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ACTIVE STRIDES-CP: protocol for a randomised trial of intensive rehabilitation (combined intensive gait and cycling training) for children with moderate-to-severe bilateral cerebral palsy

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ABSTRACT

Introduction For children with cerebral palsy (CP), who are marginally ambulant, gross motor capacity peaks between 6 and 7 years of age with a subsequent clinical decline, impacting their ability to engage in physical activity. Active Strides-CP is a novel package of physiotherapy targeting body functions, activity and participation outcomes for children with bilateral CP. This study will compare Active Strides-CP to usual care in a multisite randomised waitlist-controlled trial.

Methods and analysis 150 children with bilateral CP (5–15 years), classified in Gross Motor Function Classification System (GMFCS) levels III and IV will be stratified (GMFCS III vs IV, age 5–10 years; 11–15 years and trial site) and randomised to receive either (1) 8 weeks of Active Strides-CP two times/week for 1.5 hours in clinic and one time/week for 1 hour alternating home visits and telehealth (total dose=32 hours) or (2) usual care. Active Strides-CP comprises functional electrical stimulation cycling, partial body weight support treadmill training, overground walking, adapted community cycling and goal-directed training. Outcomes will be measured at baseline, immediately post-intervention at 9 weeks *primary endpoint* and at 26 weeks post-baseline for retention. The primary outcome is the Gross Motor Function Measure-66. Secondary outcomes include habitual physical activity, cardiorespiratory fitness, walking speed and distance, frequency/involvement of community participation, mobility, goal attainment and quality of life. Analyses will follow standard principles for randomised controlled trials using two-group comparisons on all participants on an intention-to-treat basis. Comparisons between groups for primary and secondary outcomes will be conducted using regression models. A within-trial cost utility analysis will be performed.

Ethics and dissemination The Children's Health Queensland Hospital and Health Service, The University of Queensland, The University of Melbourne and Curtin

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ This randomised controlled trial of Active Strides-CP in children with bilateral cerebral palsy is powered to detect a change on the primary outcome measure of gross motor function.

⇒ The Active Strides-CP intervention uniquely combines elements of motor training and environmental modification to address both gross motor capacity and restrictions in participation in physically active leisure.

⇒ A combination of clinic-based and home-based programme will promote transfer of gross motor capacity gains into improved mobility and participation in physically active leisure in the community.

⇒ A within-trial cost utility analysis will be conducted which will inform policy level decisions on the implementation of Active Strides-CP.

⇒ One limitation is that usual care cannot be standardised and is likely to be highly variable across states.

University Human Research Ethics Committees have approved this study. Results will be disseminated as conference abstracts and presentations, peer-reviewed articles in scientific journals, and institution newsletters and media releases.

Trial registration number ACTRN12621001133820.

INTRODUCTION

In Australia, 420 infants are born annually with cerebral palsy (CP), making it the most common physical disability in childhood and the fifth most costly health condition ($A1.47 billion per annum, average yearly costs $A43 431 per individual).[1] Of these infants,

34% will have moderate-to-severe motor physical disability (Gross Motor Function Classification System (GMFCS) III to V) which is associated with reduced general health, greater pain and discomfort,[2] reduced independence in daily life skills,[3] restricted participation in physical activity[4] and poorer vocational outcomes.[5] Gross motor capacity for children classified as GMFCS level III peaks at a mean age of 7 years 11 months, and GMFCS IV peaks at 6 years 11 months, with a subsequent clinically significant decline thereafter.[6] For marginally ambulant children with CP (uses walker or crutches) and non-ambulant (uses wheelchair), the decline in physical functioning over time means up to 87–96% of each waking day can be spent sitting.[7] Interventions to reduce physical disability and promote independence in daily life skills, inclusion and community participation are major research priority areas identified by consumers.[8]

Traditional neurodevelopmental interventions, based on passive movement experiences and with the concept of 'normalisation' are ineffective in improving motor outcomes for children with CP.[9 10] Our International Clinical Practice Guideline of Functional Therapy[11] and systematic review[10] identified that to improve walking speed and distance, over ground walking is recommended and can be supplemented with treadmill training (with/without partial body weight support). Cycling training may additionally improve muscle strength, gross motor function, metabolic health and cardiorespiratory fitness.[12] Task-specific training of self-selected gross motor goals such as sit-to-stand and transfers, appear promising to improve attainment of individualised goals.[10] The majority of current evidence to support intensive rehabilitation, is however, for children with mild-to-moderate CP (GMFCS I–II), with more limited data for improved motor capacity in children classified GMFCS III–IV.[11] Evidence indicates that motor learning-based interventions for children with CP need to be intensive, specific, repetitive, incremental and challenging in order to improve motor performance.[13] There is little evidence, however, to suggest that targeting motor capacity alone will have a downstream impact on children's participation in community-based physical activity.[14]

Children with CP participate less frequently and in fewer types of physically active leisure compared with their typically developing peers.[15] With increasing severity of motor impairment, there are less opportunities and children with CP participate in even fewer physically active leisure pursuits.[16] These children are at a greater risk of physical inactivity with more time spent sedentary and less time engaged in moderate-to-vigorous physical activity (MVPA).[4] The natural history of decline in gross motor function with age, particularly for children GMFCS III and IV,[6] further compounds this problem. Models of rehabilitation that address personal, contextual and environmental barriers to participation, may be more effective in enabling participation in physically active leisure and increasing levels of physical activity.[14] Embedding rehabilitation in context and supporting children and families to integrate active practice of their gross motor and leisure goals into their daily routines is essential for the long-term maintenance of treatment gains and ongoing participation.[17]

Results from two pilot randomised controlled trials (RCTs) have informed the development of a new intervention called Active Strides-CP for children classified in GMFCS III and IV. A package of functional electrical stimulation (FES) cycling, goal-directed training and recreational cycling was compared with usual care (UC) in a single-blind waitlist RCT for school-aged children with CP (GMFCS II–IV).[18 19] Twenty-one children were randomly allocated to either an intervention group, who received 24 hours of intensive FES cycling, goal-directed training and recreational cycling or a control group who received UC. The intervention group had significant and clinically meaningful gains in gross motor capacity on the Gross Motor Function Measure (GMFM-66, mean difference (MD)=5.9; 95% CI 3.1 to 8.8; p<0.001), achievement of goals on the Canadian Occupational Performance Measure (COPM, MD=4.4; 95% CI 3.9 to 5.3; p<0.001) and higher levels of pedalling resistance during FES cycling (MD=3.4; 95% CI 1.0 to 5.8; p=0.009) immediately post-training compared with the waitlist group.[18] Additionally, recreational adapted cycling provided children with CP an enjoyable option for longer-term participation in physically active leisure.[20 21] In a second RCT, FES robotic-assisted gait training (RAGT) plus goal-directed overground walking in gait trainers was compared with partial body weight-supported treadmill training (PBWSTT) plus goal-directed overground walking in gait trainers.[22] Forty children in GMFCS III to V were randomly allocated to 18 hours of either intensive FES RAGT and overground walking (n=20), or PBWSTT and overground walking (n=20). The findings supported a non-differential effect on gross motor function, walking speed, burden of care and goal attainment.[22] Both interventions demonstrated significant improvements in all outcomes over time at both the 6-week post intervention and 6-month follow-up compared with baseline suggesting that FES RAGT and goal-directed overground walking were not more effective than PBWSTT and goal-directed overground walking. The PBWSTT group had significant improvements in gross motor capacity on the GMFM-66 (MD 2.2; 95% CI 0.8 to 3.5; p=0.001), walking speed on the 10-metre walk test (MD 0.6s; 95% CI 0.4 to 0.8; p=0.002), goal attainment on the Goal Attainment Scale (post intervention: MD 22.3; 95% CI 17.6 to 27; p<0.001) and perceived goal performance on the COPM (MD 3.3; 95% CI 2.5 to 4; p<0.001) and when compared with baseline.[22] This study demonstrated that simplified equipment using PBWSTT and goal-directed overground walking using gait trainers was effective and feasible in non-ambulant children with CP to improve gross motor function, walking speed, goal attainment, performance and satisfaction and reduced burden of care.[22]

