BMJ Open

A protocol for a single-arm interventional study assessing the effects of a home-based joystick-operated ride-on-toy navigation training programme to improve affected upper extremity function and spontaneous use in children with unilateral cerebral palsy (UCP)

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ABSTRACT

Introduction Children with unilateral cerebral palsy (UCP) face significant limitations in upper extremity (UE) function and require effective interventions that promote intensive goal-directed practice while maximising motivation and adherence with therapy. This study builds on our past work and will assess the effects of a 6-week researcher-caregiver codelivered, home-based ride-on-toy navigation training (RNT) programme in young children with UCP. We hypothesise that the RNT programme will be acceptable, feasible to implement, and lead to greater improvements in unimanual and bimanual function when combined with conventional therapy, compared with conventional therapy provided alone.

Methods and analysis 15 children with UCP between 3 and 8 years will be recruited. During the 6-week control phase, participants will receive treatement-as-usual alone. During the subsequent 6-week intervention phase, in addition to conventional therapy, RNT will be provided 4–5 times/week (2 times by researchers, 2–3 times by caregivers), 30–45 min/session. We will assess UE function using standardised tests (Quality of Upper Extremity Skills Test and Shriner's Hospital Upper Extremity Evaluation), reaching kinematics, wrist-worn accelerometry, caregiver-rated ABILHAND-Kids questionnaire, and training-specific measures of movement control during RNT. Programme feasibility and acceptance will be assessed using device use metrics, child and caregiver exit questionnaires, training-specific measures of child engagement, and the Physical Activity Enjoyment Scale. All assessments will be conducted at pretest, following the control phase (midpoint), and after completion of the intervention phase (post-test).

Ethics and dissemination The study is approved by the Institutional Review Board of the University of Connecticut (# H22-0059). Results from this study will be disseminated through peer-reviewed manuscripts in scientific journals in

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ The single-arm interventional study is designed to investigate the feasibility of implementation and efficacy of a home-based, researcher-caregiver codelivered joystick-operated ride-on-toy navigation programme in children with unilateral cerebral palsy.

⇒ Children will serve as their own controls, and the study will involve a 6-week control phase, followed by a 6-week intervention phase based on principles of motor learning, motivational theories, and family-centric practice.

⇒ Stakeholder input will be obtained from children, clinicians, and researchers to assess feasibility, acceptance, satisfaction, and benefits of the training programme.

⇒ A combination of standardised, quantitative, and caregiver-report measures will be used to assess changes in manual abilities and upper extremity function.

⇒ The study is limited by the lack of a separate control group, small sample size, and challenges in generalising findings to children with UCP with varied motor presentations.

the field, through national and international conferences, and through presentations to parent advocacy groups and other support organisations associated with CP.

Trial registration number NCT05559320.

INTRODUCTION
Background and rationale
Cerebral palsy (CP) refers to a group of complex neurodevelopmental disorders of

movement and posture due to non-progressive damage to the growing brain, which leads to significant activity limitations and participation restriction. The WHO has identified CP to have the highest global burden of disease among all non-communicable diseases. In fact, CP is one of the three paediatric-onset conditions to be included within an initiative aimed at developing a package of rehabilitation interventions as part of WHO's strategic priority of 'Universal Health Coverage' by 2030.[1 2] The healthcare costs per year for children with CP are estimated at US$6 billion, and additional non-reimbursed costs incurred by families run into thousands of dollars.[3] Despite the significant challenges faced by children with CP and their families, clinical practice guidelines to improve motor function and participation among children with CP have only recently been released.[2 4 5] These guidelines overlap in their emphasis on: (A) functional goal-directed, task-oriented, and environmental enrichment approaches that aim to increase participation among individuals with CP, (B) training activities aligned with patient/family goals, (C) encouraging practice of enjoyable activities within real-world environments, and (D) empowering children/families to be actively involved in the decision-making process and management of the condition.[2 4 5] However, the guidelines highlight the need for more rigorous future research to develop effective, evidence-based, child-centric and family-centric interventions to promote independence and participation in children with CP.

