The effect of RaceRunning on cardiometabolic disease risk factors and functional mobility in young people with moderate-to-severe cerebral palsy: protocol for a feasibility study.

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Word count: 3589

Abstract

Introduction: There is consistent evidence that people with cerebral (CP) do not engage in the recommended physical activity guidelines for the general population from a young age. Participation in moderate-to-vigorous physical activity is particularly reduced in people with CP who have moderate-to-severe disability. RaceRunning is a growing disability sport that provides an opportunity for people with moderate-to-severe disability to participate in physical activity in the community. It allows those who are unable to walk independently, to propel themselves using a RaceRunning bike, which has a breastplate for support but no pedals. The aim of this study is to examine the feasibility and acceptability of RaceRunning for young people with moderate-to-severe CP and the feasibility of conducting a definitive study of the effect of RaceRunning on cardiometabolic disease risk factors and functional mobility.

Methods and analysis: Twenty-five young people (age 5-21 yr) with CP or acquired brain injury affecting co-ordination will be included in this single arm intervention study. Participants will take part in one RaceRunning session each week for 24 weeks. Outcomes assessed at baseline, 12 and 24 weeks include body mass index, waist circumference, blood pressure, muscle strength, cardiorespiratory fitness, physical activity and sedentary behaviour, functional mobility, activity competence and psychosocial impact. Adverse events will be systematically recorded throughout the 24 weeks. Focus groups will be conducted with participants and/or parents to explore their views and experiences of taking part in RaceRunning.

Ethics and dissemination: Approval has been granted by Queen Margaret University Research Ethics Committee (REC) and the South East of Scotland REC. Results will be disseminated through peer-reviewed journals and distributed to people with CP and their families through RaceRunning and Athletic Clubs, NHS trusts, and organisations for people with disabilities.

Trial registration number: ClinicalTrials.gov Identifier: NCT04034342. Protocol version 1.0; pre-results.

Keywords: paediatric neurology; developmental neurology and neurodisability; preventive medicine; rehabilitation medicine; sports medicine

Strengths and limitations of this study

- This multi-centre study will examine the feasibility and acceptability of RaceRunning for young people with moderate-to-severe cerebral palsy as well as the feasibility of conducting a definitive study of the effect of RaceRunning.
- People with a range of severities of motor impairment will be included.
- A range of outcomes relating to cardiometabolic risk will be assessed.
- There will be no control group included in the study.

Introduction

Cerebral palsy (CP) is an umbrella term for disorders affecting the development of movement and posture, which cause limitations in activities of daily living. Although CP is considered a non-progressive neurological condition, it often results in secondary conditions such as contractures, bone deformities, muscle weakness and fatigue leading to further decreased mobility.[1,2] Mobility often worsens with age and can lead to diminished independence.[3] These secondary conditions may lead to a negative cycle of physical inactivity and further deconditioning.[4,5] Indeed, there is ample evidence that, from a young age, people with CP do not engage in the recommended activity levels for the general population and children with CP of 60 minutes moderate-to-vigorous physical activity (MVPA) per day.[4] Participation in MPVA is particularly reduced in people with CP who have moderate-to-severe disability. Claridge et al. (2015) reported that children in GMFCS level I participated in 40min of MVPA per day and this decreased with increasing GMFCS level to 5.5 minutes, 0.71 minutes and 0.64 minutes for children in GMFCS levels III, IV and V.[6] Reduced participation in MVPA is a modifiable risk factor for non-communicable diseases (WHO) and likely contributes to the increased risk of cardiovascular disease observed in people with CP.[4] In both adults and children with CP, participation in MVPA is associated with reductions in risk factors for cardiometabolic disease such as elevated blood pressure and abdominal obesity.[7,8] Further, breaks from sedentary behaviour are associated with reductions in risk factors, independent of total time in MVPA in people without disability,[9] which may be feasible for children with CP to achieve.

