean mass, tissue % lean, and tissue % fat) using dual x-ray absorptiometry (DEXA). Feasibility will be evaluated by the following metrics: adverse events or problems experienced throughout the intervention related to participant safety; perceived enjoyment; and recruitment, enrollment, and attrition rates.

Results: Recruitment procedures started in November 2023. All data are anticipated to be collected by February 2025. Full trial results are anticipated to be analyzed and submitted for publication by March 2025. Secondary analyses of data will be subsequently published.

Conclusions: This trial tests an accessible and low-cost exercise program, which leverages principles of high-intensity exercise to provide a convenient program for children with physical disabilities. Knowledge obtained from this study will inform the development of a larger trial for improving the cardiometabolic health, cardiorespiratory fitness, and well-being of children with physical disabilities. Clinical Trial: NCT05619211

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Original Paper

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Background: Children with mobility disabilities, including those with cerebral palsy, have limited options and limited time to exercise to manage their cardiometabolic health and cardiorespiratory fitness. Regular cardiovascular exercise during childhood is a critical health behavior for preventing health decline in adulthood. Thus, there is an urgent need for accessible, age-appropriate, convenient exercise modalities in this group. Sprint-intensity interval training (SIT), combined with telehealth procedures may be ideal for children with disabilities. SIT includes repetitive bouts of maximal exercise effort combined with rest periods, which can be effective in eliciting comparable results to moderate-exercise training with very short training durations.

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Trial Registration: NCT05619211

Introduction

Overview of Cerebral Palsy

Cerebral palsy (CP) is a clinical syndrome that results from a developmental brain injury, which is prevalent in approximately 1 million people in the United States, and 23 million people worldwide [1]. CP is characterized by disorders of movement and posture due to non-progressive disturbances of the fetal or developing brain, which is often accompanied by disturbances of sensation, perception, cognition, communication, and behavior [2]. Secondary neurological conditions such as epilepsy, along with other musculoskeletal issues are also found among people who are diagnosed with CP [2]. Recent innovations in treatments and technology have increased survival rates of people with CP, resulting in a steadily growing population of adults with CP [3]. Health professionals can now focus on how to help empower young people with CP to live well with a disability, amidst a variety of health conditions and social-ecological challenges they face towards community engagement in adulthood [4, 5].

Adulthood and Cardiometabolic Risk

The transitory age between childhood and adulthood is a critical stage for health prevention interventions. As children with CP age into their 20s, they have substantially increased risk of cardiovascular disease (CVD), related conditions, metabolic syndrome [6], and a 3-fold increased risk of CVD mortality compared to the general population [7-12]. Risk for these cardiometabolic diseases can be lowered by participation in regular moderate-intensity exercise [13-15]. However, recent studies demonstrate that children with CP do not engage in sufficient levels of exercise to obtain health-enhancing benefits, and these levels of exercise participation are far lower than those observed among children without CP [16-18]. Children with CP who exercise regularly are two times more likely to exercise as adults [16]. Consequently, there is a need to identify interventions that promote early adoption of exercise as a behavior for preventing disease onset, particularly among children with CP who have a mobility disability and have the greatest risk of cardiometabolic disease [19].

Limited Aerobic Exercise Options for Mobility Disability

Conventional exercises that are known to improve cardiorespiratory fitness (eg, walking, running, and cycling) generally require long durations of training that [20] are unsuitable for a large majority of children with CP. As children transition into adulthood, approximately 27% will be unable to walk, 35% will experience decreased walking ability, and 9% will lose their ability to walk [21, 22]. Children and adults with CP often experience secondary conditions, such as hemiparesis, fatigue, impaired balance, and joint pain, and these factors impede their mobility to walk or perform types of exercise (e.g., cycle) for prolonged periods [22-25]. A recent scoping review demonstrated that a top research priority in the field of disability and exercise is to develop aerobic exercises that are inclusive of wheelchair users [26]. The standard options for exercise training methods require leg work or full body involvement. Aside from arm-ergometry or wheelchair propulsion, there are limited aerobic exercise modalities that are efficacious or effective for people who require seated training, particularly modalities that accommodate hemiparesis [26]. Of note, no randomized controlled trial of exercise has demonstrated a clinically meaningful improvement in cardiorespiratory risk factors in children with CP, likely due to lack of a sufficient exercise dose [26].

Limited Generalizability of Research Interventions

A second notable limitation of published exercise interventions for CP is low rates of both recruitment and participation [26, 27]. Despite over 30 years of research including 49 published randomized controlled trials, the average sample size for a study was $n=30$ [26], most participants were ambulatory, and wheelchair users were typically excluded from participation. These issues hinder the generalizability and transferability of study findings [26-30]. Moreover, low levels of

participation in exercise trials were speculated to be due to the following barriers: 1) in-person supervision by a rehabilitation professional or exercise specialist, 2) onsite training at a laboratory or fitness facility, and 3) costly, specialized equipment [26, 27]. The identification of an effective evidence-based modality that is inclusive of wheelchair users will require a protocol that can be replicated in a large trial that can provide confirmatory study findings.

Rationale for Sprint-Intensity Arm Exercise

High-intensity interval training (HIIT) has been studied as an effective dose of exercise that has a moderate to large effect on cardiorespiratory fitness and cardiometabolic health, while requiring a lower duration of training compared with continuous moderate-intensity exercise [31, 32]. Sprint-intensity interval training (SIT) is a form of HIIT, that also emphasizes short bouts of exercise with maximum effort at or above a power output associated with a peak oxygen consumption (VO_2 peak) [33]. A meta-analysis [34] found that SIT has a moderate-to-large effect on VO_2 peak that is similar to both moderate-intensity [35, 36] and HIIT exercise [33]. Regarding the mechanism (displayed in Figure 1), SIT is believed to utilize anaerobic energy pathways of type IIB muscle fibers, the fast twitch glycolytic pathways, that are replenished through oxidative metabolism [37], primarily from the kidneys [38]. In other words, extremely short bouts of exercise at maximal-intensity can be sustained through the utilization of lower-intensity aerobic energy pathways. Specifically, higher exercise intensities result in increased renal hypoxemia and, thus, greater production of the hormone erythropoietin. Erythropoietin initiates production of red blood cells that enhance oxygen utilization of performing muscles [39] and, thus, VO_2 peak. SIT may further increase VO_2 peak through mitochondrial biogenesis and capillary density [37]. A higher VO_2 peak is linked with reduced risk for cardiometabolic disease [36, 40], which is relevant among children with CP who generally have low levels of cardiorespiratory fitness [41]. HIIT with a treadmill has been found to be safe and effective in improving VO_2 peak among ambulatory Children with CP [42]. To the best of our knowledge, no randomized controlled efficacy or effectiveness trial has examined a wheelchair accessible SIT program among children with CP [26, 27]. SIT with arm-cycling results in greater gains in VO_2 peak versus leg-cycling (e.g., 52% vs 6% increase in VO_2 peak, respectively) [43], likely due to a higher proportion of fast muscle fibers in the arms [43], and this info holds great value for non-ambulatory children who can use their arms for exercise.

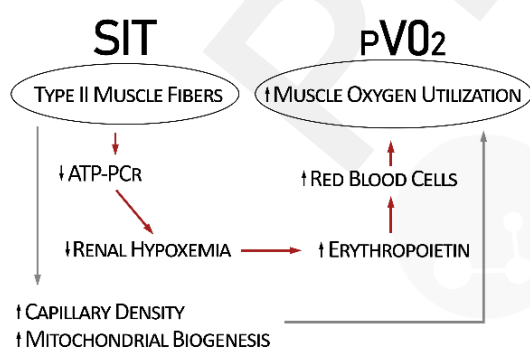


Figure 1. Conceptual model for the mechanism that underpins sprint-intensity interval training.

Telehealth Interventions Foster Enrollment and Attendance

To address low participation, telehealth interventions are especially promising because they remove many of the geographic, environmental, and financial burdens associated with traveling and paying for community programs and classes [44]. Telehealth interventions are especially helpful in delivering health services to children because of reduced burden on the caregiver, who would otherwise have to miss work or other commitments to travel or allocate transportation for the child's

appointment [44]. Transportation and time are substantial barriers to exercise participation among people with disabilities [5]. Studies have shown that overall ratings of caregivers towards telehealth appointments are highly positive and most view telemedicine consultations as at least as effective as those onsite; caregivers had modest reductions in out-of-pocket costs and less missed work cost [45]. In addition, telehealth programs are believed to enhance adherence by providing a sense of accountability, which is created by performance monitoring and social bonds with trusted professionals, as explained by the Supportive Accountability Theory [46]. Home-based exercise programs that incorporate tele-communications and monitoring have achieved the largest sample sizes of people with disabilities [26, 27, 47]. It is important to note that telehealth interventions that aim to promote strong attendance to an exercise regimen will require remote behavioral coaching strategies to maintain participant engagement. One meta-analysis found that exercise interventions that include behavioral techniques have stronger effects on exercise behavior than those that do not have such techniques [48].