Around 40% of children with CP have hemiplegia, that is, weakness on one side of the body, with the upper extremity (UE) typically more affected than the lower extremity.[6 7] Children with unilateral CP (UCP) demonstrate difficulties with reach, grasp, and manipulation, abnormal tone and posturing, poor strength and UE control, slow and uncoordinated movements, and sensory deficits on the affected side.[8–14] Limited hand use is the strongest predictor of activity limitations and participation restriction in children with CP and is one of the most challenging rehabilitation goals for clinicians.[15 16] Although a variety of UE training approaches have been tested, the single most crucial factor associated with improved clinical outcomes, irrespective of the type of training, is treatment dosing.[9 17] Current evidence-based approaches recommend high-intensity repetitive practice for several hours/day, which is often challenging to achieve within clinical practice.[18–21] Moreover, they require considerable therapist expertise and are often time-consuming and cost-intensive.[10 22 23] These limitations have led to efforts exploring the effects of caregiver-led home programmes as cost-effective adjuncts to conventional service delivery models.[7 17 19] Such programmes can help augment treatment dosing, promote UE use within children's naturalistic settings thereby facilitating training carryover outside the clinic/school, and encourage greater family involvement in their child's rehabilitation.[24–28]

Another major factor associated with dosing that is highly predictive of therapeutic improvements is child motivation.[11 29–32] Although high-intensity practice is key to functional success in CP, children frequently find conventional programmes boring.[33–36] Training activities need to be stimulating, fun, challenging, meaningful, and intrinsically motivating for children. Higher child motivation will translate to greater active and self-initiated practice of training activities, increased training volume, and ultimately higher functional gains.[30 37] Contemporary motivational theories including the Self Determination Theory and Expectancy Value Theory suggest that individuals are likely to persist in activities that are age appropriate, intrinsically rewarding, foster autonomy, and provide a sense of achievement while offering the 'just-right challenge'.[30 38–40] Therefore, there is a need for novel, cost-effective interventions involving child-preferred activities that provide families with opportunities to encourage their child to use their affected UE outside conventional therapy settings.[27 41 42]

Objectives

The overall objective of this project is to assess the utility of a single joystick-operated ride-on-toy navigation training (RNT) programme to promote affected UE function in young children with UCP. Ride-on-toys are extremely enjoyable for neurotypical children and children with disabilities alike and provide excellent opportunities to encourage children to use their UE skillfully to navigate through their physical environment. In our pilot work, we incorporated the single joystick-operated RNT (with the joystick provided on the child's affected side) into activities provided within an existing summer camp for children with UCP.[43] Research staff provided the RNT in our pilot study. Although we found promising evidence of feasibility and perceived efficacy of the RNT,[43] given the design of the pilot study, we were unable to isolate the effects of the RNT compared with the effects of other camp-related activities that children received. The present single-arm intervention study is designed to address this limitation and aims to assess the effects of the RNT as a therapeutic adjunct when provided within the child's home. Children will serve as their own controls and we will compare the effects of RNT provided in addition to conventional therapy/treatment-as-usual (intervention phase) to the effects of conventional therapy/treatment-as-usual provided alone (control phase) (see figure 1 for study timeline). Moreover, this community-embedded intervention will also involve caregivers as intervention providers. We will modify commercially available dual joystick-operated ride-on-toys to provide controls only on the child's affected UE. Our aims are: (1) to assess acceptance, feasibility of implementation, adherence, satisfaction, and perceived benefits of the home-based researcher and caregiver codelivered RNT through feedback from children and families and (2) to assess the preliminary efficacy of the of the home-based researcher and caregiver codelivered RNT programme in promoting manual abilities and spontaneous use of the affected UE in children with UCP. We hypothesise that feedback from
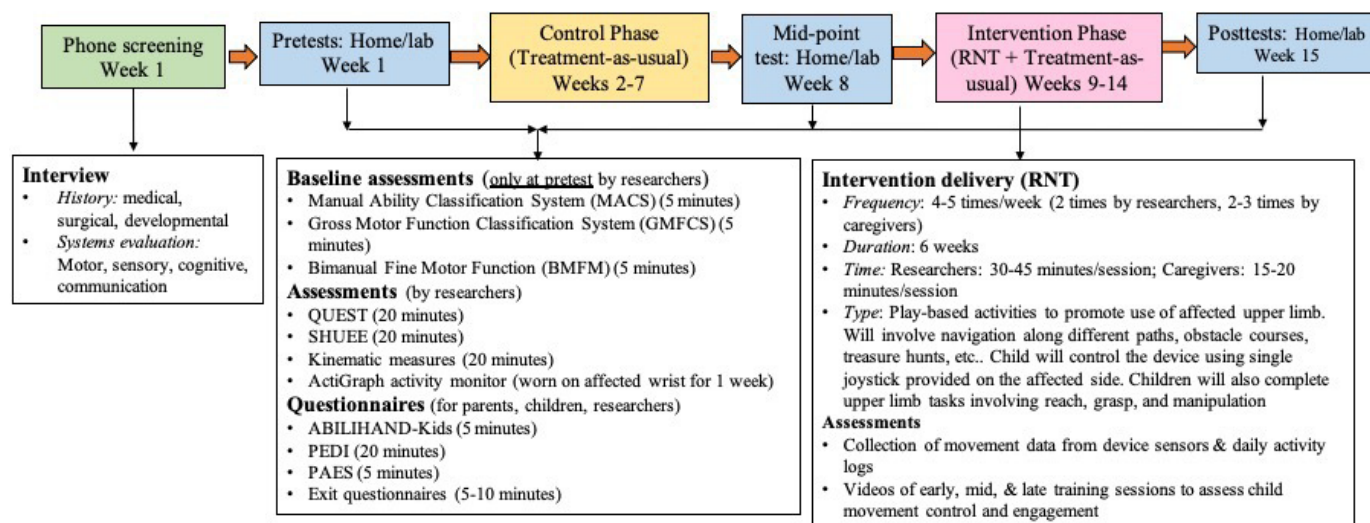
Figure 1 Study timeline and overview of procedures. PAES, Physical Activity Enjoyment Scale; PEDI, Paediatric Evaluation of Disability Inventory; QUEST, Quality of Upper Extremity Skills Test; RNT, ride-on-toy navigation training; SHUEE, Shriner Hospital Upper Extremity Evaluation.

