[Original article: 3 tables, 3 online tables, 2 online figures]

Functional electrical stimulation cycling, goal-directed training, and adapted cycling for children with cerebral palsy: a randomized controlled trial

ELLEN L ARMSTRONG^{1,2,3}

ROSLYN N BOYD³

SEAN A HORAN^{1,2}

MEGAN J KENTISH^{3,4}

ROBERT S WARE²

CHRISTOPHER P CARTY^{1,2,5}

1 School of Allied Health Sciences, Griffith University, Gold Coast, QLD; **2** Menzies Health Institute Queensland, Griffith University, Gold Coast, QLD; **3** The Queensland Cerebral Palsy Rehabilitation Research Centre, Centre for Children's Health Research, Faculty of Medicine, The University of Queensland, South Brisbane, QLD; **4** Queensland Paediatric Rehabilitation Service, Children's Health Queensland Hospital and Health Service, Brisbane, QLD; **5** Gold Coast Orthopaedic Research Engineering and Education Alliance, Griffith University, Gold Coast, QLD, Australia.

Correspondence to Ellen L Armstrong, Level 6, Centre for Children's Health Research, 62 Graham Street, South Brisbane, QLD 4101, Australia. E-mail: ellen.armstrong@griffithuni.edu.au

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ABBREVIATIONS

COPM	Canadian Occupational Performance Measure
FES	Functional electrical stimulation
FTSTS	Five Times Sit-to-Stand test
HEP	Home exercise programme
PEDI-CAT	Pediatric Evaluation of Disability Inventory Computer Adaptive Test
RCT	Randomized controlled trial

[Abstract]

AIM To test the efficacy of functional electrical stimulation (FES) cycling, goal-directed training, and adapted cycling, compared with usual care, to improve function in children with cerebral palsy (CP).

METHOD The intervention was delivered between 2017 and 2019 and included three sessions per week for 8 weeks (2×1h sessions at a children's hospital, and 1h home programme/week). Hospital sessions included 30 minutes of FES cycling and 30 minutes of goal-directed training. Home programmes included goal-directed training and adapted cycling. The comparison group continued usual care. Primary outcomes were gross motor function assessed by the Gross Motor Function Measure (GMFM) and goal performance/satisfaction assessed using the Canadian Occupational Performance Measure (COPM). Secondary outcomes were sit-to-stand and activity capacity, participation in home, school, and community activities, and power output. Linear regression was used to determine the between-group mean difference immediately post-training completion after adjusting for baseline scores.

RESULTS This randomized controlled trial included 21 participants (mean age=10y 3mo, standard deviation [SD]=3y; Gross Motor Function Classification System level: II=7, III=6, IV=8) who were randomized to the intervention ($n=11$) or usual care group ($n=10$). Between-group differences at T2 favoured the intervention group for GMFM-88 (mean difference=7.4; 95% confidence interval [CI]: 2.3–12.6; $p=0.007$), GMFM-66 (mean difference=5.9; 95% CI: 3.1–8.8; $p<0.001$), COPM performance (mean difference=4.4; 95% CI: 3.9–5.3; $p<0.001$) and satisfaction (mean difference=5.2; 95% CI: 4.0–6.4; $p<0.001$).

INTERPRETATION Children with CP achieved meaningful functional improvements after FES cycling, goal-directed training, and adapted cycling training. Cycling programmes for children with CP should be individualized and goal directed.

What this paper adds:

- Functional electrical stimulation cycling incorporated into goal-directed training and adapted cycling programmes builds functional capacity.
- Non-ambulant children with cerebral palsy can participate in cycling with appropriate equipment adaptations.

[main text]

Stationary and dynamic forms of cycling are frequently included in training programmes for people with cerebral palsy (CP) because they engage large muscle groups and can be modified to suit the individual's level of function.^{1,2} Children with CP with spasticity and reduced muscle strength can have challenges learning to pedal independently and reach therapeutic exercise intensities.³⁻⁵ Motor-assisted functional electrical stimulation (FES) cycling can assist people with impaired motor function to achieve full crank rotations and higher power outputs during cycling.^{6,7} Electrical stimulation at higher intensities has the added benefit of increased muscle fibre recruitment, resulting in stronger contractions and higher cycling power outputs.⁷⁻⁹ Even at sub-motor threshold intensities, alternating patterns of muscle stimulation may provide sensory cues to promote more fluid pedalling movements.³ Nonetheless, research on the application of FES cycling to improve cycling ability in children with CP is limited.