Study Rationale and Purposes

In consideration of exercise barriers (lack of time and transportation) and facilitators of engagement among children (enjoyable exercises and music), the present study aims to test a program among children with CP that incorporates: 1) age-appropriate pre-recorded videos with movement-to-music exercises that are based on child-appropriate themes (e.g., superheroes, sports, pop music, etc.); 2) replicable cloud-based tele-monitoring procedures; 3) behavioral tele-physical education supervision during exercise; and 4) SIT adapted for varying arm abilities.

This study has 3 aims:

Aim 1: to examine the effects of a 12-week home-based SIT program on cardiorespiratory fitness (VO₂ peak) compared to a waitlist control (WC) among 50 Children with CP. We hypothesized that the immediate start group would achieve greater improvements in VO₂ peak versus WC.

Aim 2: to explore the effect estimates of SIT on cardiometabolic risk indicators vs WC. Cardiometabolic outcomes will include high sensitivity C-reactive protein and blood insulin, hemoglobin A1c, triglycerides, and cholesterol using a finger stick dried blood spot test, and blood pressure using a sphygmomanometer. Additionally, body composition (total mass, total lean mass, tissue % lean, and tissue % fat) will be measured using dual x-ray absorptiometry (DEXA). We hypothesized that arm-SIT will demonstrate greater improvements in outcomes versus WC.

Aim 3: to evaluate the feasibility of arm-SIT among children with CP. Feasibility will be measured by *process* feasibility metrics [49] to inform a larger efficacy/effectiveness trial. Variables will include:

- 1) intervention adherence (class attendance and video mins via cloud-based analytics)
- 2) safety (adverse events or problems experienced throughout the intervention related to participant safety)
- 3) perceived enjoyment
- 4) engagement (recruitment, enrollment, and attrition rates).

Methods

Study Design & Overview

This phase I, pilot feasibility randomized controlled trial will include a two-armed parallel group design to examine the preliminary effects of 12 weeks of arm-based SIT on cardiorespiratory fitness compared to a WC that undergoes habitual activities for 12 weeks. The project will include 50 children with CP (n=25 per arm) and caregivers (50 parent-child dyads). One caregiver is required to participate by supporting their child's safety through the intervention and managing the child's exercise schedule. Participants will come to the lab for 2 total visits (baseline and week 13) to complete the data collection procedures. Participants will perform the intervention at the home, with supervision by a research staff member through telecommunications.

Ethics Approval and Informed Consent Procedure

The protocol and informed consent and assent forms were approved by the Institutional Review Board for Human Use of the University (IRB-300008913) on January 12th, 2023. Prospective participants provide written informed consent or assent documentation prior to participation in the study. Consent and assent forms are completed on the baseline data collection visit.

Participants

Eligible child participants will meet the following inclusion criteria:

- 1) have a medical diagnosis of cerebral palsy, as determined by ICD-10 codes
- 2) aged 6-17 years old
- 3) Gross Motor Function Classification System Level I-III (as determined via participant screening, explained in the protocol section below)
- 4) medical clearance from a physician to participate in high-intensity exercise (using the attached medical screening form and explained in the intervention safety, monitoring, and response plan)
- 5) access to a Wi-Fi Internet connection in the home via mobile phone or tablet computer
- 6) a caregiver who will support and monitor the participant's safety during the intervention and manage the child's exercise schedule.

Child participants will be excluded from the study based on the following criteria:

- 1) physically active (defined as >150 minutes per week of self-reported moderate-to-vigorous intensity exercise in a typical week)
- 2) cannot use their arms for exercise
- 3) Gross Motor Function Classification Level of IV-V
- 4) complete blindness or deafness
- 5) pregnant
- 6) uses a pacemaker
- 7) has not been seen by a physician within the previous 12 months of their baseline visit
- 8) uses a g-tube
- 9) any past history of a contraindication to exercise testing: ischemia; myocardial infarction or other acute cardiac event; unstable angina; uncontrolled cardiac dysrhythmias; aortic stenosis; heart failure; pulmonary embolus or pulmonary infarction; myocarditis or pericarditis; aneurysm; low

bone-mineral density of the spine (a z-score of ≤ -3 , indicative of high risk for fracture that could occur from torsion of spine, determined at baseline data collection via the DXA scan).

Contraindications were informed by guidelines that were established by the American College of Sports Medicine [50] and Pediatric Physical Medicine and Rehabilitation physician of the study team (DD).

Eligible caregivers will include parents or legal guardians of the child, who can commit sufficient time to support the child in their roles for the study and communicate in English. Caregivers who have complete blindness or deafness will be excluded from participation.

Participant Roles

Both the child and caregiver are considered participants in the study, and both will sign the informed consent or assent document.

The child's role in the study is:

- 1) to exercise following along with exercise videos, 3x per week for 12 weeks while being supervised through a videoconference web platform by a tele-coach

The caregiver's roles in the study are to:

- 1) attend each videoconference exercise session with the child
- 2) schedule and manage the child's exercise schedule
- 3) support the safety of the child
- 4) achieve CPR/AED certification through the Red Cross (certified by the PI, BL, who is certified as a Red Cross CPR instructor)

Screening and Recruitment

Screening for this study will occur in three phases. First, contact lists are screened for a medical diagnosis of cerebral palsy based on ICD-10 codes. Second, during the initial recruitment contact with a caregiver, research staff will screen participants based on the study eligibility criteria. As noted previously, to enhance intervention safety, this study is aiming to enroll children with CP who have only mild to moderate mobility disability and have low risk of adverse events from exercise. Therefore, the initial recruitment contact via telephone will include the Gross Motor Function Classification System level [51], using the GMFCS Family Report Questionnaire [52-54]. This assessment will be asked over the phone to avoid wasting participants' time arriving at the laboratory and learning that they do not qualify for the study. Third, a physician (study physician or participants' physicians) will review the patient's medical record and screen the participant for contraindications to exercise testing and risks for participation in high-intensity exercise (based on the Medical Clearance Form: Appendix 1). In summary, if an Absolute Contraindication is identified, the participant will be excluded/withdrawn from the study. If no Absolute Contraindication is identified, the physician will identify major and minor risks based off the latest information within the patient's medical record and then decide based on Major & Minor risks to clear the participant for high-intensity exercise training at home. To ensure that record review information is relevant, if the patient has not been seen in the past year, they will have to be seen prior to obtaining medical clearance and participating in the study.

Candidates will be pre-screened and recruited primarily from medical and billing records from the Children's Hospital of Alabama. Medical and billing record databases will be pre-screened for

potentially eligible patients by patient diagnoses codes (ICD-10-CM) related to CP. Contact lists will be generated from the databases. Specific methods of recruitment will include word of mouth and flyers, physician, and staff referrals, mailouts, and phone calls.

Randomization & Clinical Trial Considerations

Parent-child dyads will be randomized into one of two groups: arm-based SIT or WC (n=25 per group) with 1:1 allocation ratio using a permuted block randomization approach. The randomization sequence was generated by the project statistician (RO), using a computer-generated randomized permuted block design (SAS, version 9.4). The project statistician sent the randomization sequence to the database manager (RY) during the study preparation phase. The database manager will unfold the randomization sequence for successfully enrolled participants to one of the two groups. Study outcomes will be assessed by a research assistant, who will be blinded to group allocation (single-blinded trial design). Recruitment will occur primarily through the Children's Hospital of Alabama. All other activities will be conducted at the Human Performance Laboratory in the Wellness Health and Research Facility at the University of Alabama at Birmingham.