children and families will indicate that the training is acceptable, feasible to implement, and associated with high rates of adherence, child enjoyment, family satisfaction, and perceived benefits in terms of affected UE use and function. We also hypothesise greater improvements in unimanual and bimanual UE function measured on standardised and quantitative assessments during the intervention phase compared with the control phase.

METHODS AND ANALYSIS
Public/patient involvement statement
The current study design and content is based on pilot data collected from 11 children with UCP (mean (SD): 6.54 (2.76); range 3–14 years) at a summer camp (please note that data from our previous pilot study[43] will not be included in the present trial). Children, caregivers, and clinicians provided feedback on the training activities as well as reported on acceptance and feasibility of implementation of the RNT programme. One of the members of the research team has CP and has been closely involved in the development of the current study design and methodology.

Study design
We will conduct a non-randomised single-arm interventional study where children will serve as their own controls. All children will be observed over a 6-week control phase during which they will continue to receive treatment-as-usual through their school/private therapies, and we will keep track of conventional therapies received (frequency, duration, and type) through weekly parent interviews and check-ins. After the control phase, children will receive the 6-week intervention phase. During this phase, in addition to ongoing conventional therapy, children will receive the researcher-caregiver

codelivered RNT programme (see figure 1 for study timeline). This study is planned to be conducted from January 2023 to December 2023.

Participants and recruitment
Fifteen children with UCP between 3 and 8 years will be recruited. Children will be included if they demonstrate clear asymmetry in UE strength/control and are able to use their affected UE or trunk to activate a joystick placed within reaching distance from their body. Participants' gross and fine motor skills will be assessed at baseline using the Gross Motor Function Classification System, Bimanual Fine Motor Function, and the Manual Ability Classification System.[44–46] Children will be excluded if they have a recent history (within last 6 months) of UE trauma/surgery, are unable to sustain supported sitting for 20 min, have blindness/visual impairment, have fixed musculoskeletal deformities of the affected hand/wrist, are unable to follow two-step verbal instructions, or if their weight exceeds the limits of the toy.

Participants will be recruited through fliers circulated across local schools, parent advocacy groups, CP-specific organisations, area hospitals, and through professional networks of physical therapists and special educators. Once an interested family contacts us, we will conduct an initial screening with the family via phone/Webex. During this screening, caregivers will be asked questions related to our eligibility criteria. Families who fit our eligibility criteria will be asked to provide medical records confirming their child's diagnosis and will fill out an intake form that will obtain information on child demographics, health history, and preferred motivators. Thereafter, the family will be scheduled for their first testing visit. Parental permission and child written/oral assent will be obtained during the first testing session. We will confirm that children fit our eligibility criteria during

the in-person pretest session through play-based observational assessment and standardised motor testing.