FES cycling is a safe and feasible form of exercise that can be tolerated by people with central neurological conditions who are sensate, including adolescents with CP.⁹⁻¹¹ Previous FES cycling case series in people with CP have reported immediate improvements in cycling performance when FES was applied (for example increased cadence and power output), as well as longer term improvements in lower limb muscle strength, oxygen expenditure, and reduced muscle cocontractions.^{7,9,12,13} These results suggest that FES cycling may be a useful inclusion to therapy programmes for people with mobility restrictions, as it may allow individuals to train at higher intensities and subsequently increase their exercise capacity. To date, no randomized controlled trial (RCT) has tested the efficacy of FES cycling combined with other forms of exercise to target improved functional outcomes in children with CP.

Community-based cycling using an adapted bike is an enjoyable and inclusive mode of exercise for children with CP; however, these bikes are expensive and often require technical expertise for initial set up.² Furthermore, in contrast to stationary cycling, adapted cycling requires the child to overcome ground resistance, navigate obstacles, and master technical skills such as steering and braking. The transition from stationary cycling to adaptive cycling can be significant, especially for children with CP classified in Gross Motor Function Classification System (GMFCS) levels III and IV, and for this reason additional goal-directed training might be indicated. No previous studies have investigated the benefits of a combined programme of clinic-based FES stationary cycling, goal-directed training, and community-based adapted cycling.

The purpose of this RCT was to determine the efficacy of an 8-week programme of FES cycling, goal-directed training, and adapted cycling training to improve activity capacity and functional outcomes for ambulant and non-ambulant children with CP, classified in GMFCS levels II to IV. The intervention was designed to address the paucity of evidence-based programmes that target function, activity capacity, and physical activity levels in children with CP, particularly those who are not independently ambulant. It was hypothesized that the intervention would result in significantly higher gross motor function, goal performance and satisfaction, activity capacity, and participation compared to usual care.

METHOD

This parallel-group RCT investigated the effectiveness of a training package of FES cycling, goal-directed training, and adapted cycling compared to usual care in children and adolescents with CP in Brisbane Australia between December 2017 and August 2019. Ethical clearance was obtained from the Children's Health Queensland Human Research Ethics Committee and the Griffith University Human Research Ethics Committee (GU2018/037; HREC/17/QRCH/88; SSA/17/QRCH/145). Written consent was gained from parents and children over 12 years old, and verbal assent from younger children. The study was registered with the Australian New Zealand Clinical Trials Registry

(ACTRN12617000644369p) and any changes to the methods after trial commencement were reported. A published study protocol is available.¹⁴

Participants

Children with CP classified in GMFCS levels II to IV, aged 6 to 18 years, and living within 100km from the training facility were invited to participate. Additional inclusion criteria were: goals to improve functional mobility and cycling; able to tolerate 45 minutes of physiotherapy and communicate discomfort; and able to attend training and testing sessions. Participants were excluded if they: were unable to tolerate any level of FES stimulation at baseline; had joint contractures, severely reduced range of movement, or hip displacement that would prevent participation in cycling; surgery, trauma, or fractures in the preceding 12 months or surgery/serial casting scheduled during the study period; had any contraindications for FES; known cardiovascular or pulmonary conditions; and uncontrolled epilepsy.

Study procedure

After baseline assessments, participants were randomly allocated to the 8-week intervention or usual care. Participants were stratified by GMFCS level and randomized by an independent randomization service using computer generated, randomly varied block sizes that ranged from 2 to 4. Because of the nature of the cycling intervention, blinding of the participants or therapist was not possible. The assessor who scored the Gross Motor Function Measure (GMFM) via video recordings was masked to group allocation. The primary analysis was between-groups immediately postintervention.

