




# BMJ Open Study protocol for Running for health (Run4Health CP): a multicentre, assessor-blinded randomised controlled trial of 12 weeks of two times weekly Frame Running training versus usual care to improve cardiovascular health risk factors in children and youth with cerebral palsy

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## ABSTRACT

**Introduction** Children and youth with moderate-severe (Gross Motor Function Classification System (GMFCS) levels II–V) cerebral palsy (CP) participate less frequently in physical activities compared with peers without CP and have elevated risk of cardiorespiratory morbidity and mortality in adulthood. Frame Running (RaceRunning) is a new athletics discipline that is an accessible option for physical activity participation for people with moderate-severe CP. There is no high-quality evidence for the effect of Frame Running on cardiovascular disease in children and young people with CP. The primary aim of this study is to conduct a randomised controlled trial of the effect of 12 weeks of Frame Running training on risk factors for cardiovascular disease.

**Methods and nalysis** Sixty-two children and youth with CP (age 8–20 years) in GMFCS levels II–V will be recruited across four sites and randomised to receive either 12 weeks of Frame Running training two times weekly for 60 min, or usual care. Outcomes will be measured at baseline, immediately postintervention (primary endpoint) and 12 weeks later for retention of training effects. The primary outcome is cardiorespiratory fitness as measured by distance covered on Six Minute RaceRunner Test with 1 min heart rate recovery. Other outcomes include blood pressure, objectively measured physical activity, body mass index, waist circumference, percentage body fat, gross motor function capacity, community participation, feasibility, tolerability and safety. Adverse events will be monitored, and participants and their caregivers will be interviewed to discern their experiences of participation in Frame Running.

**Ethics and dissemination** The Children's Health Queensland Hospital and Health Service and the University

## Strengths and limitations of this study

- This randomised controlled trial of Frame Running training in children and youth with cerebral palsy is powered to detect change on the primary outcome measure of cardiovascular fitness.
- Retention (sustainability) of changes will be examined at a follow-up 12 weeks after the training sessions are complete.
- Children and youth with severe functional mobility limitations and intellectual disability will be included.
- A validated maximal exercise test will not be conducted.

of Queensland Human Research Ethics Committees have approved this study. Results will be disseminated in peer-reviewed journals and scientific conferences; through professional and athletic organisations; and to people with CP and their families.

**Trial registration number** ACTRN12621000317897; Australian New Zealand Clinical Trials Registry number.

## INTRODUCTION

One in 700 Australians has cerebral palsy (CP), a permanent but not unchanging disorder of posture and movement caused by a disturbance to the developing fetal or infant brain.<sup>1</sup> Children with CP participate in physical activities less often compared with peers without CP.<sup>2</sup> Children with CP also have high levels of sedentary behaviour,<sup>3</sup> apparent

from early infancy and peaking by 4 to 5 years of age when followed through to middle childhood.<sup>4</sup> Adults with CP experience increased risk of non-communicable diseases associated with low physical activity, including cardiovascular disease, mental illness, osteoporosis and osteoarthritis (Odds Ratios 1.3–5.8).<sup>5</sup> There is evidence that the disparity in non-communicable disease risk begins early, with a large population-based cohort study demonstrating increased risk of mental health disorders in children (6–17 years of age) with CP compared with children without CP.<sup>6</sup> In this study, pain and low physical activity level explained part of the relationship between CP and depression.<sup>6</sup>

Life expectancy in people with CP in general is only slightly reduced compared with the people without CP, however, those individuals with moderate-severe motor impairments have significantly lower life expectancy.<sup>7</sup> The causes of early death in people with CP are most frequently respiratory and cardiovascular diseases,<sup>8</sup> with respiratory illness the leading cause of death in children with CP.<sup>9</sup> An Australian prospective population-based register study following n=3507 individuals with CP determined that inability to walk independently (an indicator of severe CP) was the strongest predictor of mortality in people with CP (adjusted Hazard Ratio 6.2).<sup>10</sup> There is expert consensus that increasing aerobic fitness and physical activity in children with severe CP is likely to ameliorate the severity of acute respiratory illness.<sup>9</sup> Despite this, recent systematic reviews have demonstrated that there are no effective physical activity interventions for people with CP that do not walk independently, and interventions for children who can walk independently may not have a clinically meaningful effect on physical activity behaviours, including habitual physical activity and participation in physical activities.<sup>11 12</sup> Contributing factors to inefficacy may have included: selection bias (inclusion of children with the highest level of physical activity and physical functioning), failure to address environmental, contextual and behavioural barriers to physical activity, issues with outcome measurement<sup>11</sup> and dosing below minimum recommended guidelines.<sup>13</sup> It is clear that there is an urgent need for high-quality research into physical activity interventions of sufficient dose and duration in youth with CP who have major limitations in walking ability. Furthermore, such interventions need to be safe, community-based, informed by consumer needs, and aimed to enable ongoing, normal community participation and inclusion.