Sample size calculation

Since this is the first study to assess the effects of an RNT programme on UE function in children with UCP, we do not have effect size estimates for all the primary outcome measures. Our sample size estimates are based on pilot data we collected as well as published literature on the effects of innovative games (such as virtual reality and video games) and home-based therapy programmes for children with UCP. Effect sizes from published literature varied from 0.83 to 1.16 in favour of the intervention group.[25 47 48] Our own pilot data suggest large effect sizes (≥ 0.8 for 2 of the outcome measures, ie, total scores on the Quality of Upper Extremity Skills Test (QUEST) and scores on the Shriner's Upper Extremity Evaluation (SHUEE)). Considering these multiple sources, we determined sample size estimates assuming fixed-effects analysis of variance on a 1×3 design, 1 group and 3 time points (pretest, midpoint, post-test). The study will include a total of 15 cases. The criterion for significance (alpha) has been set at 0.05 (two tailed). With the effect size estimate of 0.80, the study has power of 82% in the most conservative scenario.

Toy modification and retrofitting

The commercially available ride-on-toys come with two joysticks for navigation and are designed for healthy kids. Our team will modify the toys to a single-joystick operation and we will encourage the child to only use their affected side to navigate the toy in the forward, backward, left, and right directions. In addition, protective support frames and reinforcements will be added to provide extra postural support, comfort, and safety. A microcontroller embedded in the toy will be programmed to calculate device usage parameters that is, total use time/session and joystick average push/pull time.

Study procedures

Control phase: treatment-as-usual (6 weeks)

Following the pretest session, children will be followed for 6 weeks during which they will continue to receive conventional therapies as part of their 'treatment-as-usual' through school-based programming or outpatient services. We will keep track of conventional therapies that children receive during this phase using weekly therapy logs. Caregivers will document the type (physical therapy, occupational therapy, speech therapy, etc), frequency (number of times per week), duration per session, and location of therapy sessions (clinics, school, home, etc) within the therapy logs. Following the 6-week control phase, a midpoint assessment will be conducted. Thereafter, children will receive the 6 week intervention phase (see figure 1 for timeline).

Intervention phase: RNT (6 weeks)

During the intervention phase, in addition to treatment-as-usual, children will also receive RNT every week.

During the RNT training sessions, the child's unaffected arm will be constrained using a soft mitten to encourage use of the affected arm to move the joystick to navigate through their naturalistic environment. We will work with families to identify a space for the training based on family convenience, for instance, at home (indoors or outdoors for example, in the garage, basement, or driveway), on a playground, at school, or other community spaces. The ride-on-toy is robust and can be used across a variety of different surfaces. The study procedures will abide by safety specifications (regarding height, weight, and age) of the ride-on-toy manufacturer. Children will wear a helmet and fasten their seatbelt while driving the toy.

During the intervention phase, the ride-on-toy will be left at the child's home. Training will be provided for 30–45 min/day, 2 days/week under the researchers' supervision and we will ask caregivers to encourage their child to drive the ride-on-toy for 2–3 more days every week for around 15–20 min/session. Please note that we will provide caregivers with training ideas and materials needed for their sessions on a weekly basis. Caregivers will also be provided a safety manual and written training instructions for ride-on-toy operation at the start of the study. In addition, researchers will check-in and trouble shoot any problems in ride-on-toy operation on a weekly basis, and caregivers will be provided contact details of the research team in case of any emergencies. The training volume for the study was determined based on pilot data from an RNT programme incorporated within a summer camp for children with CP.[43] Both researchers and caregivers will maintain a training log to document details of their weekly sessions including duration, content, as well as perceived level of difficulty and child enjoyment.