In our new Active Strides-CP study, we combine the effective elements of both pilot RCTs in a programme

of physiotherapy which combines (1) FES cycling; (2) PBWST; (3) goal-directed training; (4) overground walking and (5) adapted community recreational cycling, with the additional consideration of environmental barriers to participation.[17] We propose that these elements will complementarily work together to facilitate children attaining individualised motor and leisure goals, and overarching motor, physical activity and participation outcomes.

METHODS AND ANALYSIS
Objectives
This single-blind multisite RCT will investigate whether an 8-week Active Strides-CP intervention compared with UC will lead to greater changes in gross motor function immediately post intervention for school-age children with moderate-to-severe CP (GMFCS III, IV). Secondary outcomes will include habitual physical activity ((HPA), time spent sedentary, time in light and/or MVPA, cardiorespiratory fitness (heart rate/Physiological Cost Index (HR/PCI)), walking speed and distance, frequency/involvement of community participation, mobility performance, attainment of gross motor goals, healthcare use and quality of life immediately post intervention and with retention of outcomes 6 months post-baseline.

Trial design
Active Strides-CP is a pragmatic, single assessor-blind randomised controlled, multicentre trial with two parallel groups. The primary time point is immediately post-intervention (9 weeks post-baseline) and the secondary time point is 18 weeks post-intervention (26 weeks post-baseline). The study will be conducted in four Australian cities across five sites, Brisbane (Queensland Children's Hospital; n=40), Sydney (Cerebral Palsy Alliance; n=40), Melbourne (Royal Children's Hospital and Monash Hospital; n=40) and Perth (Healthy Strides Foundation; n=30). Randomisation will be stratified according to GMFCS (III or IV) and age bands (5–10 and 11–15 years), then randomised centrally (Queensland site) to receive either Active Strides-CP or UC using an electronic allocation system determined by non-study personnel via REDCap (Research Electronic Data Capture).[23] Children allocated to UC will receive Active Strides-CP after the 26-week follow-up assessment.

Eligibility criteria
Participants eligible for the trial must comply with all of the following eligibility criteria at randomisation: (1) diagnosis of bilateral CP (diplegia/quadriplegia) and classified in GMFCS levels III or IV; (2) between 5–15 years; (3) have goals to improve community mobility and cycling; (4) able to attend training, testing and follow-up sessions; (5) able and willing to follow instructions to perform study assessments and intervention (as determined by the assessor at baseline); (6) not expected to undergo lower limb orthopaedic or neurosurgery (eg,

selective dorsal rhizotomy) during the study period. If they have received lower limb orthopaedic surgery then study entry will be delayed until they are 12 months post-surgery; (7) medically fit to undertake moderate intensity exercise; (8) adequate range of motion (ROM) in their hips, knees and ankles to complete a full revolution of the crank arm; and (9) able to verbally or non-verbally communicate pain or discomfort.

Participants will be excluded if the child has: (1) lower limb joint contracture, severe spasticity or severely reduced ROM that limits the ability to complete a full cycling revolution; (2) uncontrolled epilepsy (not controlled by medication) as this would be a confounder; (3) surgery, trauma or fractures in the preceding 12 months without medical clearance to participate; (4) cardiovascular or pulmonary diseases without medical clearance to participate in the 8-week intervention.

Interventions
Active Strides-CP group
Dose
A total training dose of 32 hours of direct therapy will be delivered over 8 weeks. This will be achieved through:
1. Two times weekly 1.5-hour clinical sessions of motor training comprising a rehabilitation package of up to 30 min per session each of (i) FES-assisted cycling, (ii) PBWSTT and (iii) over ground walking training and planning/revision of the goal-directed home exercise programme (HEP) (based on functional goals targeted at activity performance or participation).
2. Minimum of two to maximum of four fortnightly 1-hour home visits to practice recreational cycling (individualised adapted bike), over ground walking (using gait trainers) and goals in context, and on alternating weeks.
3. Minimum of four to maximum of six fortnightly 1-hour telehealth sessions to support implementation of the home programme (on alternate weeks to the home visits).

Mode
Individual intervention with a ratio of 2:1 therapist and therapy assistant to child for the onsite training.

Content and tailoring
All elements of this package of rehabilitation are task-specific and involve active practice underpinned by motor learning theory. Three key elements of Active Strides-CP work together complementarily to target all ICF (International Classification of Functioning, Disability and Health) levels, with the ultimate aim of increasing community participation and levels of physical activity with consequent health benefits (figure 1).