In recent years, efforts have been directed towards examining the effectiveness of exercise and physical activity interventions for independently ambulant people with CP.[10,11] However, there is a lack of research examining the effectiveness of exercise interventions for people with CP with moderate-to-severe disability.[10] Limited access to adapted physical activities may contribute to lack of participation in MVPA among people with moderate-to-severe disability. RaceRunning (www.racerunning.org) is a growing disability sport that provides an opportunity for people with moderate-to-severe disability to participate in physical activity in the community. It allows those who are unable to walk independently, to propel themselves using a RaceRunning bike, which has a breastplate for support but no pedals. Participants sit on the saddle and use their legs to propel themselves forward. It is estimated that worldwide, approximately 2000 running bikes are used for participation in sport (i.e. training sessions and competitions), physical education and therapy. Pilot studies suggest that people with CP can achieve MVPA whilst taking part in RaceRunning.[12,13] Non-controlled small studies also indicate that RaceRunning has positive effects on aerobic capacity, bone health and muscle thickness.[14,15] However, the quality of the evidence from these studies is low and no study has assessed the long-term sustainability of RaceRunning or its impact on cardiometabolic risk factors and functional mobility in people with CP. Before conducting a definitive cohort study on the effect of RaceRunning, the feasibility of conducting such a study needs to be determined.

The aim of this study is to examine the feasibility and acceptability of RaceRunning for young people with moderate-to-severe CP and the feasibility of conducting a definitive study of the effect of RaceRunning on cardiometabolic disease risk factors and functional mobility.

Methods and analysis

Study design

This is a multi-centre single arm intervention study. Assessments will be conducted at baseline, 12 weeks and 24 weeks. Participants will be recruited from RaceRunning clubs, athletics clubs and NHS trusts in Scotland and Gloucestershire. The study will also be advertised through social media and via organisations for people with disabilities. Potentially eligible participants will be provided with an invitation letter and an age appropriate Participant Information Sheet. Parents of potential participants <16 years will be provided with a parent information sheet.

Participants

Twenty-five young people aged 5-21 years with CP or acquired brain injury affecting co-ordination will be included in the study. Participants are eligible to be included in the trial if they use wheeled mobility indoors and/or outdoors but they may use an assistive mobility device indoors instead of a wheel chair. For children with CP, this encompasses children in Gross Motor Function Classification System (GMFCS) levels III, IV or V. Additional inclusion criteria are: having less than 15 hours of RaceRunning experience, ability to independently propel the bike for at least 30 meters and ability to comprehend and follow instructions relating to participation in RaceRunning training. Participants will be excluded if they had lower limb surgery or Selective Dorsal Rhizotomy less than 6 months prior to the start of the study or have a severe visual impairment affecting the ability to safely take part in RaceRunning training. Children and young people who have been receiving regular Botox injections and Baclofen treatment for more than 6 months will be included in the study but these will be recorded in the trial notes.

Intervention

Participants will take part in one RaceRunning session each week for 24 weeks. This hourly session will be led by an experienced RaceRunning coach, who has attended the RaceRunning coaching course (organised by RaceRunning Scotland or CP Sport England) and/or is a level 1 or 2 UK athletics coach. The coach will be supported by a physiotherapist or other health professional and (student) volunteers.

Content of the sessions will be standardised for all training groups and will consist of warm-up, coordination, endurance and sprint training drills, and a cool-down period. Co-ordination training will be approximately 10 minutes in duration and consist of drills such as mouse steps (feet touching the ground as many times as possible), over exaggerated strides, one foot pushes, high knees and two foot pushes. Endurance training will be approximately 10-15 minutes duration and consist of steady running. Sprint training will consist of 4-5 short sprints over between 10m and 60m, depending on ability.

Outcome measures

Demographic, CP-related characteristics and self-reported physical activity will be assessed at baseline. The Physical Activity Questionnaire for children (PAQ-C) and adolescents (PAQ-A) will be used to assess self-reported physical activity. Both questionnaires provide a summary of physical activity score derived from nine items, each scored on a 5-point scale. The PAQ-C and PAQ-A are valid and reliable in children.[16-18]

The following outcomes will be assessed at baseline, 12 weeks and 24 weeks. Questionnaires will be completed at Queen Margaret University or the University of Gloucestershire, or at the participant's home if requested. Objective measures will be assessed at the location where RaceRunning sessions are completed. Participants will be asked to refrain from strenuous or unusual exercise 24 hours before the assessment and to have a light meal at least 2 hours before the assessment of objective measures. Assessors will follow a standardised protocol to complete all assessments.

Body mass index

Body mass will be measured to the nearest kg using flat medical scales (Seca). For those not able to stand independently, weight will be recorded while sitting on a chair on the scales. For participants able to stand (with or without support), body height will be measured (to the nearest mm) in standing using a portable stadiometer. For those who are unable to stand unsupported, a height measurement will be taken supine, also using a portable stadiometer. In case of severe contractures in both legs, knee height will be measured and used to estimate the height using published, validated equations based on a population of children with CP.[19] Body mass index (BMI) will calculated as mass divided by height squared (kg/m^2).