Researcher-delivered sessions will be provided by trained research staff. Each session will involve periods of 'structured play' and 'free play'. The structured play will involve navigational challenges and UE tasks. The navigational challenges will involve games such as shape mazes, treasure hunts, relay races and obstacle courses that require gross motor UE movements (shoulder, elbow and wrist movements to grasp and move the joystick in different directions—forward, backward, left and right). The UE tasks that will be completed at intermediate stations during navigation will involve components of reaching, grasping, in-hand manipulation, and release (eg, pick up/pull/push/throw props, as well as lift and manipulate balls, bean bags, cups, blocks, cones, and pool noodles). Training activities will be progressed weekly to increase the navigational challenge (straight path, circular path, slalom path, mazes, obstacle avoidance tasks, and timed challenges) and the movement control challenge (increase in required range of UE movements, muscle force control, and precision/manual dexterity during tasks). During free play, the child will be encouraged to drive the toy to explore their physical environment at their will. All researcher-delivered sessions will be videotaped. Caregiver-delivered sessions will involve similar activities. Caregivers will be trained by researchers

and will receive a safety manual, easy-to-follow written and pictorial instructions, weekly training ideas/materials, and friendly text-email-based reminders to facilitate compliance and reduce caregiver burden. We aim to provide an overall dose of 9–13 hours of RNT (researcher-delivered training for 6–9 hours over 6 weeks @ 30–45 min/session, 2 days/week; caregiver-delivered training for 3–4 hours over 6 weeks @ 15–20 min/session, 2 days/week).

The RNT will be based on principles of motor learning and family-centric practice including encouraging variable and repetitive practice, providing progressive challenges tailored to child needs, using activities promoting problem-solving and trial-and-error learning, providing immediate visual and auditory feedback based on performance, fostering playful exploration, and incorporation of child-preferred toys/themes into games to increase motivation and involvement of the family during training. Trainers will follow a least-to-most prompting hierarchy involving gestural, verbal, and finally hand-on-hand assistance per the child's needs during training. At the end of every session, children will be provided stickers as reinforcement, and children will receive a ride-on-toy driver's license following completion of the programme.

Training of intervention providers and assessing fidelity of implementation

All intervention providers will be physical therapy, kinesiology, allied health, or neuroscience graduate and undergraduate students who will receive training from the study principal investigator (PI). The training will involve watching videos of intervention sessions from our pilot study, one-on-one meetings with the last author to discuss key ingredients of training sessions, review of standardised training manuals, and weekly review/feedback sessions with the last author during the training. To assess fidelity of training, an unbiased coder will randomly select and code video data from early, mid, and late researcher-delivered training sessions using custom-designed fidelity checklists. Trainers and caregivers will maintain weekly training logs to assess child adherence/compliance with the training. Trainers and caregivers will also fill out post-test exit questionnaires to assess ease of implementation of RNT as a home programme.

Testing protocol

Aim 1: Assessment of UE function and use

Assessments will be conducted during the pretest (before control phase), at midpoint (after control phase), and during the post-test (after intervention phase) (see figure 1). The testing visits will be conducted at our lab or at the child's home, based on the family's convenience. The testing visits (pretest, midpoint, and post-test) will be conducted within 2 weeks prior to or following completion of the control and interventions phases, respectively. All tests will be videotaped and conducted by the same assessor. The assessments will capture changes in children's unimanual and bimanual function and spontaneous hand use through (A) standardised tests and parent report questionnaires and (B) objective measures including wrist-worn accelerometers and movement kinematics during a reach-grasp task (see details in table 1).

Primary measures

1. QUEST[49]: The QUEST is a 36-item, criterion-referenced, valid (concurrent validity 0.84), and reliable (test–retest reliability is 0.75–0.95) test to assess quality of unimanual function.[50] The Minimal Clinically Important Difference (MCID) for this test has been reported as 4.89 score units.[51] The test can be used for children between 18 months and 8 years and assesses UE function in four domains: dissociated movement, grasp, protective extension, and weight-bearing. We will report total and domain-specific standardised scores of the QUEST.