FES cycling[19]
All cycling will be completed on an FES cycle, which allows users to cycle from their own wheelchair or chair.[24] Each clinic session comprises three 10-min cycling phases.
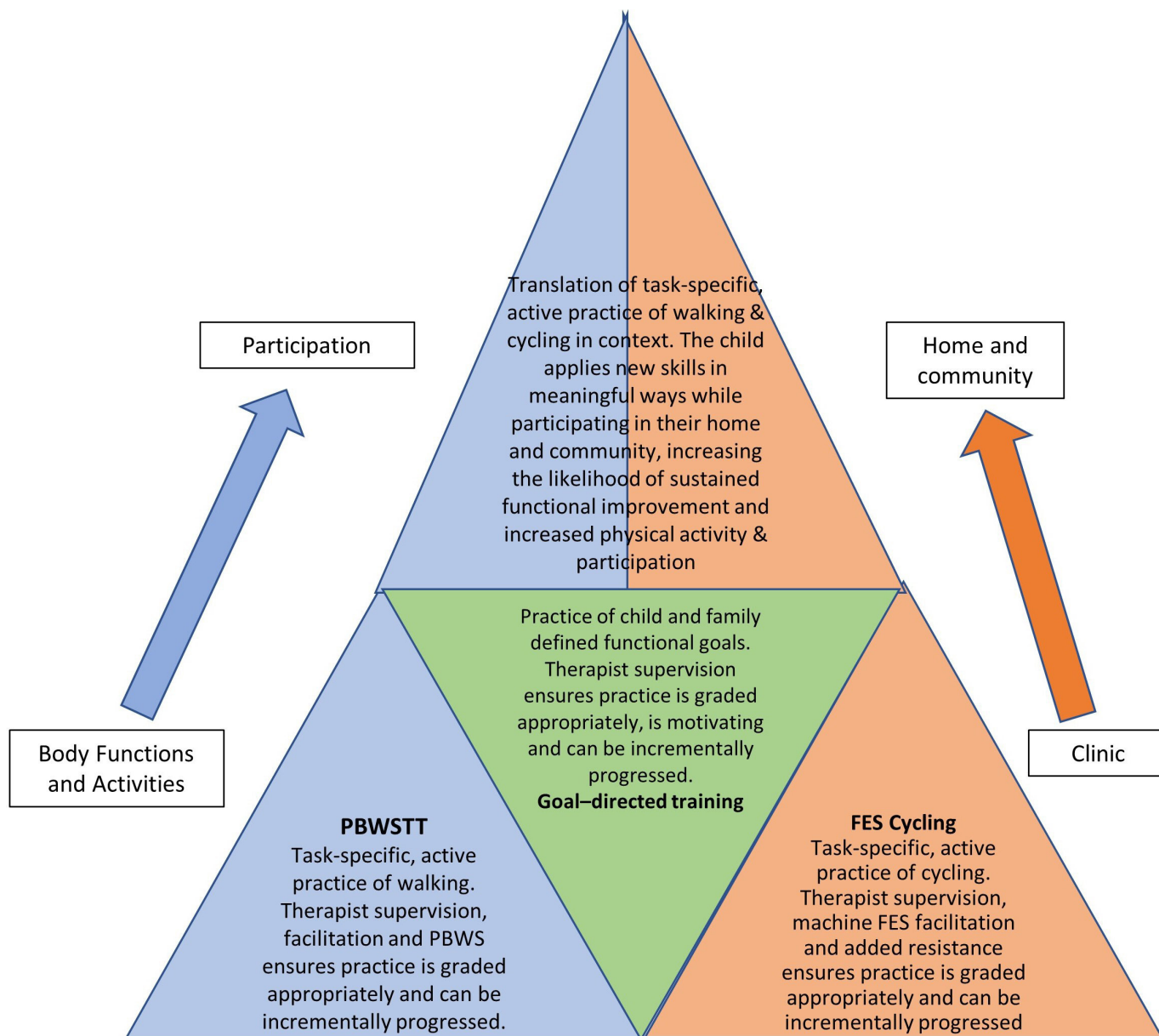
Figure 1 Components of Active Strides-CP across ICF levels and settings. FES, functional electrical stimulation; ICF, International Classification of Functioning, Disability and Health; PBWSTT, partial body weight-supported treadmill training.

1. Phases I and III: Children cycle at self-selected cadence speed up to a maximum of 45 revolutions per minute (rpm), aiming to progress speed, resistance and power output each session. Resistance will be added if children can cycle faster than 20 rpm and will be set to a level that the child can complete 10 min of cycling.

2. Phase II: All-out sprints up to 30 s long, separated by 20–30 s periods of active recovery (self-selected, comfortable cadence with minimal resistance). Sprints will be completed at 80–100% of maximum power output, resistance set to the highest level achieved in phase I and further increased as required (eg, if the child appears to easily and effortlessly achieve high rpms). Motor support will be automatically initiated if cycling falls 20–25% below the target speed.

Intensity will be monitored by a Polar M600 HR wrist monitor or Polar Verity Sense Optical Heart Rate Monitor, with the aim of >60% of age-predicted HR maximum in phases I, II and III and >80% in phase II sprints. FES stimulation parameters are adjusted based on the participant's tolerance.[25–27] Muscle groups include bilateral gluteal, hamstring, quadriceps, gastrocnemius and tibialis anterior. Global starting frequency of 40–50 Hz FES stimulation is used for all muscle groups.[25] A frequency of 50 Hz falls close to the beginning of the plateau of the force-frequency curve for the quadriceps and hamstrings of children with CP.[25 28]

A cycling sprint test will be completed at the initial FES-cycling session to determine the participant's target power outputs for the cycling protocol. Participants

will complete three, 10-s sprints on the cycle ergometer (no stimulation or motor support) and the peak power output will be recorded from the ergometer's display panel. Participants will begin cycling at a comfortable speed for 10–30s with resistance set at the minimal level of 0.5 Nm. If participants can cycle faster than 20 rpm or if they feel as though their feet are flying off the pedals, resistance will be increased to a level that is comfortable. Participants will then be asked to cycle as hard and as fast as possible for three 10s sprints, separated by 30s of passive cycling. The peak power output achieved during the sprints will be used to determine the target training power output for the first training session. In subsequent sessions, the sprint resistance and target power output will be calculated based on the highest peak power and resistance achieved during the previous cycling session. If participants require motor support to initiate or maintain a constant pedalling motion, the power output reflects the amount of work completed by the individual above the level of assistance provided by the motor.

PBWSTT[22]

Cycling training will be followed by PBWSTT which comprises three by 10-min sets, separated by a 2-min rest period. Training will be completed on a treadmill with an overhead treadmill hoist and walking sling/harness. Level of weight support will be adjusted to maximise bilateral lower limb weight bearing and facilitate ease of foot clearance during the swing phase of gait. Each set comprises of facilitated stepping (2 min) followed by independent stepping (30 s). During the 2-min facilitated stepping, initial body weight support will be provided at 60% of the child's body weight at a speed that matches the child's 10-metre fast walk test (10mFWT) speed. Facilitation is provided by a physiotherapist and therapy assistant positioned either side of the child. Standardised hand positioning will be adopted during the swing and stance phase. Speed will increase by 0.1 km/hour increments at a time. If the participant is able to maintain foot clearance during the swing phase of gait and heart rate remaining below 70% of maximum, speed can be increased by 0.1 km/hour at a time. If the walking speed is limited to 0.8 km/hour (the lowest speed in many commercial treadmills), body weight support will be increased by 10% at a time to enable foot clearance during the swing phase of gait. After the 2 min of facilitated stepping, the child will then be asked to generate independent stepping without facilitation for 30-s intervals with the treadmill speed set to match their overground walking speed (measured by their 10mFWT) with body weight support remaining the same as the proceeding 2-min interval. During the 30-s independent stepping interval, verbal prompts and props will be used to encourage consistent stepping and timing of steps. The aim in this interval is to reduce body weight support by 10% at a time while maintaining the set speed. If the child is able to maintain stepping with only 10% body weight support, the speed can then be increased by 0.1 km/hour. During the rest break between 10-min sets, children will be encouraged to stand as actively as possible while engaged in a play activity.

Overground walking and progression of goal-directed HEP

The remainder of the training session will be dedicated to practice of overground walking and reviewing and progressing the goal-directed HEP described below.

HR response will be monitored in each clinical session to determine target training intensity (set to 70% maximum HR) using a Polar M600 HR wrist monitor or Polar Verity Sense Optical Heart Rate Monitor. The HR wrist monitor will be applied at the commencement of the session to capture HR response throughout the FES cycling, PBWSTT and goal-directed training. If heart rate exceeds the 70% maximum HR, body weight support will be decreased by 10% at a time.