Equipment and Telehealth Monitoring Platform

1. The study will utilize a web-based platform with physiologic devices to support remote telehealth supervision of the intervention. The platform, referred to as Tele Rehab, includes an Android application that is installed on a computer tablet with Bluetooth capability, which sends data and allows two-way communications to a secure webserver. This setup allows a user on an Android computer tablet to communicate in real-time with a member of the research team on a desktop computer. Communication features include videoconference and text-messaging. Heart rate data are recorded and transmitted for real-time view by both the user and research staff, using a heart rate device (Polar Sense, Polar, USA). The heart rate device was placed on a headband and worn on the head to avoid disruption caused by the rapid and vigorous arm movements required for the exercise intervention. Based on internal testing among research staff (non-published findings), placement on the head may result in more accurate readings during intervention exercise than when the device was worn on the forearm or upper arm. The device instructions from the manufacturer state that the Sense can be worn on the head, forearm, or upper arm. The Tele Rehab app system was an upgraded version of one used previously in a tele-monitored feasibility exercise study among people with spinal cord injury [55]. A demonstration of an exercise session in the TeleRehab web-portal, as viewed by a telecoach is displayed in figure 2. Additional equipment to support the safety of the intervention exercise sessions will include:

- 1) Blood pressure monitor (Omron HEM-7200 / BP7200, 5 Series, Omron, USA)
- 2) Medical Alert Device (Monitored Medical Alert System, ADT, USA), a necklace worn device that allows two-way communication between the user and the ADT emergency response team, who is available at all hours of the day to provide remote support for obtaining local emergency response to the user.
- 3) Defibrillator: Philips HeartStart OnSite AED Home

Participants will be trained to use all devices at baseline data collection.

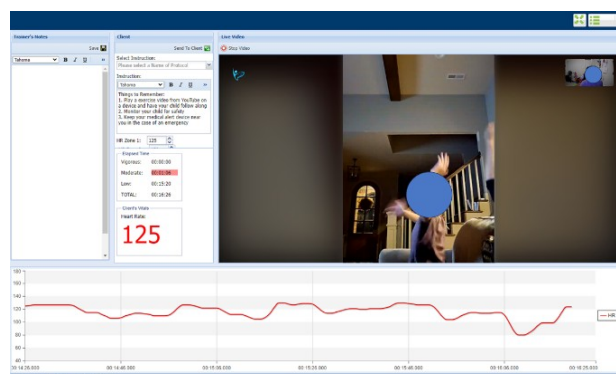


Figure 2. Telecoach view of an exercise session with a participant through the telerehab app.

Procedures

Participants in the immediate start group will complete a 12-week exercise intervention, with two data collection visits to the laboratory (pre- [week 0] and post-intervention [week 13]). Participants in the control group will wait 12 weeks before starting the intervention, and they will also complete a week 0 and week 13 data collection. Before arrival on a data collection visit, participants will be asked to fast for 10-12 hours overnight.

The first data collection visit (week 0) will include the following:

1. Study briefing (purpose, procedures, roles, risks, and benefits)
2. Informed consent and assent documentation
3. Study questionnaires
4. Cardiorespiratory fitness and cardiometabolic outcome measures
5. Red Cross CPR and AED training for the caregiver
6. ADT Subscriber Agreement form (to receive a Medical Alert Device, shipped to the home from ADT)
7. Intervention / Wait Briefing (12-week prescription and equipment instructions)

The second data collection visit (week 13) will include:

1. Study questionnaires
2. Cardiorespiratory fitness and cardiometabolic outcome measures
3. Debriefing (reviewing pre and post changes in outcomes) for the intervention group or intervention briefing for the control group

Measures

Questionnaires

The study will include the following questionnaires:

1. Demographic questionnaire of participant characteristics (e.g., age, sex, BMI, etc.)
2. Godin Leisure-Time Exercise Questionnaire (GLTEQ). The GLTEQ [56] is a 3-item self-report questionnaire that is used to measure physical activity participation. The GLTEQ asks participants to report the number of times that low, moderate, and vigorous intensity of physical activity was performed for longer than 15 minutes in a typical week. The numbers reported for moderate and vigorous exercise intensity are then multiplied by 7 and 9, respectively, and summed for a score, which is referred to as the Health Contribution score. A health contribution score of <24 is classified as physically inactive, whereas a score of ≥ 24 is considered physically active [57]. There is evidence to support the GLTEQ as a valid and reliable measure of physical activity among adults with multiple sclerosis [58] and adolescents [59].

Cardiometabolic Outcomes

Aim 1 primary outcome: peak oxygen consumption (VO₂ peak)

pVO₂ will be measured during a graded exercise test on an arm ergometer using open circuit spirometry with a metabolic cart (TruOne, ParvoMedics). Arm ergometers are considered the gold-standard modality for exercise testing among people with disabilities who use wheelchairs or cannot run or cycle for prolonged periods [60, 61]. Arm ergometry has been suggested as a valid and reliable method for measuring VO₂ peak among children with CP with mild to moderate disability [62]. Prior to starting the test, participants rest for 3-mins. They will then be instructed to maintain a pedaling cadence of 60 revolutions per min. Resistance is increased every min by 10 watts until the participant reaches volitional fatigue or achieves 3 of 5 criteria: age predicted heart rate max of more than 85%; 17 or higher on the Borg rating of perceived exertion (RPE) 6-20 scale; respiratory exchange ratio (RER) of 1.1 or higher; plateau in oxygen consumption; volitional fatigue [63] and/or subjective signs of exhaustion [64]. Heart rate and oxygen consumption will be measured continuously.

Aim 2 outcomes: indicators of cardiometabolic health

Cardiometabolic health will be measured via a dried blood spot test, blood pressure cuff, and DXA scan.

DXA scan: variables will include total mass, total lean mass, tissue % lean, and tissue % fat. Scans will be analyzed using CoreScan software.

Blood Pressure (mmHg): Blood pressure will be measured by a sphygmomanometer. Elevated blood pressure during childhood is associated with intermediate markers and hard outcomes of CVD in adulthood [65]. Small changes in BP (-6.4 mmHg systolic and -4 mmHg diastolic pressure) can occur from endurance training interventions with durations of up to 12 weeks training [66].

Dried blood spot fingerstick test: (ZRT Labs) will be used to assess blood-related cardiometabolic profile and include high sensitivity C-reactive protein (hsCRP), hemoglobin A1c (HbA1C), and fasting insulin, triglycerides, and cholesterol (total, LDL, and HDL). Dried blood spot tests will be shipped for analysis to ZRT Laboratory, Beaverton, OR, USA [67]. The ZRT Lab blood spot test has been conducted with children [68, 69] and has demonstrated excellent validity with venous serum samples (e.g., hsCRP, $r = 0.99$; fasting insulin, $r = 0.93$; fasting triglycerides $r = 0.95$) [67, 70].

hsCRP (mg/L): C-reactive protein is a critical marker of inflammation that contributes to pro-inflammatory and pro-thrombotic elements of CVD risk. A single hsCRP measure is a strong predictor of myocardial infarction or coronary heart disease mortality, and several other diseases of the circulatory system in people without a history of such conditions [71]. Changes in hsCRP may occur from as early as 8-weeks of exercise [72].

HbA1C (mmol/mol): HbA1c is a measure of red blood cell mean hemoglobin glycation over the previous 3 months. A 1-month exercise intervention without a dietary component can expect small to moderate effect [73].

Fasting Insulin (μ U/mL): High fasting insulin indicates the presence of insulin resistance, whether an individual shows glucose intolerance. Exercise interventions without a dietary component can expect a small beneficial change in fasting insulin levels from 1-month of training [73].

Fasting Triglycerides (mg/dL): A triglyceride level >150 mg/dL, is supported as an indicator of CVD risk [74, 75]. Exercise interventions without a dietary component can expect a small beneficial change in triglyceride levels following 1-month of training [73], even among people with normal triglyceride levels [76].

Fasting Cholesterol (mg/dL): Abnormalities in the lipid profile, including high total cholesterol, high LDL cholesterol, and low HDL cholesterol, are predictors of future CVD among young and middle-aged people [77, 78]. Exercise interventions without a dietary component can expect a small effect after 1-month [73].

Aim 3 intervention feasibility

Feasibility will be measured using process metrics that will inform a larger trial [49]. Metrics will include: adherence to the exercise prescription (measured via an attendance log managed by the tele-coaches and Tele Rehab cloud-analytics); technological issues; perceived enjoyment after completing the program will be assessed using the Physical Activity Enjoyment Scale (PACES) [79]; recruitment, enrollment, and attrition rates; and adverse events or problems experienced throughout the intervention.