Intervention group

The intervention consisted of two 1-hour sessions per week at a tertiary children's hospital and a 1-hour home exercise programme (HEP) per week for a period of 8 weeks (total dose 24h). Training sessions were led by a physiotherapist and included a 30-minute goal-directed training programme informed by Canadian Occupational Performance Measure (COPM) goals, and up to 30 minutes of FES cycling. Weekly HEPs were supervised by a parent or guardian. Both groups of participants continued any usual physiotherapy or occupational therapy sessions throughout the study, which were recorded in a usual care diary. A full description of the intervention, including goal-directed training and FES cycling parameters, is reported in the protocol.¹⁴

Individualized training programmes included at least two functional exercises aimed at improving lower limb strength (e.g. loaded sit-to-stand, step-up, supine bridges) and two activities that were tailored to the participant's targeted COPM goals (e.g. transfer practice). Weighted vests were used to progress sit-to-stand and step-up exercises if the participant could complete 8 to 12 unloaded repetitions. No weight was added in the first week of training. In subsequent weeks, weight was added in increments of 0.5kg to 1kg (Table S1, online supporting information).

FES cycling sessions were completed on a stationary, motorized FES bike (RT300-SL cycle, Restorative Therapies Inc., Baltimore, MD, USA).¹⁴ Adhesive surface electrodes were placed bilaterally on participant's quadricep, hamstring, gluteal, gastrocnemius, and tibialis anterior muscles. Stimulation frequency was 40Hz to 50Hz, pulse width 200µs to 250µs, and amplitude was adjusted based on tolerance.¹⁴ Participants were encouraged to cycle at a steady pace for the first and last third of each 30-minute session static FES cycling session, aiming to maintain 50% to 60% of their maximum power output. In the middle third, participants completed 10 to 30 second cycling sprints, aiming for 80% to 100% of their maximum power output. A cycling test was conducted at baseline and at week 4 to determine maximum power output. In-between cycling tests, targets were adjusted based on the highest

power output achieved during preceding sessions. Intensity was monitored using a heart rate wrist monitor.¹⁴

Weekly HEPs consisted of 30 minutes of adapted cycling and a goal-directed training programme. Illustrated HEP handouts were updated fortnightly and exercises were taught during hospital sessions.

Usual care group

The usual care group continued any usual physiotherapy, occupational, and speech therapy. The type and content of therapy was recorded. At the end of the 8-week study period participants allocated to the usual care group were offered the intervention programme.

Outcome measures

This study reports data collected at baseline (T1) and immediately after the intervention at 8-weeks (T2). Primary outcomes were: (1) gross motor function, measured on the GMFM-88 and GMFM-66 and (2) goal performance and satisfaction measured using the COPM, where participants set three activity or participation goals related to the mobility or leisure domains. The minimal clinically important difference ranges from 2.05 to 4.8 logits on the GMFM-66; 5.05 to 7.1 logits on the GMFM-88; and 4.5 to 8.05 logits for the GMFM goal score total. A change score above 2 points on the COPM is considered clinically meaningful.¹⁵ Secondary outcomes included sit-to-stand capacity, assessed using the Five Times Sit-to-Stand test (FTSTS); participation in life activities as assessed by the Participation and Environment Measure – Children and Youth; children's capacity in the areas of daily activities, mobility, social cognitive, and responsibility, measured by the Pediatric Evaluation of Disability Inventory Computer Adaptive Test (PEDI-CAT); and peak cycling power output, assessed using a cycling sprint test.¹⁴

Statistical methods

Sample size was calculated using the statistical package G*Power3 (Universitat Kiel, Germany).¹⁶ An expected change score of 3 logits (standard deviation [SD]=3) was estimated based on studies that reported minimal clinically important differences for the GMFM^{17–19} and studies that investigated stationary cycling,²⁰ goal-directed,²¹ and intensive therapy²² interventions in children with CP. With 80% power and a two-sided test with alpha set at 0.05 (no adjustment for two primary outcomes), a sample of 34 participants (17 per group) was required to detect a between-group difference of 3 points or greater.