HEP, home visits and telehealth support for the HEP

Therapists will conduct a total of eight 1-hour home or telehealth sessions over the 8-week intervention to support participants with the HEP. These will consist of a minimum of two to maximum of four home visits and four to six telehealth consultations.

On week one of the 8-week programme, a home visit will be conducted using client-centred problem-solving style of communication such as Motivational Interviewing. Two to three functional goals with a focus on mobility and/or recreational cycling will have been set as part of the baseline assessment process using the COPM.[29] The goals will align to either the Activity or Participation domain of the ICF. The domain of the goal/s will be recorded. An example of an Activity goal might be 'to independently mount adapted cycle with supervision'. The goals should have the following features to ensure that they are appropriate, specific and repeatable (table 1).

Following a participation-focused framework,[17] a collaborative approach between the parent, child and therapist will be used to:

1. Identify possible barriers and facilitators to undertaking the participant's cycling and mobility-related goals in the home/community,
2. Undertake an environmental screen to develop the HEP and understand any interactions between the child, family, goal and context/environment. If possible, directly observe an attempt at the goal activity or ask for a video of an attempt at the goal, and,
3. Trial and assess cycling and gait equipment and make modifications or adaptations to the equipment if required. Some examples of simple adaptations that could be made to cycling equipment include: adding a supportive backrest for a participant with reduced trunk control or endurance; replacing standard handlebars with looped handlebars for a participant with limited grip strength, or moving a handbrake to the participant's dominant side. More complex modifications, such as switching a free-wheel mechanism to a fixed-wheel mechanism will need to be completed by a qualified bike mechanic.

Table 1 Features of activity and participation goals for Active Strides-CP

Domain	Features	Example
Activity	► Reflects activity competence construct of the Family of Participation-related constructs (fPRC).	► 'I will transfer from my school chair to my Kaye walker in my classroom with set-up assistance'.
	► Contains a specific activity (eg, walking using my Kaye walker, transferring from my wheelchair to my Kaye walker).	► 'I will independently transfer from my bed to my Kaye walker'.
	► Contains a context where relevant (eg, classroom, bedroom, toilet), because features of that context are important (eg, rails, ramps, surfaces).	► 'I will independently walk using my Kaye walker from my car to my classroom at school'.
	► Says whether any assistance is part of the target goal (eg, independently, with the assistance of one adult, with standby assistance, with set-up assistance).	► 'I will transfer from my car to my Kaye walker with the light assistance of one adult for safety'.
	► Contains relevant information about performance to make the goal measurable (eg, the number of minutes, turning corners, smoothly, safely).	► 'I will cycle independently for 500 m using my trike with even, consistent pedalling motion'.
Participation	► Reflects participation frequency or involvement constructs of the fPRC. ► Contains an activity or routine (eg, cycling, walking). The activity does not need to be as specific compared with an activity goal. ► Contains a context (eg, home, school, neighbourhood, skate park). ► Says who with if relevant (eg, with friends, with family, with others, on my own). ► For frequency goals, gives a target frequency (eg, once per week, twice per month) and if decided to be relevant, a target number of minutes (eg, 30 min).	**Frequency:** ► 'I will ride my trike outside of therapy twice a week for 30 minutes around the neighbourhood with my mum'. ► 'I will walk my dog Betty around the block with my dad 5 days a week'.
	► For involvement goals, gives the concept (eg, engagement, persistence, social connection, affect, motivation, enjoyment). ► Participation goals SHOULD NOT contain elements related to activity competence.	**Involvement:** ► 'I will persist for the whole bike ride with my friends in my neighbourhood, even if I think I am I can't keep up'.

Subsequent home visits will be used to:

1. Support child and family in the HEP which comprises task-specific functional training specifically relevant to goals (eg, dismount off a bike with close supervision). Training will follow established principles including graded tasks and is provided in context to generalise it to other settings and ends in participating in the activity itself 'in context'. Adult involvement is to build capacity by modelling, instructing and allowing opportunities to practice supporting the child with therapist feedback on the behaviour. The child is also provided with verbal feedback on the behaviour. Task practice is ideally 'whole task' but part task practice is used where necessary to build capability towards the goal.

2. Support the family to achieve their chosen Activity and Participation goals using a client-centred, problem-solving style of communication such as Motivational Interviewing.[17] This approach promotes intrinsic motivation for health behaviour goal attainment of children with CP and their families, through autonomy support according to the principles of Self-Determination Theory.[30] Practical help and guidance from the clinician (to help families solve problems related to their child's participation and movement) is a common feature of effective participation-focused interventions in children and young people with CP and childhood-onset disability, especially for physical activity.[31]

HEP duration
Therapists will directly support 1 weekly, 60-min HEP session via home visiting or telehealth. The 60-min session will be divided into 20–30 min to practice each identified goal. Children and families will be encouraged to continue to practice their HEP, including recreational cycling throughout the remainder of each week. The amount of time will not be specified for home practice outside the structured sessions.

HEP activities
Goal-directed training component
Goal-directed training will comprise whole±part task practice aiming for a minimum of 20 min per goal. Goal-directed training will adhere to best practice principles for functional therapy, which prioritises the concept of active motor learning. This means that the properties of the task or the task environment (including instructions and verbal or visual feedback) will be altered in the first instance to elicit changes in the child's performance. 'Hands-on' feedback, facilitation or physical

handling will be minimised as much as possible during this component.[11]

Adapted cycling component

The cycling component will be completed using adapted tricycles or recumbent bikes. The adapted recreational cycling aims to bridge the gap between commencing cycling in a clinical setting (FES cycling which aims to build capacity of cycling speed and endurance) and translation to cycling in a community-based context. Therapists will work with families prior to entry to the study to secure their own adapted bike. If participants do not own their own bike and helmet, each site will have a small number of available bikes that can be loaned to the family for the duration of the study. Additional bikes or attachments such as backrests, adapted pedals and handlebars may be sourced on a case-by-case basis if an appropriate bike is not available in the existing equipment pool. Participants will continue to have access to loan bikes during the retention period (T2–T3). The cycling component will take place in a safe area that is identified by the family and therapist at the initial home visit. If there are no safe places to cycle near the participants home, participants will be encouraged to transport their bike to a local bike path or park.

Overground walking

Overground walking will be completed with the support of the child's own assistive mobility device. For children classified within GMFCS level III, equipment may include elbow crutches or a gait trainer with limited body support. The aim for overground walking in children classified within GMFCS level III may be to develop more functionally independent walking within indoor and outdoor environments, steps or stairs. For children classified within GMFCS level IV, suitable equipment may include supportive gait trainers. The aim of overground walking is for task-specific practice which may include transfers (in and out of their walker), walking in indoor environments or walking in busier cluttered environments that require steering and negotiating obstacles. The principle of overground walking is whole task practice within environmentally relevant contexts. A 'hands-off' approach will be adopted to encourage the child to initiate their own movements. Occasional guidance and support may, however, be provided to demonstrate strategies to turn and steer in the walker or to provide feedback when experiencing new movements. Incremental challenges will be introduced throughout the 8 weeks. This includes reducing physical support within the gait trainer, increasing resistance on the gait trainer, reducing physical or verbal prompts for stepping or increasing the variability of practice such as introducing obstacles, visual clutter or altered surfaces as directed by the goals set by the child and their family.