Home-Based Intervention

The SIT intervention will be performed by participants at home and include 3 sessions of exercise per week, for a total of 12-week. Each SIT session will include 30 repeated sequences of 4-second of maximal arm exercise [38], supplemented with active recovery periods of low-intensity posterior shoulder movements to prevent overuse of anterior muscles. Recovery will be gradually reduced from 30 seconds in week 1, 24 seconds in weeks 2-4, to 15 seconds in weeks 5-12[38]. The total session duration changes from 24 mins in week 1 to 16.5 mins in weeks 6-12. Included within the total session duration is a 5-min warmup and 2-min cooldown. A similar leg exercise protocol was found to be effective for raising VO₂ peak [38], and this dose may be optimal for achieving a high training volume while preventing fatigue due to muscle acidosis [38, 80]. To achieve a maximum intensity, participants are encouraged by the guidance in the videos to perform the 4-second bouts at their maximal effort. Regarding progression, resistance bands (low, medium, heavy) will be gradually introduced into the exercises so that a participant will report a near maximal intensity (≥ 7 on the Borg RPE 0-10 scale; explained in detail in the next section) [81, 82]. Children with CP from a home-based movement-to-music pilot intervention, from which the present study protocol was based on, reported that 20 min videos would be ideal for their attention spans and busy school and care schedules [83]. Sessions will be guided by pre-recorded videos that include an adapted exercise instructor (the PI, BL) and young adult actor with CP (LB) (Figure 3).



Figure 3. Exercise video themes guided by a disability exercise specialist and a young adult with cerebral palsy.

Videos will include 5 themes that were requested by Children with CP from our pilot: 3 themes related to superheroes, one theme for pop music, and one theme for sports. Each theme will include 3 sequential videos that coincide with the rest periods (1 video for week 1; 1 video for weeks 2-4, and 1 video for weeks 5-12), totaling 15 videos. Children with CP can choose to exercise with any themed video that corresponds to their exercise week. Videos include songs that relate to the video themes. Music tempo and volume coincide with sprint and recovery periods. Sprint training incorporates repetitive arm movements that relate to video themes (e.g., Spiderman videos include web shooting; football, arm running/passing). Participants are instructed by the two actors in the videos, who demonstrate and guide participants through the exercises. Videos include CP specific adaptations to enhance engagement [83]: verbal cues, visualized movement adaptations for hemiparesis, slow and repetitive instructions, positive reinforcement, judgement free atmosphere, and imagery. Videos will be archived within a YouTube playlist. Benefits of archiving videos within YouTube (as opposed to custom designed mobile/web app) are that videos are easily accessible on any device, many people are already familiar with YouTube, and this process can easily be replicated. Videos will include themes that are based on copyrighted material, which will only be used for research purposes to adhere to “fair use” for “education” and “research purposes” of the The Fair Use Act Copyright Disclaimer, section 107 of the Copyright Act 1976.

WC participants will be asked to maintain their habitual activity behavior for 12-weeks, and then receive the SIT intervention. All participants will be asked to maintain their habitual diet and eating patterns.

Intervention Development

The SIT intervention was adapted from a clinical adult-exercise program, referred to as Movement-to-Music [84]. The program originated from a clinical efficacy trial, which demonstrated improvements to lower extremity function and fatigue among adults with mobility disabilities [84]. The success of Movement-to-Music led to its implementation in larger confirmatory trials among adults with disabilities [85-87]. In contrast, a pilot feasibility trial found that several aspects of Movement-to-Music required modification to enhance participant engagement among children with CP who had mobility disabilities [69]. Findings from that study informed the development of the present SIT program.

Tele-coach and Fidelity

This study will test a high volume of supervised training to ensure children with CP achieve the desired intensity, before testing a more scalable self-regulated protocol in a larger randomized controlled trial. All three of the weekly prescribed SIT sessions will be supervised via Zoom videoconference by a tele-coach. The tele-coach will support participants in maintaining the maximal effort intensity by monitoring RPE and providing motivational support. Motivational strategies will include positive verbal encouragement to bolster exercise confidence (i.e., self-efficacy) and performing the exercises with the participant to enhance learning through observation (i.e., vicarious learning). These strategies are informed by Social Cognitive Theory [88], and the research team has used these strategies in other trials [55, 83, 86, 89]. To monitor exercise intensity, the tele-coach will ask participants how hard they are working after every change in song (~5 mins) throughout the session. If participants report <7 RPE, coaches/caregivers will provide positive verbal encouragement to enhance motivation. Post-session, participants will report an overall RPE for the entire bout, to determine the need to add resistance bands. RPE has been found to be effective in controlling vigorous exercise intensity in people with spinal cord injury [90]. Since caregiver knowledge and attitude are determinants of participation [91-93], they will be asked to help manage the child's exercise schedule. The tele-coach procedure was framed on Supportive Accountability Theory: building a social relationship and bond with a trusted and knowledgeable health professional, with the inclusion of motivation strategies, can enhance intervention adherence [46].

Analysis

Statistical Power

The chosen sample size of 50 participants permits at least 20 in each of the two study groups at 12-week follow-up after allowing for up to 20% dropout (in each arm). Power calculations were performed using nQuery, version 8.7, for Aims 1 and 2, and SAS, version 9.4, for Aim 3.

For Aims 1 and 2, baseline measurements will be compared with those obtained in week 13. We obtained estimates of the SD for peak oxygen consumption (VO_2 peak) of 7.11 ml/kg/min [64], percent body fat (% BF) of 8.5% [94], total cholesterol (TC) of 27.3 mg/dL [95], systolic blood pressure (SBP) of 14.0 mmHg [95], and diastolic blood pressure (DBP) of 11.7 mmHg [95]. With these assumptions and those of a two-sided two-group t-test, an alpha of 0.05, and 20 participants per group, the study will have at least 80% power to detect between-group differences of at least 6.47 ml/kg/min in VO_2 peak, 7.7% BF, 24.9 mg/dL in TC, 12.8 mmHg in SBP, and 10.7 mmHg in DBP as being statistically significant (at any time point); with the previous assumptions and those of a two-sided paired t-test, an alpha of 0.05, and 20 participants, we will have at least 80% power to detect within-group changes of at least 4.70 ml/kg/min in VO_2 peak, 5.6% BF, 18.1 mg/dL in TC, 9.3 mmHg in SBP, and 7.8 mmHg in DBP as being statistically significant (between any two time points).

For Aim 3, we anticipate adherence to the supervised exercise sessions of the present study will be 75%, based on the 70% adherence found from our pilot, which had non-supervised sessions and had videos that were not designed for adolescents. For an adherence rate of 75%, and assuming 40 participants in total, with 20 per study arm, complete the study, the corresponding exact binomial 95% confidence intervals are (0.588, 0.873) and (0.509, 0.913).

Analyses

For all aims, data analyses will follow intent-to-treat principles. Descriptive statistics will be obtained and reported for study variables. The normality of data distribution of the continuous outcomes will first be confirmed using graphical techniques and tests of normality; continuous

outcomes that deviate greatly from a normal distribution will be adjusted (e.g., transformed) so that the data distribution approximates a normal distribution. All statistical tests will be two-sided. Differences will be considered significant at $p < 0.05$. Statistical analysis will be performed using SAS software (version 9.4). Missing data that are not rectified through ongoing review of source documents may be managed with multiple imputation, and the influence of the missing data will be assessed with sensitivity analyses.

For Aims 1 and 2, our primary method of analysis will be general linear mixed models techniques, such as mixed models repeated measures analyses, as there will be two study groups (SIT and WC) and three time points (baseline, 6 weeks, and 12 weeks). Post hoc analyses will be performed using the Tukey-Kramer multiple comparisons test. These models will allow us to assess the between-group effect, the within-group effect, and the group by time interaction. Covariates to be included in some models include age, sex, and GMFCS level. Analysis of categorical variables between groups will be performed using the chi-square test (or Fisher's exact test, if needed). Pairwise correlations between study parameters will be assessed using Pearson (or Spearman, if needed) correlation analysis.

For Aim 3, we will calculate adherence rates, adverse events rates, and other rates of interest, and corresponding exact binomial 95% confidence intervals. These rates will be calculated overall and stratified by study group. Exploratory comparisons of rates between groups will be performed using the chi-square test (or Fisher's exact test, if needed).

Ethical Considerations

Prior to enrollment of participants, this study was approved by the Institutional Review Board of the University. Written informed consent documentation will be obtained from all participants prior to their engagement in the study. Participants will receive an electronic gift card that will be loaded for \$500 for each of the data collections completed, for a total of up to \$1,000. The rationale for this value is to account for the commitment and time required of both the child and caregiver to complete the study. No artificial intelligence software or program was used in the writing of this manuscript.