Waist circumference

For waist circumference, midpoints between the lower rib margin and the iliac crest will be marked and the circumference will be measured to the nearest mm by positioning the measuring tape over the mid-distance mark on both sides. This method has shown to have adequate reliability and measurement error in children.[20]

Thigh and calf circumference

For an estimation of the muscle bulk, a tape measure will be used to measure thigh and calf circumference to the nearest mm. For the calf, the location of the circumference measurement will be in the most prominent point of the muscle belly. For the thigh, the measurement point will be at 50% on the line from the anterior superior iliac spine to the superior part of the patella. The average of three measurement will be used for analysis.

Resting heart rate and blood pressure

Heart rate will be assessed using SunTech Tango M2 in DKA mode for recording of resting ECG. Three chest ECG electrodes will be used to record resting ECG. If this is not feasible (e.g. lack of cooperation of the young person), resting heart rate will be recorded through a chest belt.

Blood pressure will be assessed using an automated blood pressure monitor with an appropriately sized cuff. The cuff, with integrated microphone for the automatic detection of the Kortkoff sound, will be placed on the left arm with the centre of the bladder over the brachial artery. The bladder will encircle at least 80% of the arm but not more than 100%. The participant will be asked to sit quietly for between 5 and 10 minutes in an upright position with his/her back against the chair and the measurement arm supported at the level of the heart.

After sitting quietly for at least 5 minutes, resting blood pressure and heart rate will be recorded in triplicate, with a 1-min interval between measurements. The average of the 3 readings of systolic blood pressure (mmHg), diastolic blood pressure (mmHg) and heart rate (beats per minute) will be used in analysis. Efforts will be made to assess blood pressure and heart rate at a similar time of day at all assessments. Assessment will take place in a private, quiet, well-ventilated room of adequate size.

Knee extensor muscle strength

Knee extensor strength will be assessed using digital myometry in sitting with the knee flexed in 90 degrees. This test will be repeated twice with each leg. It is a reliable method of measuring strength in children with CP.[21]

Cardiorespiratory fitness

There are currently no validated measures of maximum aerobic capacity for people with moderate-to-severe CP who are non-ambulant and/or unable to propel a wheelchair. For this research a RaceRunning specific field-based maximal incremental test for determining peak oxygen consumption ($\dot{V}O_{2peak}$) will be used. The test is based on existing field-based incremental protocols for young people with CP.[22] Instead of 10 metre shuttles, as in the original tests, cones will be placed around the track every 10 meters, which will avoid the participants having to do a 180 degree turn. Participants will walk/run between each cone at a set incremental speed determined by a signal. Those participants who are able to complete 100m in less than 40 seconds will perform an adapted shuttle run test for GMFCS level I (Shuttle RaceRunning test I, SRRT-1), others will perform the adapted shuttle run test for GMFCS level II (Shuttle RaceRunning test II SRRT-II). Both tests consist of 23 levels each lasting one minute with an increase in speed of 0.25km/h at each level. The starting speed of the SRRT-I is 5km/h and the starting speed of the SRRT-II is 2km/h. The test will end upon reaching volitional exhaustion. If participants do not complete one shuttle within the allocated time, they will be encouraged to continue. The test will be terminated if the participants does not complete two consecutive shuttles within the allocated time.

Expired gas exchange data will be collected in real-time throughout the test using a portable online gas analyser (Metamax 3B (CORTEX Biophysik GmbH, Germany)). Heart rate will be monitored throughout the test using a chest-based transmitter (H10, Polar) and recorded by the Metamax software at a sample rate of 60Hz.

The test protocol described above allows direct measurement of the following outcomes: $\dot{V}O_{2peak}$, ventilatory threshold, minute ventilation, breathing frequency, sub-maximal and maximal heart rate, respiratory exchange ratio, ventilatory efficiency ratios and O_2 pulse. $\dot{V}O_{2peak}$ will be determined as the highest observed value of $\dot{V}O_2$ over a 10 second epoch during the last completed 20 seconds in the final stage of the test. In addition, we will assess work efficiency at different exercise intensities that will correspond to a complete stage of the SRRT.