Participants will record in a diary the amount of time spent each week on goal-directed training, cycling and walking training.

Intervention providers

Physiotherapists or exercise scientists/physiologists and allied health assistants will complete standardised training to deliver the Active Strides-CP programme.

Location

The intervention groups will be conducted in the clinics in each of the participating sites for two sessions per week and in the home for four fortnightly visits and four tele-health consultations.

UC

UC over the 6-month waitlist period will vary across Australia and could range from intensive therapy blocks, weekly clinic-based therapy sessions to school-based consultative services provided on a monthly, quarterly or yearly basis. All families in both groups will complete a UC diary for the duration of study involvement to record the frequency and duration of therapy, exercise activities and any concurrent medical interventions such as intramuscular botulinum toxin A injections and/or serial casting. All children in the UC group will be offered Active Strides-CP after the 6-month retention time point.

Therapist training and fidelity
Therapist attributes

It is required that therapists possess the following attributes:

▶ Full registration with the Australian Health Practitioner Regulation Agency (physiotherapists) OR full members with accreditation from Exercise & Sports Science Australia (exercise scientists/physiologists).
▶ Current Basic First Aid and Cardiopulmonary Resuscitation certificate.

It is highly desirable that therapists possess the following attributes:

▶ 3+ years' experience working with children with cerebral palsy and their families.
▶ Experience working within models or frameworks of motor learning.

Therapist training, treatment manual and ongoing peer-to-peer support

Standardised therapist training will be provided to therapists employed to deliver the intervention. The training will specifically target FES cycling, PBWSTT, overground walking, goal setting and goal-directed training, Motivational Interviewing techniques, home programmes and adapted cycling. Training sessions will be overseen by the chief investigators (CIs) who are experienced in each aspect of Active Strides-CP. Training sessions will be video recorded and accessible at any time for established or new therapists delivering the intervention. A comprehensive treatment manual will be developed to support therapists in the implementation of Active Strides-CP. Therapists across all sites will attend monthly meetings with CIs to problem solve challenges in delivery of the Active Strides-CP intervention.

Fidelity

Individual adherence to the training manual will be recorded on a session-by-session basis by therapists including (1) percentage of sessions attended (including partial attendance); (2) session content checklist to record the percentage of each component of the intervention was achieved, documenting speed, body weight support, distance covered and active engagement within the programme; (3) percentage of session duration spent within the target HR threshold for training intensity. The first session that is conducted by a site therapist for a child that is GMFCS level III and IV will be video recorded and reviewed by one CI (DP) to provide feedback. A session in week 5 and 9 will also be video recorded for the same child for review (DP) and provide feedback for progression of the programme and adherence to the protocols. In addition to this, each therapist will record a session three times a year throughout the study course duration. Intervention parameters and adherence will be reviewed based on the core competency checklist. Reasons for missed or incomplete sessions will be recorded. Adherence data will be reported alongside study outcomes.

Outcomes

Three measurement time points will be taken: baseline (T1); immediately post-intervention at 9 weeks post-baseline *primary endpoint* (T2); 26 weeks post baseline *retention* (T3). Children allocated to the waitlist group will be offered Active Strides-CP following the 26 weeks retention time point.

Primary outcomes at primary endpoint (T2) and retention (T3)
GMFM-66

The GMFM-66 is a criterion referenced observation measure developed using Rasch modelling to measure gross motor function of children with CP.[32] The GMFM-66 has established construct validity, high test–retest reliability (intraclass correlation coefficient (ICC) 0.99)[32] and is responsive to change (Minimal Clinical Important Difference - MCID=3).[32–34] The GMFM-66 will provide an overall measure of gross motor function capacity. As the GMFM-66 domains D (standing) and E (walking) contain all the items of the GMFM-88, these domains will also be reported for these specific motor skills.[35]

Secondary outcomes
HPA

HPA will be measured using triaxial accelerometers positioned on the least impaired wrist (ActiGraph GT3X+, Pensacola, Florida, USA) and least impaired anterior thigh (Axivity AX3 Axivity Ltd, Newcastle, UK). Accelerometry is valid, reliable and feasible in children with CP.[36 37] Accelerometers will be fitted during assessment and worn during waking hours for seven consecutive days.[38] Raw accelerometer data from both devices will be processed into HPA metrics using machine-learnt random forest PA classification algorithms specifically trained and validated for assessing HPA in youth with CP

who use mobility aids for ambulation.[39 40] The algorithms use statistical and frequency domain features in the raw acceleration signal to identify activity type and quantify time spent in sedentary activities (sitting or lying down), standing utilitarian movements (light intensity), comfortable walking and brisk walking. Participants are provided with a log sheet and are asked to record the following information about each instance either accelerometer is removed and return the log sheet with the accelerometer: device/s removed, date removed, time removed, what the child was doing while the device/s were removed, date replaced and time replaced.

Walking endurance: 6-minute walk test

This simple, submaximal test measures the distance walked over 6 min, provides information about endurance during functional activities.[41] It has excellent test–retest reliability (ICC=0.98) in CP.[42] Percentile curves have been created on 1445 typically developing children aged 7–16 yrs.[43] The test will be performed according to guidelines of the American Thoracic Society on a 30-min course.[44] The test requires participants to walk as far as possible in 6 min using a 10-metre track with cones demarcating the turning points. Participants will be given verbal and visual instructions before testing. Participants will be instructed to walk as far as possible without running in 6 min. Participants will be given verbal encouragement and every 30 s will be advised of the distance covered (in laps) and the time remaining. Distance will be measured to the nearest 5-metre mark.

Walking speed: 10mFWT

The 10mFWT tests maximal walking speed over a 10 metre distance, considered the minimum for functional ambulation. It has moderate test–retest reliability for CP (ICC 0.81).[45]

Mobility: pediatric evaluation of disability inventory computerised assessment test

The paediatric evaluation of disability inventory computerised assessment test (PEDI-CAT) is a standardised, norm-referenced assessment of independence in self-care. The test is valid, reliable and responsive in this population.[18] The PEDI-CAT is completed by parents using an iPad application. The item bank of the PEDI-CAT was developed using Rasch measurement modelling on large samples of typically developing children and those with disabilities. The Mobility domain will be completed by caregivers.

PCI

PCI will be measured using HR throughout each intervention session on a Polar Heart Rate Monitor aiming to achieve MVPA (50–70% hour max).[46] Test–retest reliability is high in children with CP (ICC 0.82–0.99).[47 48] The Polar Heart Rate Monitor will be worn during the 6-minute walk test (6MWT).