Results

This study was approved by the university IRB on Jan 11, 2023. The study was initiated in May 2023, and the first participant was enrolled in October 2023. The study end date is April 30, 2025.

Discussion

Overview

This study will investigate the preliminary efficacy of a home-based, low-cost program for improving cardiorespiratory fitness and metabolic health, which is child appropriate for CP. A program that is short in duration, accessible for various physical abilities, and can be accessed anywhere through the Internet could benefit this population. Cerebral palsy is a low prevalence disability group that is often geographically isolated from suitable clinics, specialists, and adapted exercise services. Moreover, children with CP have limited options for exercise that are evidenced to improve their health [26, 27] and have experienced lower participation in exercise since the outbreak of the coronavirus pandemic [96].

Strengths & Limitations

Considering that this program can be delivered entirely through an Internet video cloud server, YouTube, it has the potential to be carried forward in a scale-up randomized controlled trial of exercise for children with CP. This is important, considering that the average sample size for a randomized controlled trial for children and youth with disabilities has been found to be 27 people, and one of the largest trials included 159 people [27], numbers that are certainly not representative of the diverse needs of children with CP [97]. Another strength of this study is the incorporation of child-appropriate themes and music. Music-based therapy has been found to have an effect on functional ability and goal attainment among children with CP [98]. Child-appropriate themes were requested by this age group and could enhance engagement in therapeutic exercise [83].

This is a preliminary study with a high focus on examining the safety of the intervention and its efficacy under carefully controlled conditions (supervised exercise sessions). Supervised training via telehealth communication provides a sense of accountability that enhances adherence to the intervention prescription [46]. Thus, this study assumes that participants will adhere strongly to the intervention exercise, and the research question is not whether they will perform the exercise, instead the question focuses on how well they will enhance their health with strong attendance. Should this trial be successful, there will be a need to compare the effects of supervised exercise training versus self-regulated, asynchronous training, which will be the primary format that pre-recorded videos will be utilized in real world settings. Supervised training is burdensome for both the research staff and participants.

An additional limitation of this study is the requirement for onsite data collection procedures, which warrants investigation. Onsite visitations are difficult for children with CP and their caregivers due to difficulties with transportation and lack of time with busy school schedules, therapies, and daily activities [5, 99]. There is a need to develop home-based data collection procedures that provide comparable results to those obtained from in-person testing. The blood spot test can be performed remotely by shipping blood spot test kits to the homes of children with CP. However, to the best of our knowledge, there is no home-based alternative method for measuring cardiorespiratory fitness or body composition imaging, particularly among people with mobility disabilities [20].

Conclusions

Should the findings of this study suggest that the program can improve cardiorespiratory fitness or cardiometabolic health, the study may discover an innovative and, most importantly, scalable method of exercise intervention among children with physical disabilities.

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All authors contributed to the final draft of the manuscript and project design. BL and TS created the initial manuscript draft. All authors contributed to either the study design, creation of the study videos, or procedures.

Data Availability

The data sets generated during and/or analyzed during this study will be made publicly available at completion of the study and can be obtained from the corresponding author on reasonable request.

Conflicts of Interest

None declared.

Abbreviations

CP: cerebral palsy

SIT: sprint-intensity interval training

VO2 peak: peak oxygen consumption

References

1. Gulati, S. and V. Sondhi, *Cerebral Palsy: An Overview*. Indian J Pediatr, 2018. **85**(11): p. 1006-1016. PMID:32053326
2. *The Definition and Classification of Cerebral Palsy*. Dev Med Child Neurol, 2007. **49**(s109): p.

- 1-44. PMID:17371509
3. Mutch, L., et al., *Cerebral palsy epidemiology: where are we now and where are we going?* Dev Med Child Neurol, 1992. **34**(6): p. 547-51. PMID:1612216
 4. Rimmer, J.H., *Health promotion for people with disabilities: the emerging paradigm shift from disability prevention to prevention of secondary conditions.* Phys Ther, 1999. **79**(5): p. 495-502. PMID:10331753
 5. Martin Ginis, K.A., et al., *A systematic review of review articles addressing factors related to physical activity participation among children and adults with physical disabilities.* Health Psychol Rev, 2016. **10**(4): p. 478-494. PMID:27265062
 6. Heyn, P.C., et al., *Prevalence of metabolic syndrome and cardiovascular disease risk factors in adults with cerebral palsy.* Dev Med Child Neurol, 2019. **61**(4): p. 477-483. PMID:30663044
 7. McPhee, P.G., et al., *Associations of non-invasive measures of arterial structure and function, and traditional indicators of cardiovascular risk in adults with cerebral palsy.* Atherosclerosis, 2015. **243**(2): p. 462-5. PMID:26520900
 8. Ryan, J.M., et al., *Mortality due to cardiovascular disease, respiratory disease, and cancer in adults with cerebral palsy.* Developmental Medicine & Child Neurology, 2019. **61**(8): p. 924-928.
 9. Mcphee, P.G., et al., *Cardiovascular disease and related risk factors in adults with cerebral palsy: a systematic review.* Dev Med Child Neurol, 2019. **61**(8): p. 915-923. PMID: 30221763
 10. McPhee, P.G., *Cardiovascular disease in cerebral palsy: shifting our focus from attention to prevention.* Dev Med Child Neurol, 2019. **61**(4): p. 390-391. PMID:30761526
 11. van der Slot, W.M., et al., *Cardiovascular disease risk in adults with spastic bilateral cerebral palsy.* J Rehabil Med, 2013. **45**(9): p. 866-72. PMID:23828200
 12. Peterson, M.D., N. Kamdar, and E.A. Hurvitz, *Age-related trends in cardiometabolic disease among adults with cerebral palsy.* Dev Med Child Neurol, 2019. **61**(4): p. 484-489. PMID:29704244
 13. Ryan, J.M., et al., *Habitual physical activity and cardiometabolic risk factors in adults with cerebral palsy.* Res Dev Disabil, 2014. **35**(9): p. 1995-2002. PMID:24864052
 14. Slaman, J., et al., *Can a lifestyle intervention improve physical fitness in adolescents and young adults with spastic cerebral palsy? A randomized controlled trial.* Arch Phys Med Rehabil, 2014. **95**(9): p. 1646-55. PMID: 25067790
 15. Thorpe, D., *The role of fitness in health and disease: status of adults with cerebral palsy.* Dev Med Child Neurol, 2009. **51 Suppl 4**: p. 52-8. PMID:19740210
 16. Waltersson, L. and E. Rodby-Bousquet, *Physical Activity in adolescents and young adults with cerebral palsy.* Biomed Res Int, 2017. **2017**: p. 8080473. PMID:29423412
 17. Shkedy Rabani, A., et al., *Duration and patterns of habitual physical activity in adolescents and young adults with cerebral palsy.* Dev Med Child Neurol, 2014. **56**(7): p. 673-680. PMID:24506509
 18. Carlon, S.L., et al., *Differences in habitual physical activity levels of young people with cerebral palsy and their typically developing peers: a systematic review.* Disabil Rehabil., 2013. **35**(8): p. 647-55. PMID:23072296
 19. McPhee, P.G., et al., *Descriptive data on cardiovascular and metabolic risk factors in ambulatory and non-ambulatory adults with cerebral palsy.* Data Brief, 2015. **5**: p. 967-70. PMID:26759816
 20. Novak, I., et al., *Clinical prognostic messages from a systematic review on cerebral palsy.* Pediatrics, 2012. **130**(5): p. e1285-312. PMID:23045562