Physical activity and sedentary behaviour

Steps per day, time spent in an upright position per day, number of transfers per day and time spent in sitting and lying (i.e., sedentary time) will be recorded using the activPAL3µ activity monitor. The activPAL3µ is a small, lightweight device that is worn on the anterior aspect of the person's thigh. It incorporates accelerometry and inclinometry data to provide information on the volume of time people spend in sedentary, upright and ambulatory activities. The activPAL is a valid measure of activity in children with CP.[23]

Participants will be asked to wear the activPAL3µ on their least affected leg for 7 continuous days. They will be asked to remove the monitor only for bathing and swimming. Participants will be provided with instructions on how to attach the monitor and a diary to log time spent sleeping and non-wear time. Participants will return the monitors at the training session after the assessment or via post in a stamped addressed envelope provided by the researcher.

Functional mobility

The Functional Mobility Scale (FMS) will be used to assess functional mobility. The FMS describes the level of a child's functional mobility in everyday life over 5m, 50m, and 500m, representing the home, school, and community settings respectively.[24] For each distance, an ordinal rating from 1 (wheelchair) to 6 (confident walking on all surfaces) is assigned depending on the amount of assistance required for the child's mobility.

Activity competence

The Canadian Occupational Performance Measure (COPM) will be used to identify important physical activities for each participant and to record the change in participants' perceived performance of this activity and the satisfaction with this performance over time.[25] The COPM is reliable and valid for use in young people.[26]

At baseline, in a semi-structured interview the researcher will ask the participant (or where appropriate the parents/carer together with, the or on behalf of the participant) to identify up to five physical activity related activities of daily living, which are important but problematic to the participant. The participant will then be asked to rate his/her current performance on each of these activities on a Visual Analogue Scale ranging from 1 (cannot do at all) to 10 (no problem at all) at 12 and 24 week assessments, the researcher will list the same five activities and the participant will be asked to repeat the rating of his/her performance and satisfaction on these activities. A change score of ≥ 2 is considered clinically meaningful in the COPM manual,[27] while a study with older adults reported an optimal cut-off value ranged from 0.9 and 1.9.[28]

Psychosocial impact

The Psychosocial Impact of Assistive Devices Scale (PIADS) will be used to record the impact of using a RaceRunner on a range of psychosocial outcomes such as happiness, self-esteem, independence and quality of life. It is a valid and reliable tool that consists of 26 items and can be completed by the parents/carer together with, the or on behalf of the participant.[29]

Adverse events

The safety of the intervention will be determined by recording the incidence of adverse events including falls and injuries between baseline and 24 weeks. An adverse event will be defined as "any untoward medical occurrence affecting a participant that does not necessarily have a causal relationship with the intervention".[30] At 12 and 24 weeks, researchers will systematically enquire about changes in the participant's health or any adverse events since the last assessment. Standardised questioning will be used to probe the participant regarding specific types of adverse events, e.g. falls, injuries, increase in muscle pain or spasticity. Participants will also be asked to record any adverse events experienced during training in their RaceRunning diaries. Trainers will also be asked to contact the research team if the participant experiences an adverse event while participating in the intervention. The following are expected non-serious AEs in response to the intervention: delayed onset muscle soreness, mild fatigue, saddle discomfort, shoe scuffing, foot discomfort.

A serious adverse event (SAE) will be defined as an untoward medical occurrence/effect that: results in death, is life-threatening, requires hospitalisation or prolongation of existing hospitalisation, or results in persistent or significant disability or incapacity.

Economic evaluation

This study will assess the feasibility of conducting an economic evaluation alongside a definitive cohort study of the effects of RaceRunning to reduce cardiometabolic disease risk and improve functional mobility. For the purpose of this study, we will assess the feasibility of collecting data on health service use and health-related quality of life. Health service use will be assessed using a modified version of the Client Service Receipt Inventory (CSRI). The CSRI collects retrospective information on service utilisation, service-related issues and income.[31] Health-related quality of life will be assessed using the EQ-5D-Y.[32]

Process evaluation

Fidelity

Fidelity to the intervention will be assessed through recording the overall volume of aerobic exercise achieved during RaceRunning, according to the FITT principle. Attendance, training duration and content for every session will be recorded by the coach. Intensity of the training sessions will be assessed for each participant by monitoring heart rate during 3 training sessions at week 1, week 12 and week 20 of the intervention.