Frame Running, formerly known as RaceRunning, is a para-athletics discipline recently sanctioned by World Para Athletics (the International Paralympic Sports Federation for athletics). Frame Running was invented in 1991 by Connie Hansen, an Occupational Therapist and para-athlete and Mansoor Siddiqi, a para-athlete with CP competing in the now defunct discipline of backward wheelchair racing (foot-propelled). Frame Running uses a three-wheeled frame with low rolling resistance for support, enabling running in people with otherwise



**Figure 1** Petra Frame Runner(TM) by ConnieHansen running frame.

severe mobility limitations (see [figure 1](#)). In the absence of an existing systematic review of the literature, an author search was conducted on 21 September 2021 for articles indexed in the PubMed database, using the terms ‘Race-Running’ OR ‘Frame Running’ OR ‘race running’ AND ‘cerebral palsy’ located in title/abstract (with additional hand search of reference lists for included articles). This search returned only seven studies: one pilot single-group pre–post trial,<sup>14</sup> one study protocol for a pilot randomised feasibility study,<sup>15</sup> one reliability study for a Frame Running-specific field exercise test,<sup>16</sup> two cross-sectional studies examining relationship between impairments and Frame Running performance<sup>17 18</sup> and two cross-sectional studies on kinesiological and metabolic responses or adaptations to use of running frames.<sup>19 20</sup> The pre–post pilot trial included n=15 adolescents and young adults with CP (age range 9–29 years, Gross Motor Function Classification System (GMFCS) levels I–IV) and demonstrated that 12 weeks of two times weekly Frame Running training led to on average, a 34% increase in cardiorespiratory endurance and a 9% increase in thickness of the medial gastrocnemius muscle.<sup>14</sup> Frame Running can evoke a heart rate commensurate with high-intensity exercise<sup>19</sup> and uses large muscle groups in a reciprocal way that may have functional cross over to enhanced mobility.<sup>20</sup> A larger (n=25) pre–post pilot study of once weekly Frame Running training for 24 weeks duration is planned.<sup>15</sup> This study with no accompanying sample size calculation has the potential to be underpowered and/or underdosed to detect improvements in cardiometabolic risk factors. Furthermore, as the study is unrandomised, the quality and certainty of the evidence provided will necessarily be lower than a randomised study.

We, therefore, aim to conduct an adequately powered randomised controlled trial of Frame Running training in children and youth with CP on cardiometabolic risk factors and related outcomes (Run4Health CP). This study may, therefore, provide evidence that cardiometabolic risk factors can be modified in children and youth with CP who have moderate to severe motor impairment

and high support needs in mobility. This evidence may have critical patient and clinical impacts through support of funding for running frames and may help to foster development of the discipline and expand participation opportunities.

## METHODS AND ANALYSIS

### Objectives

The primary objective of this study is to compare the effect of 12 weeks of Frame Running training versus usual care control on cardiovascular fitness (endurance) on the Six Minute RaceRunner Test (6MRRT) and 1 min heart rate recovery ( $HRR_{1min}$ ) following exercise testing immediately at postintervention (primary endpoint) and at 12 weeks postintervention.

Secondary objectives are to compare the effect of 12 weeks of Frame Running training versus usual care control immediately postintervention and at 12 weeks postintervention on:

1. other cardiovascular risk factors including: resting blood pressure (BP), habitual physical activity level, body mass index, per cent body fat and waist circumference.
2. Gross motor activity capacity including gross motor function and Frame Running-specific activity limitation tests.
3. Community participation.

The tertiary objective of this study is to determine whether 12 weeks of Frame Running training is feasible, tolerable, safe and sustainable in the study population, including whether participants report that it induces additional pain and fatigue when compared with usual care.

### Trial design

Run4Health CP is a pragmatic, single (assessor)-blind randomised controlled, multicentre trial with two parallel groups. The primary timepoint is immediately postintervention (12 weeks postbaseline) and the secondary timepoint is 12 weeks postintervention (24 weeks postbaseline). The study will be conducted in four Australian cities, Brisbane (n=24), Cairns (n=18), Sydney (n=10) and Sunshine Coast (n=10). Assessment of outcome measures and Frame Running training will be conducted at community synthetic athletic tracks and nearby associated indoor sports facilities at a time convenient to participants and their caregivers. Recruitment commenced on 16 August 2021 and the first participant was enrolled on 16 September 2021. Last participant data collection is anticipated in January 2023.

### Eligibility criteria

Participants eligible for the trial must comply with all of the following eligibility criteria at randomisation: (1) diagnosis of CP and classified in GMFCS levels II–V, (2) between 8.00 and 20.99 years of age, (3) live within 150 km of one of the trial sites, (4) have not engaged in

more than six sessions of formal Frame Running training with a coach or health professional within the last 6 months, (5) can understand and follow the directions of the coach and assessors for the purposes of training safely and completing outcome measurement in the opinion of the principal investigator.

Participants are excluded if at any time: (1) the child/youth has orthopaedic and/or neurological surgery within 6 months prior to baseline or during the study period requiring a period of recovery that would exclude the participant from training for more than 1 week, (2) the child/youth has uncontrolled epilepsy, medical fragility and/or serious precautions not able to be accommodated (eg, significant history of atraumatic lower limb fractures or sacral pressure injuries, etc) precluding participation in moderate-vigorous intensity Frame Running, (3) caregiver English language skills are not sufficient to understand the study information, provide informed consent and/or complete study questionnaires.