2. Wrist-worn accelerometry: To assess children's habitual dominant and non-dominant arm use, children will be asked to wear ActiGraph activity monitors (wGT3X-BT) on the wrist for a duration of 1 week (minimum of 4 days, ie, 3 weekdays and 1 weekend day) at all testing time points. In addition, children will also wear ActiGraph watches during training sessions within the first and last weeks to assess changes in affected UE activity during the RNT programme. Caregivers will also be provided with an activity log to keep track of the child's ActiGraph wear time and sleep times over the week. We will calculate metrics of use ratio, magnitude ratio, and bilateral magnitude using the accelerometer data.[52] These ratios provide information regarding the duration (in hours) and magnitude (based on activity counts) of use of the dominant and non-dominant arms. In addition, we will also calculate the percent time spent by the affected arm in no activity, light activity, and moderate-to-vigorous activity bouts. The MCID score for arm accelerometry is a change of 575–752 counts.[53]

Secondary measures

1. Shriner Hospital Upper Extremity Evaluation (SHUEE)[54]: The SHUEE is a video-based test for children between 3 and 18 years that assesses spontaneous use of the affected arm during 16 bimanual functional tasks. The test has an excellent intrarater (r=0.98–0.99) and inter-rater reliability (r=0.89–0.90).[54] It also shows fair-to-good correlations with the self-care domain of the Paediatric Evaluation of Disability Inventory and the total time section of the Jebsen-Taylor Test of Hand Function.[54]

2. Movement kinematics: We will collect kinematic data during a reach-grasp task involving unilateral and bilateral reaching at self-selected speed towards different objects (foam ball, rattle and square block) placed at half arm's length (near) and at arm's length (far) from the child on the table. Motion sensors will be placed on both hands, both forearms, both arms, and the C7 spinous process. We will assess bilateral range of motion, maximum joint angles, peak and average veloc-

Table 1 Summary of assessments and outcome measures to be used in the study

Test	Domain assessed	Outcome measures	Time points of measurement	Test duration (in minutes)
Assessing upper extremity function and use				
Primary				
Quality of Upper Extremity Skills Test	Unimanual function assessed on four domains: 1. Dissociated movements 2. Grasps 3. Protective extension 4. Weight-bearing	1. Total standardised scores 2. Domain-specific standardised scores	▶ Pretest ▶ Midpoint ▶ Post-test	15–20
Wrist-worn accelerometry	Use (duration and intensity) of dominant and non-dominant arms during habitual activities of daily living and while driving the ride-on-toy.	Activity counts obtained will be used to calculate: 1. Use ratio 2. Magnitude ratio 3. Bilateral magnitude 4. Per cent time spent in no activity, light activity and moderate-to-vigorous activity bouts	▶ Pretest ▶ Midpoint ▶ Post-test ▶ First and last weeks during researcher-delivered sessions	All day for 1 week (at least 3 weekdays, 1 weekend day)
Secondary				
Shriner Hospital Upper Extremity Evaluation	Spontaneous use of the affected arm during bimanual activities	1. Spontaneous functional analysis scores 2. Dynamic positional analysis scores for UE joints.	▶ Pretest ▶ Midpoint ▶ Post-test	15–20
Movement kinematics	Reaching trajectories during a unilateral and bilateral reach-grasp task (motion sensors attached on both hands, both forearms, both arms and C7 spinous process)	1. Maximum joint angles 2. Peak and average velocity 3. Total movement time 4. No of movement units 5. Movement straightness, smoothness, variability and speed 6. Symmetry of reaching trajectories	▶ Pretest ▶ Midpoint ▶ Post-test ▶ Once/week during researcher-delivered sessions	15–20
ABILHAND-Kids questionnaire	Parent perceptions of child's use of affected UE during bimanual activities of daily living	1.Total scores	▶ Pretest ▶ Midpoint ▶ Post-test	5–7
Training specific measures of manoeuvring skills: movement bouts	Ability to sustain pushing/pulling of the joystick during navigation	1. Average length of movement bouts during navigation 2. Rates of unintentional stops/session due to fatigue or poor muscle control	Intervention phase: ▶ Early (week 1) ▶ Mid (week 3) ▶ Late (week 6)	30–45
Training specific measures of manoeuvring skills: obstacle contacts	Ability to stay on the designated course while avoiding obstacles along the path	1. Rates of bumps against obstacles 2. Percent time spent 'in-designated' vs 'out-of designated' path.	Intervention phase: ▶ Early (week 1) ▶ Mid (week 3) ▶ Late (week 6)	30–45
Training specific measures of manoeuvring skills: joystick assistance	Ability to independently control joystick movements during navigation	1. Percent time of independent vs trainer-assisted ride-on-toy navigation	Intervention phase: ▶ Early (week 1) ▶ Mid (week 3) ▶ Late (week 6)	30–45
Assessing feasibility, adherence, and training acceptance				
Primary				