21. Andersson, C., Mattsson, E, *Adults with cerebral palsy: a survey describing problems, needs, and resources, with special emphasis on locomotion*. Dev Med Child Neurol, 2001. **43**(2): p. 76. PMID:11221908
22. Opheim, A., et al., *Walking function, pain, and fatigue in adults with cerebral palsy: a 7-year follow-up study*. Dev Med Child Neurol, 2009. **51**(5): p. 381-388. PMID:19207296
23. Rimmer, J.H., M.D. Chen, and K. Hsieh, *A conceptual model for identifying, preventing, and managing secondary conditions in people with disabilities*. Phys Ther, 2011. **91**(12): p. 1728-39. PMID:22003160
24. Parkinson, K.N., et al., *Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study*. Arch Dis Child, 2013. **98**(6): p. 434-440. PMID:23606716
25. Alriksson-Schmidt, A. and G. Hägglund, *Pain in children and adolescents with cerebral palsy: a population-based registry study*. Acta Paediatr, 2016. **105**(6): p. 665-670. PMID:26880375
26. Lai, B., et al., *Leisure-time physical activity interventions for children and adults with cerebral palsy: a scoping review*. Dev Med Child Neurol, 2020. PMID:33241561
27. Lai, B., et al., *Research trends and recommendations for physical activity interventions among children and youth with disabilities: a review of reviews*. Adapt Phys Activ Q, 2020. **37**(2): p. 211-234. PMID:32131052
28. Ryan, J.M., et al., *Exercise interventions for cerebral palsy*. Cochrane Database Syst Rev., 2017. **6**(6):CD011660. PMID: 28602046
29. Reedman, S., R.N. Boyd, and L. Sakzewski, *The efficacy of interventions to increase physical activity participation of children with cerebral palsy: a systematic review and meta-analysis*. Dev Med Child Neurol, 2017. **59**(10): p. 1011-1018. PMID:28318009
30. Verschuren, O., et al., *Exercise and physical activity recommendations for people with cerebral palsy*. Dev Med Child Neurol, 2016. **58**(8): p. 798-808. PMID:26853808
31. Costigan, S.A., et al., *High-intensity interval training for improving health-related fitness in adolescents: a systematic review and meta-analysis*. Br J Sports Med, 2015. **49**(19): p. 1253. PMID:26089322
32. Ramos, J.S., et al., *The impact of high-intensity interval training versus moderate-intensity continuous training on vascular function: a systematic review and meta-analysis*. Sports Med, 2015. **45**: p. 679-692. PMID:25771785
33. Rosenblat, M.A., A.S. Perrotta, and S.G. Thomas, *Effect of high-intensity interval training versus sprint interval training on time-trial performance: a systematic review and meta-analysis*. Sports Med, 2020. **50**(6): p. 1145-1161. PMID:32034701
34. Gist, N.H., et al., *Sprint interval training effects on aerobic capacity: a systematic review and meta-analysis*. Sports Med, 2014. **44**(2): p. 269-279. PMID:32034701
35. Sun, S., et al., *Twelve weeks of low volume sprint interval training improves cardio-metabolic health outcomes in overweight females*. J Sports Sci, 2019. **37**(11): p. 1257-1264. PMID:30563431
36. Cassidy, S., et al., *High-intensity interval training: a review of its impact on glucose control and cardiometabolic health*. Diabetologia, 2017. **60**(1): p. 7-23. PMID:27681241
37. Gaitanos, G.C., et al., *Human muscle metabolism during intermittent maximal exercise*. J Appl Physiol (1985), 1993. **75**(2): p. 712-9. PMID:8226473
38. Satioglu, R., et al., *Four-Second Power Cycling Training Increases Maximal Anaerobic Power, Peak Oxygen Consumption, and Total Blood Volume*. Med Sci Sports Exerc, 2021. PMID:34310498
39. Mairbäurl, H., *Red blood cells in sports: effects of exercise and training on oxygen supply by*

- red blood cells. *Front Physiol*, 2013. **4**: p. 332. PMID:24273518
40. McMurray, R.G., et al., *Is physical activity or aerobic power more influential on reducing cardiovascular disease risk factors?* *Med Sci Sports Exerc*, 1998. **30**(10): p. 1521-9. PMID:9789853
 41. García, C.C., et al., *Metabolic, cardiorespiratory, and neuromuscular fitness performance in children with cerebral palsy: a comparison with healthy youth.* *J Exerc Rehabil*, 2016. **12**(2): p. 124-31. PMID:27162775
 42. Lauglo, R., et al., *High-intensity interval training to improve fitness in children with cerebral palsy.* *BMJ Open Sport Exerc Med*, 2016. **2**(1): p. e000111-e000111. PMID:27900177
 43. Zinner, C., et al., *The physiological mechanisms of performance enhancement with sprint interval training differ between the upper and lower extremities in humans.* *Front Physiol*, 2016. **7**: p. 426. PMID:27746738
 44. Langkamp, D.L., M.D. McManus, and S.D. Blakemore, *Telemedicine for children with developmental disabilities: a more effective clinical process than office-based care.* *Telemed J E Health*, 2015. **21**(2): p. 110-4. PMID:25545598
 45. Harper, D.C., *Telemedicine for children with disabilities.* *Children's Health Care*, 2006. **35**(1): p. 11-27. doi:10.1207/s15326888chc3501_3
 46. Mohr, D.C., P. Cuijpers, and K. Lehman, *Supportive accountability: a model for providing human support to enhance adherence to eHealth interventions.* *J Med Internet Res*, 2011. **13**(1): p. e30-e30. PMID:21393123
 47. Lai, B., et al., *Current trends in exercise intervention research, technology, and behavioral change strategies for people with disabilities: a scoping review.* *Am J Phys Med Rehabil*, 2017. **96**(10): p. 748-761. PMID: 28398967
 48. Ma, J.K. and K.A.M. Ginis, *A meta-analysis of physical activity interventions in people with physical disabilities: content, characteristics, and effects on behaviour.* *Psychology of Sport and Exercise*, 2018. **37**: p. 262-273. doi:10.1016/j.psychsport.2018.01.006
 49. Tickle-Degnen, L., *Nuts and bolts of conducting feasibility studies.* *Am J Occup Ther*, 2013. **67**(2): p. 171-176. PMID:23433271
 50. Liguori G, *American College of Sports Medicine, ACSM's guidelines for exercise testing and prescription.* 11th Ed. 2020: Lippincott Williams & Wilkins. ISBN:9781975150211
 51. Palisano, R., et al., *Development and reliability of a system to classify gross motor function in children with cerebral palsy.* *Dev Med Child Neurol*, 1997. **39**(4): p. 214-23. PMID:9183258
 52. Morris, C., B.E. Galuppi, and P.L. Rosenbaum, *Reliability of family report for the gross motor function classification system.* *Dev Med Child Neurol*, 2004. **46**(7): p. 455-460. PMID:15230458
 53. Morris, C., et al., *Who best to make the assessment? Professionals' and families' classifications of gross motor function in cerebral palsy are highly consistent.* *Arch Dis Child*, 2006. **91**(8): p. 675-679. PMID:16638783
 54. Jewell, A.T., A.I. Stokes, and D.J. Bartlett, *Correspondence of classifications between parents of children with cerebral palsy aged 2 to 6 years and therapists using the Gross Motor Function Classification System.* *Dev Med Child Neurol*, 2011. **53**(4): p. 334-337. PMID:21166672
 55. Lai, B., et al., *Teleexercise for persons with spinal cord injury: a mixed-methods feasibility case series.* *JMIR Rehabil Assist Technol*, 2016. **3**(2): p. e8. PMID:28582252
 56. Godin G, Shephard RJ. *Leisure Time Exercise Questionnaire.* *Canadian J Appl Sport Science*. 1997. doi:10.1037/t31334-000
 57. Amireault, S. and G. Godin, *The Godin-Shephard leisure-time physical activity questionnaire:*