### Interventions

#### Frame Running training group

Frame Running training will consist of two, 60 min sessions per week for 12 consecutive weeks (total dose 24 hours). Established guidelines for aerobic exercise to improve cardiovascular health in typically developing individuals recommend a minimum frequency of three sessions per week.<sup>13</sup> There is evidence, however, that two sessions per week is adequate in deconditioned individuals with CP to improve aerobic fitness,<sup>13</sup> and this was demonstrated in the pilot pre–post study of Frame Running training by Hjalmarsson *et al.*<sup>14</sup> This can likely be attributed to the dose–response relationship between physical activity and cardiorespiratory outcomes, whereby even small increases in physical activity in previously inactive individuals can result in clinically meaningful improvements in health.<sup>21</sup> Provision of only two sessions per week may also increase the likelihood that participants can comply with the intervention (considering issues such as time and financial constraints relative to a third session).

Participants will attend Frame Running training in groups of approximately three, matched by age and/or ability if possible. Sessions will be administered by a coach with qualifications in Physiotherapy and/or Exercise Physiology. Participants in the training group are permitted to receive their usual care from non-study providers (as per concomitant care) with no restriction.

Frame Running training sessions will consist of a combination of (1) anaerobic Frame Running (ie, starts and sprints drills using established athletic training principles), (2) aerobic Frame Running (ie, steady running working towards  $\geq 15$  min duration) and (3) task-specific functional training for Frame Running technique and skills (eg, braking, steering, propulsion strategies, running form and power). Training sessions will increase in difficulty in a stepwise fashion, with the aim to initially develop basic skills in operating a running frame, working towards maintaining moderate-vigorous exercise intensity

throughout a 60 min session. A training load of 60%–75% of peak heart rate can elicit a 9%–40% increase in peak aerobic capacity with 2–4 sessions per week for minimum 20 mins in individuals with CP.<sup>13</sup> Based on a literature review of exercise training studies, proposed ideal exercise parameters for individuals with CP are: an intensity between 60% and 95% of peak heart rate, between 40% and 80% of the heart rate reserve or between 50% and 65% of  $VO_{2peak}$ .<sup>13</sup> To monitor adherence to this exercise intensity, participants will wear a Polar Verity Sense (Polar Electro Oy, Kempele Finland) optical heart rate monitor on the non-dominant upper limb during training sessions with output observed by the coach and/or assistants (eg, undergraduate physiotherapy or exercise physiology students). As suggested by Verschuren *et al*, peak heart rate will be estimated at 194 bpm for children and youth with CP in the absence of maximal exercise testing.<sup>22</sup> Therefore, the target heart rate will be between 116 bpm and 185 bpm. Where possible, when a participant provides a Global Positioning System (GPS)-enabled smartphone, distance covered and time in motion will be recorded by attachment of the smartphone to the running frame using the Polar Beat phone application (Polar Electro Oy, Kempele Finland).

Running frames are registered in the low-risk Medical Device Class 1 category on the Australian Register of Therapeutic Goods. They are manufactured overseas and are imported to Australia by Dejay Medical and Scientific. There are currently two brands available in Australia, 'RAD—Trike—Disability vehicle, cycle, tricycle, foot-propelled' (ARTG: 345236) and 'By Connie Hansen—Disability vehicle, cycle, tricycle, foot-propelled' (ARTG: 309224). Both types of running frames may be used in the trial according to availability and suitability, as the differences between these brands are expected to be superficial considering the context of the trial (novice and beginner Frame Running athletes, recreational style participation with elementary competition). Where possible, the same frame will be used by each participant throughout the study period with consistent attachments, seat height and chest plate angle/depth unless these are adapted for performance reasons.

#### Usual care control group

Participants in the control group will receive their usual care from non-study providers (type/dose/content as per concomitant care). This will determine the effect of Frame Running training in addition to usual care, which already contains active treatments such as physiotherapy and/or occupational therapy. Participants in the control group will not be offered Frame Running training and will be asked to refrain from participating in Frame Running until they have exited the study, however, this will not be actively prevented for ethical reasons. As Frame Running requires access to a running frame, and participants are not expected to have their own frame, it is expected that few participants in the control group will participate in Frame Running during the study period.

Following outcome measurement at the final time point, participants in the control group will be provided with an information package regarding local Frame Running training sessions, running frame fitting results (eg, frame size required and attachments) and advice about how to obtain a running frame and participate in the sport. They will also receive up to two phone calls from a therapist providing physical activity counselling and advice. The aim of this package of supports is to improve equity in access to Frame Running opportunities for those otherwise receiving a no-treatment control.