Continued

Table 1 Continued

Test	Domain assessed	Outcome measures	Time points of measurement	Test duration (in minutes)
Device use time	Time spent driving the ride-on-toy on a weekly basis assessed through sensors mounted on the ride-on-toy	1. Device use time/week (in hours)	Weekly throughout the intervention phase	–
Secondary				
Child and caregiver exit questionnaires	Custom-designed questionnaires to assess feasibility and satisfaction with training	1. Satisfaction 2. Enjoyment and repeatability 3. Perceived efficacy in promoting motor skills (gross motor, fine motor, and bilateral coordination) and spontaneous affected UE use. 4. Barriers to implementation 5. Caregiver burden during implementation	Post-test	5–10
Training logs	Adherence with training protocol	1. Average training duration/session	Weekly throughout the intervention phase	2–3
Child affect and attention	Child engagement	1. Affective states: Percent duration of time spent in- 　1. Positive affect 　2. Negative affect 2. Attentional focus: Percent duration of time spent in- 　1. Task-appropriate attention 　2. Task-inappropriate attention	Intervention phase: ► Early (week 1) ► Mid (week 3) ► Late (week 6)	30–45
Physical Activity Enjoyment Scale	Reported enjoyment while completing conventional therapy and following RNT programme	1. Total scores	► Midpoint ► Post-test	5–10

RNT, ride-on-toy navigation training; UE, upper extremity.

ity, total movement time, number of movement units, movement straightness, movement variability, and smoothness (eg, jerk, dimensionless jerk, and log dimensionless jerk).[55 56] Kinematic and motion data will also be obtained during weekly researcher-delivered training sessions.

3. ABILHAND-Kids questionnaire[57]: This is a valid and reliable questionnaire assessing parent perceptions of their child's level of ease or difficulty in performing 21 manual activities independently over the last 3 months. It has strong reliability (r=0.94) and reproducibility (r=0.91), and the test–retest reliability is 0.91 after a delay of 25±13 days.[57]

4. Training-specific measures of movement control during navigation: Video data from researcher-delivered sessions will be coded using Datavyu behavioural coding software to assess changes in movement control and children's manoeuvring skills across training weeks. Specifically, an early (week 1), mid (week 3), and late (week 6) session will be coded for (A) duration of independent versus trainer-assisted ride-on-toy navigation,

(B) average length of movement bouts during navigation, (C) rates of stops/session due to fatigue or poor muscle control, (D) rates of unintentional bumps against obstacles, and (E) accuracy of navigation, that is, duration of time spent inside versus outside the designated path (see table 1 for details).

Aim 2: Feasibility and acceptance of intervention

We will assess feasibility of the RNT programme using a framework developed by Bowen et al., to evaluate pilot feasibility trials.[43 58] Specifically, we will assess acceptability, satisfaction/demand, practicality of implementation, child enjoyment, and perceived efficacy of the RNT programme.

Primary measures

1. Device use time: The ride-on-toy will be fitted with a microcontroller that will store a log of the daily use time of the ride-on-toy. This data will be downloaded from the device on a weekly basis during researcher-delivered sessions.

Secondary measures

1. Exit questionnaires: At post-test, children and caregivers will fill out custom-designed exit questionnaires that will obtain information on satisfaction with the training, child enjoyment, repeatability of training, perceived efficacy of the programme, barriers to implementation, and caregiver burden during the programme.[43] Child questionnaires will involve Likert-style questions rated on a 5-point scale. Caregiver questionnaires will involve a mix of Likert-style and open-ended questions.

2. Physical Activity Enjoyment Scale[59 60]: This is a 16-item questionnaire scored on a 5-point Likert scale that has been validated for children between 6 and 18 years to assess enjoyment with physical activity. We will obtain child perceptions both at the end of the control phase and the intervention phase of the study. For the post-test administration, we will modify the basic stem of the questionnaire from 'When I am physically active' to 'When I drive the ride-on-toy'. If children are unable to answer the questions in an interview format, we will seek parent input to complete the questionnaire.

3. Training-specific measures of child engagement: Video data from an early (week 1), mid (week 3), and late (week 6) session will be coded using Datavyu to assess children's affective states and their attentional focus during ride-on-toy navigation sessions. Specifically, we will report on (A) percent duration of positive affect, (B) rates of smiles, and (C) duration of task-appropriate (looking at path, obstacles, and props used in the training) versus task-inappropriate attention (distracted or looking elsewhere) during sessions.