Data are summarized as mean (SD) for continuous data and frequency (percentage) for categorical data. Continuous data were analysed using a linear regression model with treatment group included as the main effect and baseline value of the outcome entered as a covariable. Outcome data was checked to ensure it fulfilled the conditions for linear regression. Effect estimates are presented as mean difference (95% confidence interval [CI]). Effect size, which provides an indication of the magnitude of the difference between groups, was calculated as the adjusted between-group difference at follow-up divided by the pooled standard deviation at baseline for the primary outcomes. FTSTS data did not meet linear regression assumptions and were analysed using quantile regression. Effect estimates are presented as median difference (95% CI). Analyses were conducted on an intention-to-treat basis, with participants analysed in the groups they were allocated to. Statistical significance was set at $p < 0.05$. Data analysis was completed using Stata I/C v14.2 (Statacorp, College Station, TX, USA).

RESULTS

Twenty-one children were recruited and randomized to the intervention ($n=11$) or usual care group ($n=10$), fewer than the target of 34 participants. Participant flow (Fig. S1, online supporting information) and characteristics (Table 1) are reported. At baseline, body mass index was significantly greater in the usual care group (mean difference=5.64, 95% CI 0.07–11.21; $p=0.05$), although this should be interpreted carefully because of the imprecision of the estimate. There were clinically meaningful differences in GMFM scores between groups at baseline, although the standard deviations were large and the differences were not statistically significant. Regression diagnostics were checked and confirmed to be appropriate for all variables except FTSTS.

Retention and compliance

Participant retention was 100%. Average attendance was 91% (160/176 sessions) with non-attendance due to illness ($n=9$), school camps ($n=2$), lack of support workers ($n=2$), other commitments ($n=2$), and inclement weather ($n=1$). On average, 70% of the gym session content was goal-directed training and 30% included resistance-based exercises. For FES cycling, the average time cycled per session was 24 minutes (SD=5.6) and average distance per session was 3.4km (SD=1.2).

Stimulation tolerance and number of muscles stimulated was variable. Quadriceps were stimulated in 89% of FES cycling sessions; hamstrings and tibialis anterior in 78%; gastrocnemius in 82%; and gluteals in only 16%. Participants tolerated a frequency of 40Hz to 50Hz and a pulse width of 200 μ s to 250 μ s. A median amplitude of 21mA (range=7–35) was tolerated for the quadriceps, 22mA (7–33) for the hamstrings, 18mA (5–33) for the tibialis anterior, 17mA (7–32) for the gastrocnemius, and 12mA (6–25) for the gluteal muscles.

Compliance with adapted cycling was high, with some exceeding the prescribed 30 minutes per week. Average time cycled at home was 50 minutes per week (SD=38.8). Details of the adapted bikes used for the HEP are provided (Table S2, online supporting information). For the exercise component of the HEP, participants completed 70% (SD=38.0) of the prescribed exercises. Reasons for missed HEPs were a lack of time or motivation and preference for cycling over structured exercise.

During the 8-week period, usual care consisted of a mean of 3.8 hours (SD=3.6) of therapy (Table S3, online supporting information).

Primary and secondary outcomes

The primary outcomes at baseline (T1) and immediately after the intervention (T2) are reported (Table 2). After adjusting for baseline, significant and clinically meaningful between-group differences were found favouring the intervention group on the GMFM-88 (mean difference=7.7, 95% CI 2.3–12.6; $p=0.007$; effect size=0.32; $R^2=0.95$), GMFM-66 (mean difference=5.9; 95% CI 3.1–8.8; $p<0.001$; effect size=0.51; $R^2=0.95$), GMFM goal scores (mean difference=13.5; 95% CI 5.9–21.0; $p=0.001$; effect size=0.85; $R^2=0.80$), COPM goal performance (mean difference=4.4; 95% CI 3.9–5.3; $p<0.001$; effect size=3.32; $R^2=0.83$) and COPM satisfaction (mean difference=5.2; 95% CI 4.0–6.4; $p<0.001$; effect size=3.89; $R^2=0.81$). The scatter plots with regression lines for the primary outcome measures are provided as Figure S2 (online supporting information).

Of the 63 COPM goals, 37 (59%) were activity focused and 26 (41%) were participation focused. The intervention group set a slightly higher proportion of activity (67%) versus participation (33%) goals compared to the control group. Thirty-one goals (49%) were related to cycling, 18 (29%) to mobility goals, and 14 (22%) to transfers.