#### Modifications and adaptations

Heart rate data will be used to adjust the session in real time and to tailor the progression of session difficulty from week to week, so that the participant spends at least 60% of session duration in the target HR range. Individual tailoring will also accommodate variability in participants' propulsion strategies, motor type/distribution, activity limitations, age and interests. If any unexpected, unusual or additional pain or fatigue beyond what is considered 'normal' is experienced by a participant allocated to the Frame Running training group, this will be discussed with the participant and their caregiver and modifications to the training programme may be implemented. Unexpected or unusual pain or fatigue in the training group will be recorded as an adverse event. Other participant characteristics may necessitate modification or adaptation to the training programme, including but not limited to intellectual disability, injury, hearing and/or vision impairment, tactile and/or proprioceptive impairment, behavioural and/or emotional dysregulation. Modifications may include reduction in dose, changes to training session content, use of assistive technology (hearing aids, visual aids, etc), visual guides, caregiver involvement and/or advice and education regarding management of pain, injury and fatigue.

#### Adherence and fidelity

The training content has been manualised to facilitate consistent application by coaches across trial sites and participants and to promote adherence to the prescribed dose. Several strategies will be applied to optimise the participant's frequency of attendance at sessions and level of involvement (which is defined as the subjective experience of participation while attending and includes elements such as engagement, motivation, persistence and affect).<sup>23</sup> These strategies are hypothesised to fulfil participants' basic psychological needs of autonomy, competence and relatedness according to self-determination theory,<sup>24</sup> which has been demonstrated to underpin physical activity interventions in children with CP<sup>25</sup>:

1. Training activities will be individually tailored as described above. This is likely to facilitate a 'just right challenge' and fulfil participants' need for competence.<sup>25</sup>
2. Training will occur in groups of approximately three, matched where possible for age and/or ability. Training together with peers, with social group dynamics

managed carefully, is likely to promote social connection and fulfil participants' need for relatedness.<sup>26</sup>

3. Coaches will use autonomy-supportive, empathetic communication with participants and families. Coaches will facilitate participant self-efficacy through teaching positive self-talk about performance and will promote positive peer-to-peer encouragement.<sup>25</sup>

Furthermore, training sessions will take place in locations where Frame Running squads train on a regular basis. This provides an ongoing avenue for normal, regular participation in the sport once the free clinical trial sessions conclude.

Individual adherence to the training manual will be recorded on a session-by-session basis by coaches. Measures include:

1. Percentage of sessions attended (including partial attendance).
2. Percentage of training drills completed according to the manualised content.
3. Percentage of session duration spent within the target heart rate threshold for training intensity.
4. List of modifications or adaptations.
5. Distance covered and time in motion during the training session.

Reasons for missed or incomplete sessions will be recorded. Adherence data will be reported alongside study outcomes.

### Concomitant care

Participants in both groups may continue any usual care from non-study providers throughout the study period (except Frame Running in the usual care control group). Type, dose and duration of usual care are likely to vary widely between participants owing to individual needs, access and funding arrangements. This could include Botulinum Toxin-A injections, serial casting and a broad array of exercise and movement-based therapies. Participants in both groups will be asked to record frequency of participation in Frame Running and other physical activities, and frequency/type/dose of usual care therapies from non-study providers from allocation to exit using a Usual Care Diary. Botulinum Toxin-A injections and serial casting are not expected to have significant impacts on our activity and participation-level outcome measures at the group level. Based on prior experience in randomised controlled trials administered by our centre, it is not feasible to exclude participants receiving these interventions as timely recruitment would be affected.

### Outcomes

#### Primary outcome

##### Cardiovascular health (primary)

Distance (metres) covered in the 6MRRT.<sup>16</sup> The 6MRRT is a validated measure of RaceRunning endurance with good test–retest reliability (Intraclass Correlation Coefficient (ICC)=0.78–0.91) in children classified in GMFCS levels III and IV. The 6MRRT is theoretically a submaximal

exercise test, however, it is likely that many participants will achieve almost maximal heart rate.

#### Secondary outcomes

##### Cardiovascular health (secondary)

HRR<sub>1min</sub> will be taken immediately following the 6MRRT. HRR<sub>1min</sub> in beats per minute is the difference between the heart rate taken at the cessation of the 6MRRT and exactly 60s later, while the participant is engaged in relative rest (ie, has stopped moving).<sup>27</sup> HRR<sub>1min</sub> is strongly associated with cardiac mortality and is responsive to a 12-week cardiac rehabilitation programme in children following heart surgery.<sup>27</sup> Children and youth will wear a Polar Verity Sense Optical heart rate monitor on the less-impaired upper arm during testing.

Resting BP will be measured using an automated arm-cuff sphygmomanometer (valid and reliable).<sup>28</sup> Resting systolic and diastolic BP in mm Hg is a traditional risk factor for cardiometabolic disease in individuals with CP,<sup>28</sup> and systolic BP is associated with cardiorespiratory fitness, central adiposity and body mass index (BMI) in children with CP.<sup>29</sup> Systolic and diastolic BP were responsive to a 12-week training programme in youth with Down syndrome.<sup>30</sup>

Habitual physical activity will be quantified using accelerometry, a valid, reliable and feasible method in youth with moderate to severe CP.<sup>31</sup> Participants will wear an ActiGraph GT3X+ on the less-impaired wrist and less-impaired anterior thigh for 7 days during waking hours during their usual activities (free-living). Data will be processed to identify time spent in different postures and activities using machine learning algorithms; a combined thigh and wrist classification model has been validated in children classified in GMFCS levels III and IV.<sup>31</sup>