Performance and satisfaction with occupational performance goals
The COPM[29] will be used to measure performance of and satisfaction of individually defined goals. Children will set a maximum of three goals to target in the intervention directly related to functional mobility (eg, transfer to walker) or cycling (eg, mounting or dismounting cycle, steering). Test–retest reliability is high (ICC 0.76–0.89). It is responsive to change, where a change of ≥2 is considered clinically significant.[49]

Intensity and frequency of home, school and community participation
Participation and Environment Questionnaire (PEM-CY)[50 51] is a parent completed questionnaire with good test–retest reliability and internal consistency.[50] Summary scores for participation frequency, involvement and environmental supportiveness will be evaluated. The PEM-CY will be completed at baseline only and will be used as a covariate in post hoc analyses.

Quality of life
The CP-QOL Child is a 52-item, condition-specific self-report measure of child quality of life (QOL) that is specifically developed for measuring QOL in children with CP. The domains covered in the child self-report version include physical well-being, social well-being, emotional well-being, school and acceptance by others. It has good concurrent validity, internal consistency (Cronbach's alpha 0.80–0.90) and test–retest reliability for children 9 years of age and over. The CP-QOL Child will be completed by all children aged 9 years and older. An adult who is not participating in the study as the primary parent/caregiver will read the questionnaire alongside the child and clarify the meaning of the questions and response scale if necessary. The CP-QOL Teen self-report will be completed by children aged 13–15 years. The CP-QOL Primary Caregiver version will be completed by the child's primary caregiver for children aged between 5 and 12 years and the CP-QOL Teen Primary Caregiver version will be completed by the child's primary caregiver for children aged between 13 and 15 years.

The Child Health Utility Index (CHU-9D) is a paediatric health related quality of life measure for use in economic evaluation. The measure consists of nine questions. Children can self-report from 7 years of age and parents can proxy report for their child. In this study, the CHU9 will be completed by the child's primary caregiver.[48]

Health economics
A within trial economic evaluation will be conducted to synthesise the costs and outcomes of the Active Strides-CP training programme and estimate the cost-effectiveness of Active Strides versus UC. Resource usage (staff time, equipment and facility use) associated with the programme will be collected alongside the RCT. Healthcare usage will be assessed using a resource use questionnaire previously used in CP child studies[52] and linked Australian Medicare claims data that will provide medical services and medication usage. Utility will be derived from the CHU-9D,[53] a generic child quality of life measure designed specifically for economic evaluation and which has been validated in an Australian population.[54] Incremental cost-effectiveness ratios will be estimated and appropriate one way and multivariate sensitivity analyses will be undertaken as in previous RCTs by our group.[50]

Classification systems and demographic characteristics
The following validated classification systems will be applied: GMFCS,[55] Manual Abilities Classification System,[56] Communication Function Classification System.[57]

The following participant demographic characteristics will be collected to characterise the sample: age, sex, primary motor type, distribution and presence of comorbid diagnoses.

Participants will also be screened for conditions that may be considered a precaution to high intensity exercise, and thus requiring attention or adaptation (eg, known cardiovascular or respiratory condition).

Participant timeline
Active Strides-CP schedule of assessments and interventions are provided below in table 2 and the Consolidated Standards of Reporting Trials[58] participant flow diagram is provided in figure 2.

Recruitment
Families with a child meeting eligibility will be invited to join at four collaborating sites (Queensland, New South Wales, Victoria and Western Australia) and associated clinical services (Queensland Children's Hospital, Cerebral Palsy Alliance, Healthy Strides Foundation, Royal Children's Hospital and Monash Hospital). Recruitment from five major centres will enable the target sample size to be achieved. As there are a predicted 500 children with moderate-to-severe CP (Australian CP Register), aged 5–15 years, recruitment of 150 children is feasible. The CIs have achieved high recruitment and retention in RCTs of school-aged children with moderate-to-severe CP.[59]

Recruitment at each site will commence once ethical and governance approvals have been obtained. The first participant was enrolled and randomised on 1 August 2022 and the study is expected to be complete by 30 June 2026. Recruitment will draw on current databases within each organisation, referrals from clinical services and the respective State Cerebral Palsy Registers. Contact with participants will occur via one of the following mechanisms:

i. Child name, basic characteristics and family contact details are identified on a Clinical Trials Register, clinical and/or research database hosted by one of the partner institutions.
ii. Families who consent to receive information about clinical trials will be sent up to two emails and one

Table 2 Schedule of assessments for Active Strides-CP study

Assessment/procedure	T1 baseline assessment	T2 follow-up assessment 9 weeks	T3 follow-up assessment 26 weeks	T4 follow-up assessment waitlist group only
Informed consent	x			
Demographic information	x			
Primary outcome				
GMFM	x	x	x	x
Secondary outcomes				
6MWT	x	x	x	x
10mFWT	x	x	x	x
PEDI-CAT – mobility (P)	x	x	x	x
COPM (child >8 years and P)	x	x	x	x
PEM-CY (P)	x			
CPQOL (child/adolescent self-report and P)	x	x	x	x
CHU9 (P)	x	x	x	x
Health Resource Usage Questionnaire (P)	x			x
ActiGraph and Polar OH1	x	x	x	x

CHU9, Child Health Utility; COPM, Canadian Occupational Performance Measure; CPQOL, Cerebral Palsy Quality of Life Measure; GMFM, Gross Motor Function Measure; 10mFWT, 10-metre fast walk test; 6MWT, 6-minute walk test; P, parent; PEDI-CAT, Paediatric Evaluation of Disability Inventory – Computer Adapted Test; PEM-CY, Participation and Environment Questionnaire.

postal package with approved trial invitation letter and flyer.

a. The Study Coordinator and/or site therapist will then follow-up with a phone call with families (at least 1 week later) to ascertain interest in the study.

Families who indicate interest will be sent the participant information and consent forms and contacted again after these have been received to discuss enrolment.

Families who indicate no interest will not be contacted again.

iii. Children and families attending a clinical service associated with the project including the Queensland Paediatric Rehabilitation Service (QPRS at the Queensland Children's Hospital (QCH), Brisbane), Cerebral Palsy Alliance (CPA, Sydney) and Healthy Strides Foundation (Perth), Royal Children's Hospital (RCH, Melbourne) and Monash Health Service (Melbourne) will be identified by treating clinicians and provided with a flyer.

iv. Electronic and standard billboards at QPRS/QCH, CPA, RCH, Monash Health Service and Healthy Strides Foundation will display the approved flyer during the recruitment period.

v. A newsletter snippet will be included in the electronic and paper newsletters distributed by Queensland Cerebral Palsy and Rehabilitation Research Centre, Queensland Paediatric Rehabilitation Service, Cerebral Palsy Alliance, Perch Children's Hospital, Royal Children's Hospital, Monash Health Service

and Australasian Academy of Cerebral Palsy and Developmental Medicine.

vi. The flyer and trial information will be posted on the research websites for QCPRRC, CPA, PCH, RCH, Monash Health Service and AusACPDM.

vii. A Facebook page will host the approved trial information and flyer and be shared and 'liked' organically (word of mouth referrals).