Secondary outcomes are reported in Table 3. The intervention group had a significantly higher peak cycling resistance (mean difference=3.4; 95% CI 1.0–5.8; $p=0.009$), but not power output at T2. There were no between-group differences for PEDI-CAT or Participation and Environment Measure – Children and Youth. Three participants in the intervention group could not complete the FTSTS test at

baseline and two were unable to complete it at follow-up. These participants were given a score of 83 seconds, 1 SD slower than the slowest time recorded from successful trials.²³ After baseline adjustment there was not a significant between-group difference for the FTSTS at T2 (median difference: -5 seconds; 95% CI: -12.3-2.8; $p=0.20$).

Adverse events

Four minor adverse events were reported during the intervention, including post-training muscle soreness, general muscle fatigue, inner-thigh chaffing, and a low-speed fall while cycling at home. All events were mild, requiring no medical intervention.

DISCUSSION

This RCT demonstrated that a programme of FES cycling combined with goal-directed training and adapted cycling can lead to improvements in gross motor function, goal performance and satisfaction, and peak cycling resistance in children and adolescents with CP (GMFCS levels II–IV). Our results reinforce that children and adolescents with CP can experience meaningful functional improvements when provided with appropriate environmental modifications and opportunities to participate.

Previous stationary cycling studies in children with CP have reported mixed effects on gross motor function.²⁴ The PEDALS study, an RCT including 62 ambulant children (GMFCS levels I–II) with CP aged 7 to 18 years, reported no between-group difference for gross motor function after 30 hours of stationary cycling over 12 weeks.¹ Another RCT including 35 non-ambulant children (GMFCS levels IV–V) with CP found significant differences on the GMFM standing domain after 9 hours of stationary cycling over 6 weeks, less than half the dose in the PEDALS study.²⁵ In contrast, the current RCT included both ambulant and non-ambulant children (GMFCS levels II–IV) and the training dose was 60 minutes, three times per week for 8 weeks. The large treatment effects for the primary outcomes (effect size range 0.3–3.9) observed in the current study are likely due to some key distinctions in the intervention and study methodology that included FES cycling, goal-directed training, and adapted cycling.

A major strength of our intervention was the individualized intervention approach utilizing self-identified COPM goals. There is compelling evidence to support the efficacy of goal-directed approaches to improve activity and participation outcomes in children with CP.^{26–28} Children are more likely to engage in therapy programmes that align with their preferences and goals.²⁹ In the current study, children and parents were equally engaged in setting therapy goals and identifying perceived barriers and facilitators to their participation in the programme. A prerequisite for study participation was to have goals to ‘ride a bike’ or ‘improve function and mobility’. As such, participants had a pre-existing motivation or desire to improve. These factors likely contributed to participants’ high compliance, retention, and willingness to engage in the training programme.

A further point of difference from previous interventions was the addition of motor assistance and FES. A challenge when working with children with CP is finding activities that allow them to reach therapeutic exercise intensities. In the PEDALS study, participants who were unable to complete full pedalling revolutions were provided manual assistance by a therapist.¹ The ergometer used in the current study provided automatic motor assistance to those who were unable to pedal independently, allowing participants to contribute as much volitional effort as possible and have periods of active recovery when their pedalling cadence fell below a target speed. The addition of FES may have assisted participants to initiate pedalling or achieve more fluid pedalling movements, although participants’ tolerance of FES was highly variable. Higher amplitudes were tolerated in larger muscle groups (quadriceps and hamstrings) compared to smaller groups (tibialis anterior and gastrocnemius) and participants tolerated electrodes placed on the larger muscle groups. We recommend trialling a smaller pulse width and frequency, particularly in smaller muscle groups, to improve overall tolerance and comfort.

Participants' tolerance of the electrodes was also highly variable. Several children complained about the stickiness of the electrodes and many refused to trial FES on their gluteal muscles, potentially out of embarrassment/discomfort. Clinicians need to be aware of hypersensitivity issues when considering FES cycling for children with CP and may consider offering familiarization sessions with low levels of stimulation and teaching parents to apply gluteal electrodes. Regardless of the variable FES and electrode tolerance, this study found significant functional improvements in the intervention group, suggesting that benefits can be gained even from submotor levels of stimulation. Collectively, we re-emphasize the need for FES cycling programmes to be highly individualized.