4. Daily training logs: Researchers and caregivers will be asked to document the duration of practice (in minutes) the child received per session during researcher-delivered and caregiver-delivered sessions within the intervention phase. These data will supplement the device use data obtained from the toy microcontroller.

Data coding procedures

All data from standardised tests will be coded by graduate and undergraduate students trained by the PI and will establish inter-rater and intrarater reliability of over 90% using 20% of the dataset. All training-specific measures will be coded by trained undergraduate students following the establishment of reliability as stated above. Although coders will not be blind to child grouping, for each measure, we will have two coders independently score the data and we will resolve discrepancies between raters through consensus coding in an ongoing manner during the data coding process. All kinematic and activity data will be post-processed using custom-developed MATLAB/Labview programs.

Statistical analysis

Data analyses will be conducted in consultation with a biostatistician. All data will be evaluated for assumptions of parametric statistics and general or generalised models will be selected accordingly. For dependent variables collected at pretest, at midpoint, and at post-test, general or generalised linear models for change scores will be conducted controlling for baseline characteristics and possible covariates (eg, age, volume of therapies received). We will compare training-related effect sizes across control and intervention phases to assess the differential effects of the ride-on-toy training on motor function in children with UCP. Variables that violate distributional assumptions of normality will be analysed using generalised linear model procedures, which enable the use of non-Gaussian error models. We anticipate limited missing data based on pilot data collection for this study and also because attrition rates are typically low in home-based versus clinic-based programmes.[61 62] When applicable, missing data will be evaluated to determine if it is completely at random. If random, multiple imputation by maximum likelihood estimation will be used.

Data management

Participants will be assigned a unique four-digit identifier at the time of enrolment. Hard copies of identifiable study files will be retained in locked cabinets in the PI's office for a duration of 5 years after study completion, following which they will be shredded. Data collected from participants will be stored in a deidentified form in locked file cabinets and on password-protected computers indefinitely. Only research staff working on the project will have access to these data. For participants who fail the screening, all hard copies will be shredded, and electronic information will be permanently deleted, immediately after the screening.

Expected outcomes

We hypothesise that the RNT programme will be acceptable to the family, feasible to implement, and associated with high rates of treatment adherence, fidelity of implementation, satisfaction, and perceived benefit as assessed using child-rated and caregiver-rated exit questionnaires. Child engagement will be high throughout the intervention phase. We also predict greater improvements (larger effect sizes) in unimanual and bimanual function and spontaneous affected UE use following the intervention compared with the control phase as assessed using qualitative (standardised tests and parent-report questionnaires) and quantitative measures (kinematic measures and habitual arm use based on wrist-worn accelerometry).

Resource sharing plan

We plan to disseminate the findings from this research through presentations at national and international scientific conferences as well as through peer-reviewed manuscripts published in leading journals in this area. All authors (PI/coinvestigators) will have access to the data and will receive priority for first authorship for the papers they take lead in writing.

ETHICS AND DISSEMINATION

The study was approved by the Institutional Review Board of the University of Connecticut (# H22-0059) and is registered at ClinicalTrials.gov (Trial registration number: NCT05559320). Study procedures will be governed by the UConn IRB. All protocol modifications will be reported to the IRB and ClinicalTrials.gov. Participants will be discontinued in case of an adverse event and adverse events will be immediately reported to the UConn IRB. Results from this study will be disseminated through peer-reviewed manuscripts in scientific journals in the field, through national and international conferences, and presentations to parent advocacy groups and CP-specific support organisations.

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Contributors SMS is the study principal investigator; PK and KM are coinvestigators; all three of them conceptualised the study and design and will be responsible for conducting the study and reporting outcomes collected. KMF is a consultant on the study and is providing advice on intervention development, study design, and key outcomes. VS was involved in analysis of pilot data and will be involved in data collection and analyses for the present study. First draft of the manuscript was written by VS and SMS. All authors contributed to the critical review of the manuscript and approved the final draft.

Disclaimer The sponsors had no role in the study design and will not be involved in its execution, analyses, interpretation of the data, or decision to submit results.

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.

Patient consent for publication Consent obtained from parent(s)/guardian(s).

Provenance and peer review Not commissioned; externally peer reviewed.

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