Between week 3 and 8, 9 and 16, and 17 and 24, respectively, participants will be filmed during a RaceRunning session using a standard video camera for analysis of their running characteristics such as the propulsion technique (e.g. bilateral, unilateral, in phase), presence of foot drag and the presence of involuntary head or upper limb movement. This analysis of movement technique will enhance our understanding of the factors influencing maximum aerobic capacity and work efficiency.

Qualitative data collection

Focus groups will be conducted from a constructivist phenomenological perspective, aiming to explore a socially constructed phenomenon from the perspectives of participants. This balances different ways of looking at knowledge and knowing, and we will prioritise the constructivist perspective in analysis whenever possible.[33] Focus groups will be conducted with participants and/or their parents where appropriate to explore the views and experiences on taking part in RaceRunning.

Topic guides for the focus groups will be developed and piloted by the research team based on the study aims and available literature. An experienced qualitative researcher from Queen Margaret University will facilitate the focus groups, flexibly based around the topic guide. The focus groups will last around 60 minutes and will be recorded using digital audio recorders. There will also be an observer present who will take field notes (e.g. body language) during the focus group. The digital audio-recordings of the focus groups will be transcribed verbatim by the researcher and supplemented by observer notes.

Patient and Public Involvement

A person with CP was involved in the design of the study and will be involved in the conduct and reporting of the study.

Data analysis

As this is a feasibility study no formal statistical analysis of the effectiveness of the intervention will be undertaken. Analysis will be conducted in order to determine the feasibility and acceptability of the intervention, and the feasibility of conducting a definitive study.

Descriptive statistics (e.g., mean, standard deviation, range, median, interquartile range, percentages) will be used to report recruitment, retention, attendance at RaceRunning sessions, outcome measure completion, and outcomes and adverse events at baseline, 12 weeks and 24 weeks, respectively. Generalised estimating equations will be used to explore changes in outcomes over 24 weeks. Analyses will be conducted using Stata (Statcorp, USA) and SPSS (IBM Corp, USA).

Qualitative data analysis

Analysis will be carried out separately for data collected from children and data collected from parents. Transcripts will be read for the initial understanding and a participant summary will be prepared for each focus group. Feedback will be sought from the participants on the summary, which will be added to the transcripts. For the next stage of analysis, the researcher will familiarise with the data while noting down initial observations which will be followed by the coding of the whole data set. With the help of these codes, text will similar meanings will be thereby categorised to themes with assigned labels and definitions. A second researcher will carry out a secondary analysis of a selected sample to check the coding and interpretation and for confirming themes. Any lack of agreement between the researchers on interpretation or themes will be resolved through discussion. Any modification will be applied to the relevant sample. Relationships between themes may be explored if evidence of links exists, and or themes may be grouped into further categories. Qualitative software NVIVO will be used for interview analysis.

Sample size

This is a feasibility study and thus no sample size calculation has been performed. Twenty-five people will be recruited to the study.

Ethics and dissemination

The trial will be conducted in full conformance with the principles of the Declaration of Helsinki and to MRC Good Clinical Practice (GCP) guidelines. All researchers working on the trial will receive training in GCP-ICH guidelines. It will also comply with all applicable UK legislation and Queen Margaret University and University of Gloucestershire Research integrity guidance.

The study has ethical approval from the South East Scotland Research Ethics Committee and Queen Margaret University Research Ethics Committee (reference 19/SS/0035). Written informed consent will be obtained from participants aged 16 years and older. For those aged 16-17 years written informed consent will also be obtained from a parent/guardian. Written informed assent will be obtained from participants younger than 16 years of age and written informed consent will be obtained from his/her parent or guardian.

The findings for this study will be distributed through peer-reviewed journals, RaceRunning and Athletic Clubs, NHS trusts, organisations for people with disabilities e.g. CP Sport, and at national and international conferences.

Author contributions: MvdL and NT conceived the study. MvdL, JMR, NT, PK, SP, NA, GA, FK, KCJ, PvS, HD designed the study. JMR and MvdL drafted the manuscript. All authors have read and approved the final manuscript.

Funding statement: The study is supported by joint award from Action Medical Research and Chartered Society of Physiotherapy Charitable Trust.

Competing interests statement: None declared.

Ethical approval: The study has ethical approval from the South East Scotland Research Ethics Committee and Queen Margaret University Research Ethics Committee (reference 19/SS/0035).

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