- validity evidence supporting its use for classifying healthy adults into active and insufficiently active categories. *Percept Mot Skills*, 2015. **120**(2): p. 604-22. PMID:25799030
58. Sikes, E.M., et al., *Use of the Godin leisure-time exercise questionnaire in multiple sclerosis research: a comprehensive narrative review*. *Disabil Rehabil*, 2019. **41**(11): p. 1243-1267. PMID:29343122
59. Zelener, J. and M. Schneider, *Adolescents and self-reported physical activity: an evaluation of the modified Godin Leisure-Time Exercise Questionnaire*. *Int J Exerc Sci*, 2016. **9**(5): p. 587-598. PMID:29177023
60. Larsen, R.T., et al., *A systematic review and meta-analysis comparing cardiopulmonary exercise test values obtained from the arm cycle and the leg cycle respectively in healthy adults*. *Int J Sports Phys Ther*, 2016. **11**(7): p. 1006-1039. PMID:27999717
61. Eerden, S., R. Dekker, and F.J. Hettinga, *Maximal and submaximal aerobic tests for wheelchair-dependent persons with spinal cord injury: a systematic review to summarize and identify useful applications for clinical rehabilitation*. *Disabil Rehabil*, 2018. **40**(5): p. 497-521. PMID: 28637157
62. Brehm, M.A., et al., *Reliability of a progressive maximal cycle ergometer test to assess peak oxygen uptake in children with mild to moderate cerebral palsy*. *Phys Ther*, 2014. **94**(1): p. 121-8. PMID:24029296
63. Thompson, P.D., et al., *ACSM's new preparticipation health screening recommendations from ACSM's guidelines for exercise testing and prescription*. *Curr Sports Med Rep*, 2013. **12**(4): p. 215-217. PMID: 23851406
64. Balemans, A.C., et al., *Maximal aerobic and anaerobic exercise responses in children with cerebral palsy*. *Med Sci Sports Exerc*, 2013. **45**(3): p. 561-568. PMID:23034639
65. Yang, L., et al., *Elevated blood pressure in childhood or adolescence and cardiovascular outcomes in adulthood*. *Hypertension*, 2020. **75**(4): p. 948-955. PMID:32114851
66. Cornelissen Veronique, A. and A. Smart Neil, *Exercise training for blood pressure: a systematic review and meta-analysis*. *J Am Heart Assoc*. **2**(1): p. e004473. PMID:23525435
67. Kapur, S., S. Kapur, and D. Zava, *Cardiometabolic risk factors assessed by a finger stick dried blood spot method*. *J Diabetes Sci Technol*, 2008. **2**(2): p. 236-241. PMID:19885348
68. Larkin, E.K., et al., *Agreement of blood spot card measurements of vitamin D levels with serum, whole blood specimen types and a dietary recall instrument*. *PloS one*, 2011. **6**(1): p. e16602-e16602. PMID:21304997
69. Lai, B., et al., *The effects of virtual reality tele-exergaming on cardiometabolic indicators of health among youth with cerebral palsy: protocol for a pilot randomized controlled trial*. *JMIR Res Protoc*, 2022. **11**(8): p. e40708. PMID:35976192
70. Kapur, S., et al., *Postprandial insulin and triglycerides after different breakfast meal challenges: use of finger stick capillary dried blood spots to study postprandial dysmetabolism*. *J Diabetes Sci Technol*, 2010. **4**(2): p. 236-43. PMID:20307382
71. Bassuk, S.S., N. Rifai, and P.M. Ridker, *High-sensitivity C-reactive protein: Clinical importance*. *Curr Probl Cardiol*, 2004. **29**(8): p. 439-493. PMID:15258556
72. Ford, E.S., *Does exercise reduce inflammation? Physical activity and C-reactive protein among U.S. Adults*. *Epidemiology*, 2002. **13**(5): p. 561-568. PMID:12192226
73. Snowling, N.J. and W.G. Hopkins, *Effects of Different Modes of Exercise Training on Glucose Control and Risk Factors for Complications in Type 2 Diabetic Patients*. *Diabetes Care*, 2006. **29**(11): p. 2518. PMID:17065697
74. Singh, A.K. and R. Singh, *Triglyceride and cardiovascular risk: A critical appraisal*. *Indian J Endocrinol Metab*, 2016. **20**(4): p. 418-428. PMID:27366705

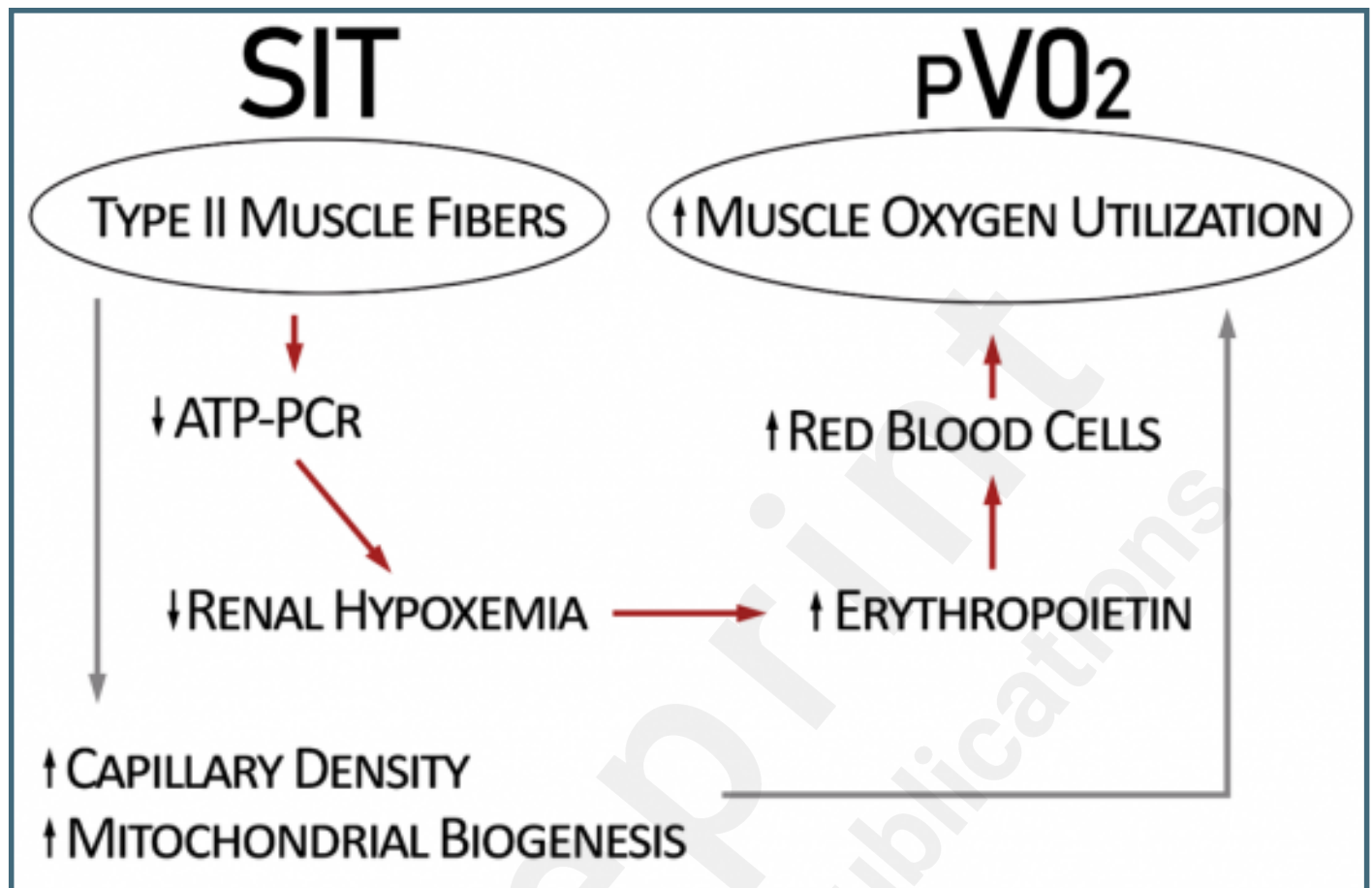
75. Lee, J.S., et al., *Triglyceride and HDL-C dyslipidemia and risks of coronary heart disease and ischemic stroke by glycemic dysregulation status: the Strong Heart study*. *Diabetes Care*, 2017. **40**(4): p. 529. PMID:28122840
76. Igarashi, Y., N. Akazawa, and S. Maeda, *Effects of aerobic exercise alone on lipids in healthy east Asians: a systematic review and meta-analysis*. *J Atheroscler Thromb*, 2019. **26**(5): p. 488-503. PMID:30381613
77. Peters, S.A., et al., *Total cholesterol as a risk factor for coronary heart disease and stroke in women compared with men: a systematic review and meta-analysis*. *Atherosclerosis*, 2016. **248**: p. 123-31. PMID:27016614
78. Ravnskov, U., et al., *LDL-C does not cause cardiovascular disease: a comprehensive review of the current literature*. *Expert Rev Clin Pharmacol*, 2018. **11**(10): p. 959-970. PMID:30198808
79. Kendzierski, D. and K.J. DeCarlo, *Physical activity enjoyment scale: Two validation studies*. *J Sport Exerc Psychol*, 1991. **13**(1). doi:10.1123/jsep.13.1.50
80. Buchheit, M. and P.B. Laursen, *High-intensity interval training, solutions to the programming puzzle: Part I: cardiopulmonary emphasis*. *Sports Med*, 2013. **43**(5): p. 313-38. PMID:23539308
81. Williams, N., *The Borg Rating of Perceived Exertion (RPE) scale*. *Occupational Medicine*, 2017. **67**(5): p. 404-405. doi:10.1093/occmed/kqx063
82. Borg, G., *Borg's perceived exertion and pain scales*. 1998: Human kinetics. ISBN:9780880116237
83. Lai, B., et al., *Telehealth Movement-to-Music to increase physical activity participation among adolescents with cerebral palsy: pilot randomized controlled trial*. *JMIR Form Res*, 2022. **6**(10): p. e36049. PMID:36306154
84. Young, H.J., et al., *The Effects of a Movement-to-Music (M2M) intervention on physical and psychosocial outcomes in people poststroke: a randomized controlled trial*. *Arch Rehabil Res Clin Transl*, 2021. **3**(4): p. 100160. PMID:34977542
85. Young, H.-J., et al., *The movement-to-music (M2M) study: study protocol for a randomized controlled efficacy trial examining a rhythmic teleexercise intervention for people with physical disabilities*. *Trials*, 2021. **22**(1): p. 779. PMID:34743701
86. Rimmer, J.H., et al., *Rationale and design of a Scale-Up Project Evaluating Responsiveness to Home Exercise And Lifestyle Tele-Health (SUPER-HEALTH) in people with physical/mobility disabilities: a type 1 hybrid design effectiveness trial*. *BMJ Open*, 2019. **9**(3): p. e023538. PMID:30928927
87. Wilroy, J., et al., *Scale Up Project Evaluating Responsiveness to Home Exercise And Lifestyle Tele-Health (SUPER-HEALTH): preliminary findings*. *Arch Phys Med Rehabil*, 2022. **103**(12): p. e121. PMID:30928927
88. Bandura, A., *Health promotion by social cognitive means*. *Health Educ Behav*, 2004. **31**(2): p. 143-64.
89. Lai, B., et al., *Exploring the uptake and implementation of tele-monitored home-exercise programmes in adults with Parkinson's disease: A mixed-methods pilot study*. *J Telemed Telecare*, 2018: p. 1357633X18794315. PMID:30134777
90. Goosey-Tolfrey, V., et al., *Regulating intensity using perceived exertion in spinal cord-injured participants*. *Med Sci Sports Exerc*, 2010. **42**(3): p. 608-13. PMID:19952816
91. Bloemen, M.A., et al., *Factors associated with physical activity in children and adolescents with a physical disability: a systematic review*. *Dev Med Child Neurol*, 2015. **57**(2): p. 137-48. PMID:25403649
92. Verschuren, O., et al., *Identification of facilitators and barriers to physical activity in children*