BMI, kg/m<sup>2</sup> will be calculated according to the equation: BMI=weight (Kg)/height<sup>2</sup> (m). Weight will be taken using the same calibrated digital scale at each site and height taken using the same stadiometer at each site for all participants. Participants who are unable to stand unassisted will access chair scales. If body shape distortion is severe and/or standing height is not feasible, then height will be measured using a recumbent measuring board if available or will be estimated using segmental limb length (knee height).<sup>32</sup> Anthropometric measures will be converted to Z-scores using age and gender-specific reference data for the general population.<sup>33</sup>

Waist circumference (cm) will be measured to the nearest millimetre at the midline level (midway between the superior border of the iliac crest and the inferior rib margin, often slightly above the umbilicus) using a non-stretchable tape measure.<sup>34</sup>

Percentage body fat will be estimated based on the triceps and subscapular skinfold thickness using CP-specific equations.<sup>35</sup> This will be measured using callipers (Harpenden Skinfold Caliper, Baly International, West Sussex UK) by trained investigators.

### Gross motor capacity

Gross motor function will be assessed using GMFM-66, a criterion referenced observation measure developed using Rasch modelling to measure gross motor function of children with CP.<sup>36</sup> The GMFM-66 has established construct validity, high test–retest reliability (ICC 0.99) and is responsive to change.<sup>36 37</sup>

Frame Running-specific activity limitation will be assessed using 100 m sprint (time in seconds), distance covered in four strides (metres, average of best two trials of three), and step count in 20 m (steps, average of best two trials of three). The assessment of function is activity specific and, therefore, outcomes should be strongly related to the activity of interest.<sup>38</sup> These assessments are investigator developed and will be subjected to further independent assessment of their validity and reliability.

### Participation

Community participation will be evaluated using the Participation and Environment Measure for Children and Youth (PEM-CY).<sup>39</sup> The PEM-CY is a caregiver-report questionnaire with good test–retest reliability and internal consistency.<sup>39</sup> Youth 18 years and older will be invited to self-report the questionnaire. Summary scores for participation frequency, involvement and per cent environmental supportiveness will be calculated.

### Feasibility, tolerability and safety

Feasibility, tolerability and safety will be measured on a weekly basis in both groups using the Wong-Baker FACES rating scale (pain),<sup>40</sup> Fatigue Severity Scale (fatigue),<sup>41</sup> and for the training group only, training load (Rate of Perceived Exertion (RPE) on the OMNI RPE<sup>42</sup> multiplied by session duration).<sup>43</sup> Monitoring of adverse and unintended events including injuries will be undertaken throughout the study.

### Classification systems and demographic characteristics

The following validated classification systems will be applied: GMFCS Expanded and Revised,<sup>44</sup> Manual Abilities Classification System,<sup>45</sup> Communication Function Classification System,<sup>46</sup> Visual Function Classification System (VFCS),<sup>47</sup> Eating and Drinking Ability Classification System (EDACS).<sup>48</sup> The VFCS and EDACS will be applied owing to the contribution of the visual system to athletic performance,<sup>49</sup> and the association between eating and drinking ability and nutrition status, which is relevant to body composition, muscle mass, functional ability and performance in training programmes in people with CP.<sup>50 51</sup> If known, the participant's Frame Running Sport Class under the two existing classification systems will be recorded (RR1/RR2/RR3 and/or T71/T72). If unknown (or not yet classified), a provisional classification will be performed following the process outlined by Athletics Australia.

The following participant demographic characteristics will be collected to characterise the sample: participant age, sex, dominant hand, self-reported household

income, residential postal code, presence of comorbid diagnoses, list of up to nine sports/PAs the participant attended in the last 12 months and caregiver frequency of participation in structured and unstructured sports/PAs in the last 4 months. Participants will also be screened for medical conditions that may be precautions to high-intensity exercise using a running frame requiring attention or adaptation but not meeting exclusion criteria (eg, known stable cardiovascular or respiratory condition, etc.).

### Participant timeline

Run4Health CP schedule of assessments and interventions are provided in [table 1](#) and the Consolidated Standards of Reporting Trials (CONSORT)<sup>52</sup> study flow diagram is provided in [figure 2](#).

### Sample size

Based on the primary outcome of 6MRRT, which has a smallest detectable difference of approximately 150 m and sample SD of 150 m,<sup>16</sup> a sample size of n=44 will detect at least this difference at 90% power and two-sided 5% significance level. To allow for up to 15% attrition, n=52 (n=26 per group) will be required, however, additional funding awarded to increase the implementation of Frame Running in a fourth site (Sunshine Coast) will allow for up to n=62 participants.