Allocation and blinding (masking)

Participants will be randomly assigned to either Active Strides-CP or UC with a 1:1 allocation as per a computer-generated randomisation schedule using the REDCap randomisation module, stratified by GMFCS (III or IV), age bands (5–10, 11–15) and site, using permuted blocks of four to six random sizes. The allocation sequence will be generated and entered by a biostatistician not otherwise involved in recruitment, assessment, or trial conduct. Randomisation will occur after enrolment and completion of all baseline assessments (except for 7-day HPA monitoring). Participants and therapists delivering the intervention will not be blinded to intervention after baseline assessments, as it will not be possible to maintain blinding while they are delivering the intervention. All objective outcome measures will be performed by outcome assessors (physiotherapists and/or exercise physiologists) blind to treatment allocation. A detailed assessment manual will be provided to all assessors outlining the order and administration requirements of each measure. Participants and caregivers will be asked to not divulge their group assignment. Participants and
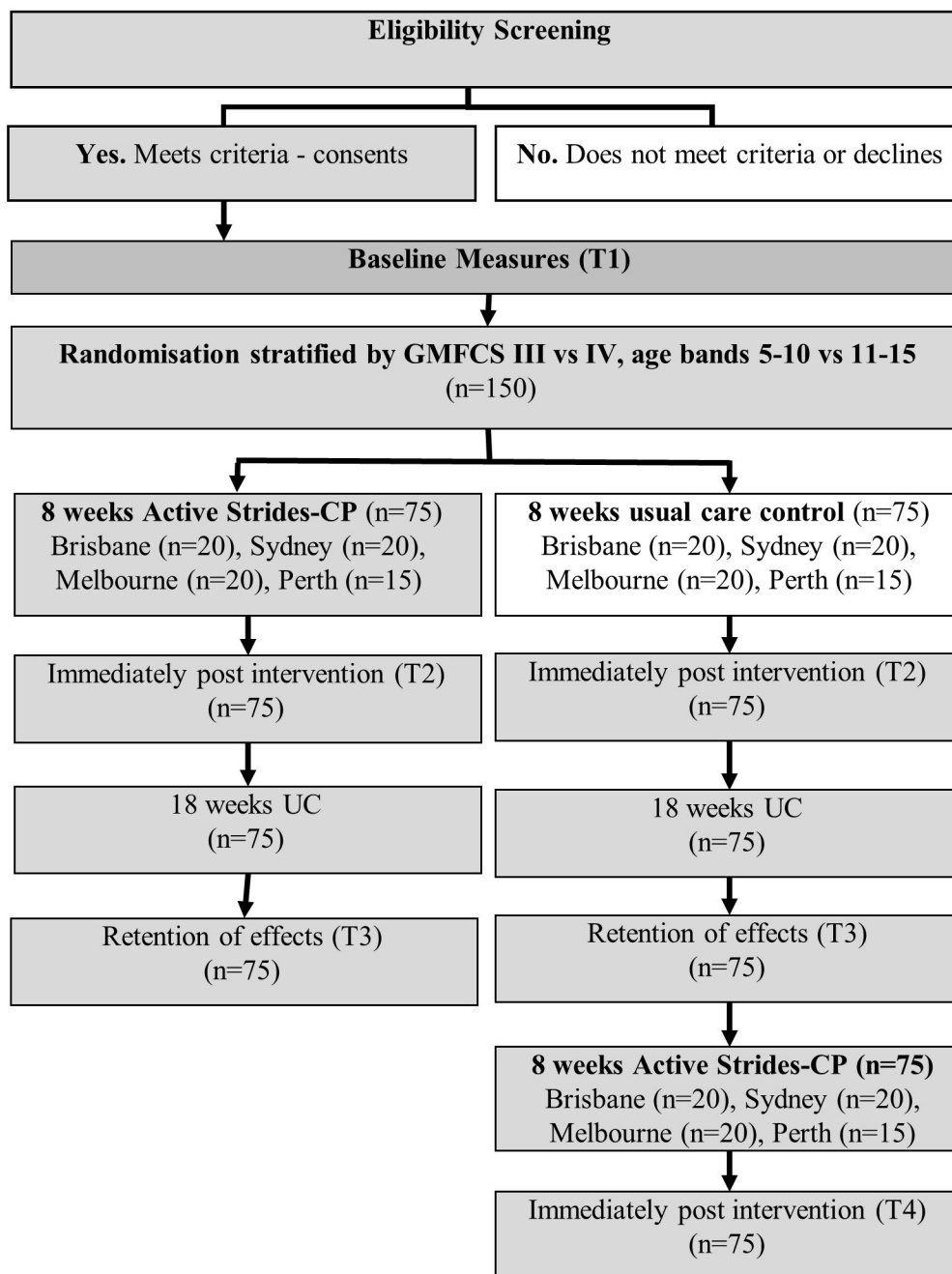
Figure 2 Consolidated Standards of Reporting Trials study flowchart. CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; UC, usual care.

groups will be given a numerical identifier in the data set, which will not be revealed to statisticians and CIs until analysis is completed.

Data management
Data types
Objective data will be collected on gross motor function using the GMFM, HPA using accelerometry, walking endurance using the 6MWT, walking speed using the 10mFWT, PCI using Polar Heart Rate Monitor. All measures are suitable for children with bilateral CP. Subjective measures will be collected from children and are appropriate for use with children over 8 years of age

and for younger children will be completed by their primary caregiver. One questionnaire-based measure of mobility performance will be collected from the child's primary caregiver. All data are re-identifiable.

Data collection
Data will be collected in one of four ways: (a) paper forms; (b) online survey platform (REDCap) instead of/in addition to paper forms; (c) devices (ActiGraph GT3X+, Axivity AX3, Polar OH1, photo/video/audio recording devices) owned by sites/organisations (not personal devices); (d) face-to-face assessments with the child.

Data transfer

Data collected on REDCap will be stored on the secure University of Queensland research server. Data collected on paper forms will be converted into an electronic format by the site therapist, forwarded using a secure file transfer service such as CloudStor and stored on the secure University of Queensland research server or uploaded directly to REDCap. Original paper files will be sent to the Brisbane site via registered post or courier after being de-identified at the conclusion of the data collection phase. Data collected from devices will be downloaded from devices by the site therapist, forwarded using a secure file transfer service such as CloudStor and stored on the secure University of Queensland research server or uploaded directly to REDCap, then deleted.

Data storage

Data (both working and archived data) recorded on paper will be stored at the trial sites in locked filing cabinets during the data collection phase and within an archive box located in the locked filing cabinets of investigators at the Centre for Children's Health Research, South Brisbane Australia at the conclusion of the data collection phase. Data will be stored on secure Australian servers using REDCap (database) and the secure University of Queensland research server. Data will not be destroyed.

Statistical methods

Sample size estimation and justification

The primary basis for sample size calculation is adequate power for H^1 comparison between functional effects of Active Strides-CP compared with UC immediately post intervention. Based on data from two pilot RCTs, we propose a mean difference of 7 logit points (100-Logit Scale)[18] on the GMFM-66 as the minimum difference likely to have substantial clinical impact.[18 22] Data from our two pilot RCTs yielded a pooled SD of 14 logits.[18 22] To detect a between-group difference of 7 logits or greater on the GMFM-66 with 80% power we require 64 children in each group (alpha=0.05). Based on previous RCTs conducted by our group we anticipate a maximum drop-out between randomisation and T2 of 15%,[60] consequently, we will aim to recruit 150 children in total (75 in each group).