- and adolescents with cerebral palsy. *J Pediatr*, 2012. **161**(3): p. 488-94. PMID:22494875
93. Verschuren, O., et al., *Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial*. *Arch Pediatr Adolesc Med*, 2007. **161**(11): p. 1075-81. PMID:1798441
94. Finbråten, A.K., et al., *Assessment of body composition in children with cerebral palsy: a cross-sectional study in Norway*. *Dev Med Child Neurol*, 2015. **57**(9): p. 858-864. PMID:25827868
95. Barja, S., et al., *Obesity and cardio-metabolic risk factors among children and adolescents with cerebral palsy*. *Nutr Hosp*, 2020. **37**(4): p. 685-691. PMID: 32686452
96. Lai, B., et al., *The impact of COVID-19 on the lifestyles of adolescents with cerebral palsy in the Southeast United States*. *Disabil Health J*, 2022. **15**(2): p. 101263. PMID:35000878
97. McIntyre, S., et al., *Global prevalence of cerebral palsy: A systematic analysis*. *Dev Med Child Neurol*, 2022. **64**(12): p. 1494-1506. PMID:35952356
98. Yanagiwara, S., et al., *Effects of music therapy on functional ability in people with cerebral palsy: a systematic review*. *J Rural Med*, 2022. **17**(3): p. 101-107. PMID:35847757
99. Shields, N., A.J. Synnot, and M. Barr, *Perceived barriers and facilitators to physical activity for children with disability: a systematic review*. *Br J Sports Med.*, 2012. **46**(14): p. 989-97. doi: PMID:2194812

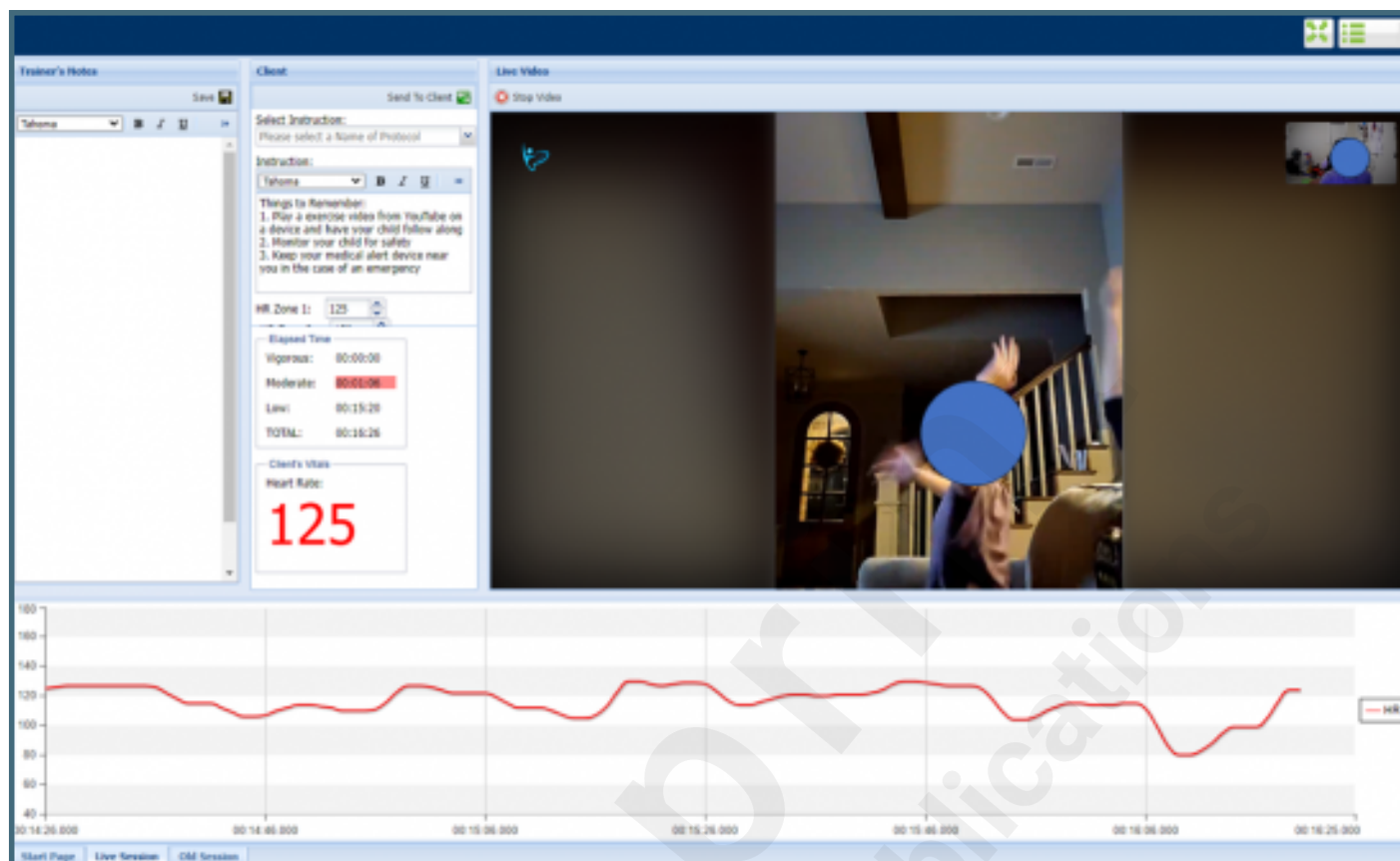
Supplementary Files

Figures

Conceptual model for the mechanism that underpins sprint-intensity interval training.



Telecoach view of an exercise session with a participant through the telerehab app.



Exercise video themes guided by a disability exercise specialist and a young adult with cerebral palsy.



Multimedia Appendixes

Evidence of protocol review.

URL: <http://asset.jmir.pub/assets/654e1ca9d756cd8f44c80154e559018c.pdf>



TOC/Feature image for homepages

Exercise video demonstration.