### Recruitment

Strategies to achieve adequate participant enrolment to reach the target sample size are as follows:

1. Clinical database: potential participants will be identified on a clinical database held and maintained by the Queensland Paediatric Rehabilitation Service (QPRS) at the Queensland Children's Hospital and the Sydney Children's Hospitals Network (SCHN). Caregivers of children/youth who have previously consented to receive communications about research studies will be sent a copy of the study flyer to their contact email or postal address.
2. Clinical service: children and youth with CP attending an associated clinical service within QPRS and SCHN will be identified by their treating clinicians based on eligibility criteria. Clinicians will ask permission to discuss the project and gain consent from the family to be contacted by a project staff member.
3. Patient advertising: patient waiting areas at associated clinical services within QPRS and SCHN will display the approved flyer during the recruitment period.
4. Newsletter: the flyer will be included in the newsletters distributed by associated clinical services within QPRS and SCHN and research groups of the investigators.
5. Websites: the flyer will be posted on the research websites of the investigators.
6. Social media/word of mouth: the flyer will be posted on social media websites, which may include but are not limited to Facebook, Twitter and Instagram. The

**Table 1** Schedule of assessments for Run4Health CP study

Timepoint	Enrolment	Allocation/baseline	Intervention	Immediately post-intervention (12 weeks)	Retention (24 weeks)
<b>Visit number:</b>	Screen	T1		T2	T3
Participant contact	X				
Eligibility screen	X				
Informed consent	X				
Allocation		X			
<b>Interventions:</b>					
Frame running training			X		
Usual care control			X		
<b>Assessments:</b>					
Classification systems (GMFCS, MACS, CFCS, VFCS, EDACS)		X			
Demographic questionnaire		X			
Frame Running provisional sport class		X			
Six minute RaceRunner test		X		X	X
Heart rate recovery in 1 min		X		X	X
Resting blood pressure		X		X	X
7 day free-living accelerometry for habitual physical activity (ActiGraphGT3X+thigh and wrist)		X		X	X
Body mass index		X		X	X
Per cent body fat		X		X	X
Waist circumference		X		X	X
Gross Motor Function Measure 66		X		X	X
Frame Running activity limitation tests		X		X	X
Participation and Environment Measure for Children and Youth		X		X	X
Wong-Baker FACES rating		X	X	X	X
Fatigue severity scale		X	X	X	X
Monitoring of adverse and unintended events		X	X	X	X
Usual care diary		X	X	X	X

CFCS, Communication Function Classification System; EDACS, Eating and Drinking Ability Classification System; GMFCS, Gross Motor Function Classification System; MACS, Manual Abilities Classification System; VFCS, Visual Function Classification System.

electronic version of the flyer may then be shared by third parties.

### Allocation and blinding (masking)

Participants will be randomly assigned to either Frame Running training or usual care control with a 1:1 allocation as per a computer-generated randomisation schedule using the Research Electronic Data Capture (REDCap) randomisation module, stratified by GMFCS (II–III/IV–V) and site (Brisbane vs Cairns vs Sydney vs Sunshine Coast), using permuted blocks of random sizes. Randomisation will occur following enrolment into the study and completion of all baseline assessments except for 7-day habitual PA monitoring. [Table 2](#) contains information about concealment and blinding (masking), who these apply to, how and when. As participant health and safety is managed directly by Frame Running coaches who are

not blind to treatment allocation, procedures for emergency unblinding are not required.

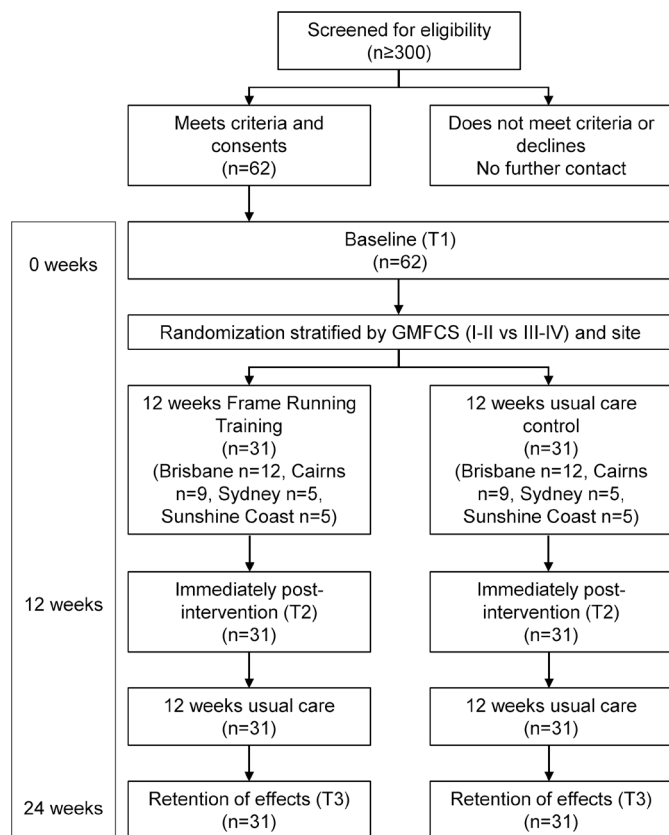
### Data collection

#### Assessment order

Assessments will be delivered in a standardised order across two sessions (first lab-based, second track-based) on different days to reduce the effect of fatigue and enable a familiarisation session with the running frame prior to track-based assessments. The track-based assessments will be delivered in the following order: 100 m sprint, distance in four strides, step count in 20 m and 6MRRT with adequate rest in between.