Statistical methods to be undertaken

Primary comparison is a between-group comparison of GMFM-66 scores post treatment at 9 weeks. Groups will be compared using linear regression with group (intervention/UC) as the main effect and baseline GMFM-66 scores included as the covariable. For secondary outcomes, we will use similar methods to compare between groups post intervention for continuous outcomes (linear regression), binary outcomes (logistic regression) and count outcomes (Poisson regression). When data from multiple time points is analysed, mixed-effects regression models will be constructed. They will include participant as a random effect to account for non-independence of observations

from the same child. Where continuous data exhibit skewness not overcome by transformation, non-parametric methods such as median regression will be used. Sensitivity analyses will be conducted using imputation techniques to investigate missing data during follow-up. Analyses will follow standard principles for RCTs using two-group comparisons on all subjects on an intention-to-treat basis.

Participant safety and withdrawal

Risk management and safety

The main risk to this project is recruitment shortfalls and participant attrition. Importantly, we have access to large state-wide populations of school-aged children with bilateral CP throughout Queensland, New South Wales, Victoria and Western Australia from state-wide rehabilitation services and the NSW/ACT Cerebral Palsy Register.[16] High recruitment is expected as all children receive the intervention either immediately or after the waitlist UC period. Should further lockdowns occur due to COVID-19, these are likely to be isolated to specific areas/states, therefore having four sites enables recruitment to continue in other sites should one be restricted due to COVID-19. In the event that children miss sessions due to COVID-19 or other illnesses, attempts will be made to reschedule as many missed sessions as possible within the allocated 8-week intervention period.

All staff involved in delivering the intervention will be trained and supervised by senior experienced personnel. Regular daily monitoring will occur and any discomfort reported by the child or their caregiver will be immediately responded to.

The ActiGraph GT3X+, Axivity 3AX and Polar OH1 Optical HR devices pose no health and safety risks and do not cause pain or harm to participants when infection control procedures are followed (disinfection of devices and straps between uses and participants). Discomfort could be caused by tight application or contact allergic reactions to cleaning fluid. This will be monitored closely with verbal prompting, and visual checking of devices and skin contact twice a day. 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. Children will be monitored during each clinic session about comfort/discomfort of the FES electrodes.

Adverse event reporting

Any adverse events associated with Active Strides-CP will be screened at each session using open-ended questions. Adverse events will be documented and reported to the CIs. Major adverse events (requiring medical treatment) will be reported to the Human Research Ethics Committee within 24 hours.

Handling of withdrawals

Participants can withdraw at any time. Participants who choose to withdraw from the study will not be penalised

in any way. If they wish to continue with therapy intervention for their child, they will be assisted to source another local therapy option that matches their preferences. 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. Data will be analysed on an intention-to-treat basis.

Participants/parents are informed that on withdrawal from the study they have the option to have data already collected either destroyed (all information collected about the child can no longer be used for research) or retained (information collected about the child can continue to be used for research). With the understanding that; no further information about the child will be collected for the study from the withdrawal date; information about the child that has already been analysed and/or included in a publication by the study, may not be able to be destroyed; and; choosing to withdraw a child from the study will not affect the child's access to Health Services or Government benefits.

Replacements
Participants that withdraw will not be replaced, as the a priori power calculation will account for a 10% dropout rate and 10% crossover rate.

Patient and public involvement
A person with CP and a parent/caregiver are associate investigators on the study and will coordinate a Consumer Council. The Consumer Council will additionally comprise a representative from each site (either a person with lived experience of CP or a caregiver of a child with CP). The consumer council will meet two to three times per year to provide feedback on all aspects of trial conduct (conduct, analysis and reporting). They will be financially compensated for their time and expertise at the rate of $A50 per hour. This study protocol has been reviewed by two consumer representatives who provided input into the study design.

ETHICS AND DISSEMINATION
Informed consent process
For children and youth <16 years of age, written informed consent will be obtained from the legal guardian.

Ethics and dissemination
Active Strides-CP is registered on the Australian New Zealand Clinical Trials Registry. The project has received ethics approval from the Children's Health Queensland Hospital and Health Service Human Research Ethics Committee (HREC/21/QCHQ/77129), The University of Queensland Human Research Ethics Committee (2021/HE002198), Curtin University (HRE2021-0760) and The University of Melbourne. Results of the study will be published/disseminated in the trial registration database, conference abstracts and presentations, peer-reviewed articles in scientific journals, organisation and institution newsletters and media releases. In accordance with the Australian National Statement 3.1.65, results will be provided directly to participants in an appropriate and accessible format to them.

DISCUSSION
There is a paucity of evidence for effective interventions to improve motor outcomes and community participation for children with moderate-to-severe bilateral CP (who cannot stand and/or walk without gait aids). For these children, high intensity gait and cycling training has recently been shown to improve gross motor function and mobility,[11] but it is not known if this leads to improved participation in home and community life, including physically active leisure.[14] We have developed and pilot-tested new treatment approaches for children with moderate-to-severe bilateral CP and now propose a package of training which combines effective elements of intensive gait (iStride),[22] and cycling and goal directed training (ACTIVATE-CP)[18] that complementarily work together to attain individualised motor and leisure goals, and overarching motor and participation outcomes.

One potential limitation of the study is that there may be interruptions to the protocol due to COVID-related illness of participants and/or staff. To address this, we will maintain the 8-week intervention period, but offer make up sessions if possible to account for missing sessions. All variations will be recorded. UC will be variable and it is not possible to standardise given the multitude of different service providers under pre-agreed funding packages through the Australian National Disability Insurance Scheme. We will record the type and dose of standard care to enable reporting in as much detail as possible.

The study has a number of strengths. The sample size has been calculated for the primary outcome and the inclusion of five different recruitment sites across Australia will ensure that recruitment is feasible. We have included outcome measures with reported and validity and reliability for our population of interest. We have a comprehensive fidelity framework including standardised training of intervention providers, manualisation of the assessment and intervention protocols, that in combination with a within trial cost utility analysis will provide important information to inform future translation of the intervention into clinical practice. We plan that the results of this RCT will be disseminated widely through peer-reviewed journals and academic conferences.

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Contributors LS, DP, RNB and CE conceived the trial. DP, CE and JV developed and tested the protocol for the partial body weight-supported treadmill training. EA, MK, RNB and SH developed and tested the protocol for the Functional Electrical Stimulation cycling component of Active Strides. EA and RT developed the protocol for adapted community cycling. SER, LS, IN and RNB developed the protocol for the home visiting and goal-directed training component of the study. ST developed the protocol for measurement of physical activity using actigraphs and MDP provided expert advice on the implications for longer-term health and well-being. TC developed the protocol for the health economic evaluation. RSW developed the analysis plan for the study. LS achieved study funding as the lead investigator. LS completed the initial draft of the manuscript. DP, LS, RNB, RT, MK, SH, EA, SW and SER developed the treatment manual and therapist training programme. All authors designed the study, have read, edited and approved the final manuscript and supplementary files.

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Competing interests None declared.

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