#### Interventionist training and experience

The interventionists (Frame Running coaches) will be exercise physiologists, physiotherapists and/or athletics



**Figure 2** Consolidated Standards of Reporting Trials (CONSORT) study flowchart. GMFCS, Gross Motor Function Classification System.

coaches with at least 2 years' experience prescribing physical activity programmes to people with disabilities including CP and conducting group exercise sessions with children and young people. Interventionists will have current cardiopulmonary resuscitation and first aid qualifications and will adhere to institutional policies and procedures for child safety.

Interventionists will be provided with 6 hours face-to-face didactic training from the principal investigator in how to deliver the intervention according to the training manual. The following topics will be covered: (1) general principles of aerobic and anaerobic exercise in CP, (2) coaching principles to provide a fun and intrinsically motivating exercise experience, (3) interpreting and applying the Frame Running intervention manual, (4) correctly fitting athletes to running frames and (5) practical component. Regular supervision meetings will be conducted throughout the trial to facilitate adherence to the training manual.

#### Outcome assessor training and experience

Outcome assessors will be physiotherapists with at least 3 years' experience administering the GMFM-66 to children and youth with CP and will have completed the GMFM Criterion Test for scoring reliability. They will be provided with written and videotaped standardised procedures for the administration of all other study outcome

measures. Regular supervision meetings will be conducted to facilitate adherence to the assessment manual.

#### Retention

##### Participant retention

The following strategies will be used to promote participant retention and complete follow-up: (1) Frame Running training will be offered at no cost, (2) where possible according to track availability, sessions will be scheduled at mutually convenient times, (3) questionnaires will be administered using the REDCap survey module enabling forced choice/completion and automated email reminders, (4) usual care control participants and/or their caregivers will be reminded that a Frame Running participation pack with interventionist follow-up support will be provided after the T3 retention (24 weeks) timepoint is complete, (5) once enrolled, investigators will make all reasonable efforts (including phone call, email and text messages) to contact participants and/or their caregivers to encourage completion of overdue assessments, if any.

##### Participant withdrawal

Participants can withdraw at any time. Participants who choose to withdraw from the study will not be penalised in any way. They will be assisted to source another local therapy option that matches their preferences if desired. Participants are informed of their right to withdraw at any time without consequences at the time of reading participant information forms and signing of consent forms. Any deidentified (including reidentifiable) data collected from participants who later withdraw will be retained and included in analyses. Reasons for participant withdrawal will be recorded and reported where available.

##### Data management and access

Study data will be collected and managed using REDCap electronic data capture tools hosted at The University of Queensland.<sup>53 54</sup> REDCap is a secure, web-based software platform designed to support data capture for research studies. To promote data quality and minimise data loss, REDCap forms will be set up with range checks and forced completion. All assessments administered by the outcome assessors for backup if recording forms are incomplete, damaged or lost. The University of Queensland Research Data Manager database will be used for long-term data storage, and a description of the data will be uploaded onto the UQeSpace repository at the conclusion of data collection and analysis. Confidentiality of participant data will be maintained at all times from collection to storage. A deidentified data set will be made available on written request for the purposes of further scientific research, including meta-analysis, ancillary studies related to the original aims and objectives and verification of results.

##### Statistical methods

Between-group differences for primary and secondary outcomes will be determined on an intention-to-treat



**Table 2** Blinding (masking) and concealment information for the Run4Health CP trial

Group or individual blinded	Information withheld	Method of blinding
Person assigning participants to groups	Group assignment	(REDCap) randomisation module, with schedule generated and entered by a biostatistician not otherwise involved in participant recruitment, assessment or trial conduct. Trial staff with access to the randomisation function (to allocate participants) do not have access to the randomization schedule.
Participants	Not blinded after baseline	
Coaches delivering intervention	Not blinded after baseline	
Outcome assessors	Group assignment	Not told of group assignment and no access to randomisation status or intervention information on REDCap. Participants and caregivers will be asked not to discuss their assignment with the outcome assessor. Questionnaires will be entered directly into REDCap by participants and/or their caregiver and will be locked for editing by study personnel except for one research data manager (who is not on the investigator team) if an error in data entry is made. All changes to data are available in a log accessible from REDCap.
Research data manager/study coordinator	Not blinded after baseline	
Statistician	Group identity	The analysis code is written and finalised before the dataset is made available for analysis. The groups are randomly assigned as 'group A' or 'group B' in the downloaded dataset provided to the statistician. The identity of the group is revealed after the primary statistical analysis is complete.
Investigators and manuscript writers	Not blinded	

CP, cerebral palsy.

basis using generalised estimating equations to account for the repeated measures design, stratification and potential missing outcome data.<sup>55</sup> Covariables will be stratification factors (GMFCS II–III vs IV–V and site), baseline and wear time for accelerometer data that may be confounded by duration of wear, for example, average minutes per day of sedentary behaviour. Effect estimates will be presented as a mean difference and 95% CI with a significance level of  $p < 0.05$ . Data will be inspected visually for normality, homoscedasticity and linearity. If any analyses are found to violate necessary assumptions, then data will be transformed, or appropriate non-parametric analysis methods will be used.

### Data monitoring and safety

There are no additional risks to participating in Frame Running beyond typical physical activity participation using adaptive equipment in this population. The following control strategies will be implemented to manage the risk of adverse events: (1) participants will be screened for the presence of comorbid conditions and will be managed by senior experienced clinical staff including the principal investigator, (2) families will be provided with an information sheet and brief counselling on the risks associated with

wearing accelerometers, with a focus on preventing the development of pressure areas, early identification of allergic skin reactions and reducing unpleasant sensations, (3) treating/assessing staff will be provided with standardised training that includes a component on awareness of risks, application of control strategies and safety, (4) participants may use running frames only with a properly fitting, Australian-standards approved bicycle helmet and appropriate footwear of their choice, including orthoses if preferred (same footwear to be worn for all assessments), (5) participants will be reminded to use sun protection and have access to fresh drinking water during training sessions, (6) participants will be instructed on safe use of the running frame at a familiarisation session of at least 30 min, (7) participants will be encouraged to wear padded bike pants to reduce discomfort in the saddle and (8) fatigue and pain will be monitored on an ongoing basis and training load adapted accordingly. Coaches and assessors are asked to report adverse events in real time using REDCap, which is monitored by the principal investigator, who will determine the severity of the adverse event, whether it is expected or unexpected, and whether it is related or unrelated

to the intervention. Serious or unexpected adverse events will be discussed at the earliest convenience by the chief investigators (SER, LS, LM, CS and RNB) and reported to the Ethics committees, at which point a decision will be made about continuing the trial. No interim analyses will take place. The study is covered by standard clinical trials insurance held by The University of Queensland.

### Qualitative interviews

To fully address the tertiary objective of this study, semi-structured interviews will be conducted in up to eight focus groups (one child/youth participant group and one parent/caregiver group at each site). Participants 18 years or older who provide independent consent to participate in the intervention will be asked if they would like the option of a parent/caregiver or support person to attend the parent/caregiver group. The aims of the qualitative interviews are to understand how participants and/or their caregivers perceive their involvement in the programme and elucidate barriers and facilitators to ongoing, sustainable participation in Frame Running. Qualitative interview transcription will be completed by a high-quality paid service and checked against original recordings. Participants will have the opportunity to review their transcripts and edit their responses prior to analysis. Transcripts will be thematically analysed using an inductive content analysis approach.<sup>56</sup>

### Patient and public involvement

A person with CP, a parent/caregiver and Frame Running organisations have been invited to participate as consumer representatives during the study period. They will be financially compensated for their time and expertise at the rate of AU\$50 per hour. A parent of a child with CP (who participates and competes in Frame Running at an international level) reviewed the protocol and provided feedback on the study design, which has been integrated. At least one consumer representative will meet with the study team not less than every 2 months once recruitment commences to provide advice and input in relation to all following phases of the trial (conduct, analysis and reporting).

## ETHICS AND DISSEMINATION

### Informed consent process

For children and youth <18 years of age or ≥18 years with an impaired capacity to consent, written informed consent will be obtained from the legal guardian. Youth ≥18 years who can provide their own written informed consent will do so. This will occur after the treating/assessing staff member has explained the study again in an accessible format (verbal, written) to the satisfaction of both the participating parent/guardian and/or child/youth.

### Ethics and dissemination

Run4Health CP is registered on the Australian New Zealand Clinical Trials Registry. Protocol updates will be reflected in the trial registration and reported in the primary result manuscript. The project has received ethics approval from the Children's Health Queensland Hospital and Health Service Human Research Ethics Committee (HREC/21/QCHQ/69281) and the University of Queensland Human Research Ethics Committee (2021/HE000725). Results of the study will be published/disseminated in (1) the trial registration database, (2) conference abstracts and presentations, (3) peer-reviewed articles in scientific journals, (4) organisation and institution newsletters and media releases and (5) in accordance with the Australian National Statement 3.1.65, directly to participants and consumers in a format that is appropriate and accessible to them as the research will be likely to generate findings or results of significance to young people with CP and their families. The study will be reported in a way consistent with both Template for Intervention Description and Replication<sup>57</sup> and Consensus on Exercise Reporting Template guidelines.<sup>58</sup>

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**Contributors** SER, LS, LM, CS, EB, KW and RNB conceived the trial. SER completed the initial draft of the manuscript. MDC generated the randomization strata and provided biostatistical advice and information. SGT, RT, and ID contributed technical expertise to the protocol manuscript for physical activity measurement, therapist outcome assessment, and Frame Running coaching respectively. AG and BD designed the Frame Running training session content. ZC designed the assessment methods and procedures. All authors designed the study, have read, edited, and approved the final manuscript and supplementary files.

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**Competing interests** None declared.

**Patient and public involvement** Patients and/or the public were involved in the design, or conduct, or reporting, or dissemination plans of this research. Refer to the Methods section for further details.

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