A final important feature of the intervention was the use of adapted cycling as an adjunct to FES cycling training. Some children were hyper-compliant with the adapted cycling component of the intervention, with one participant completing eight times the prescribed dose in the first week of training. In this case, the participant's parents reported that she was so excited to receive a bike that she could actually ride, and was motivated to ride every day with her able-bodied sibling. This confirms that adapted cycling is more motivating than stationary cycling and provides opportunities to experience freedom of movement, keep up with peers, and improve their sense of wellbeing.^{2,30} Cycling is an enjoyable activity, and 'enjoyment' is a strong predictor of long-term engagement in physical activity.³¹ A qualitative evaluation may provide greater insights into the link between enjoyment and engagement with the intervention.

At T2, the between-group difference for cycling power output was not significant, although the intervention group experienced a within-group improvement of 14.6W compared to only 5.3W in the control group, and the intervention group achieved a significantly higher resistance. There are no previous studies to compare peak power output; however improvements in cycling resistance are consistent with a stationary cycling study in 10 non-ambulant children with CP, suggesting that stationary cycling may have positive effects on lower limb strength.²⁰ Given muscle strength was not directly tested, an alternative explanation is that a practice or learning effect occurred in the intervention group.²⁰ It is also possible that this test was confounded by a lack of understanding, behavioural issues, and low motivation to complete maximum sprint efforts in some cases in both groups of participants.

The intervention did not lead to improvements on the PEDI-CAT or Participation and Environment Measure – Children and Youth. A lack of change on the responsibility and social/cognitive domains of the PEDI-CAT was expected as the intervention specifically targeted physical functioning and mobility. A lack of change on the mobility or daily activity PEDI-CAT domains despite improvements in goal performance on the COPM may however reflect that the PEDI-CAT is a measure of what the child can or cannot do (capacity) rather than performance.³² The PEDI-CAT daily activity and mobility items cover a broad range of activities that are applicable to children with CP, but were not the focus of the intervention. It is likely that improvements in the performance of mobility and cycling goals did not translate to general improvements in the capacity to perform other mobility tasks or activities of daily living.

There are some potential limitations to consider when interpreting our results. The sample size was small, and the combined therapies approach limits the extent to which conclusions can be drawn on individual study components. Although the actual sample size was smaller than the intended sample size of 34 participants, 21 participants (11 in the intervention and 10 in the control group) was sufficient to detect between-group differences at follow-up of 7.4 and 5.9 points on the GMFM-88 and GMFM-66, after adjusting for baseline GMFM scores. These results were both statistically significant and clinically meaningful. Another factor that should be considered is the clinically meaningful between-group difference in GMFM scores that occurred at baseline. However, this difference was not statistically significant, and our analysis method adjusted for the baseline value of the GMFM score. This difference in baseline GMFM scores was a product of randomization, and likely emphasized by the small sample size. Finally, several participants had parent-reported secondary conditions such as attention deficits and intellectual disability, which may have influenced

individuals' engagement in the study.³³ Despite the large treatment effects detected in this study, replication of our results in a larger sample is needed.

This RCT demonstrated that a combined programme of FES cycling, goal-directed training, and adapted cycling can lead to meaningful functional improvements in children with CP (GMFCS levels II–IV), including those who are non-ambulant. Our results reinforce the importance of an individualized approach to FES combined with goal-directed training and adapted cycling programmes in children with CP. The goal-directed focus of the intervention, addition of FES and motor support and adapted cycling, may explain the large treatment effects found in the current study compared to previous stationary cycling interventions for children with CP.²⁴

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Supporting information

The following additional material may be found online:

Table S1: Example individualized training programme to address participant's COPM goals

Table S2: Description of the bikes and trikes used by the intervention group, including specific attachments, modifications and adaptations

Table S3: Amount and content of usual care during the intervention period

Figure S1: Participant flow diagram.

Figure S2: Scatter plots with regression lines for the primary outcome measures.

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Table 1: Baseline characteristics of participants who enrolled in an 8-week randomized controlled trial investigating the efficacy of functional electrical stimulation (FES) cycling, adapted cycling, and goal-directed training

Participant characteristics	Intervention group (n=11)	Control group (n=10)
Demographics		
Mean age, y:mo (SD)	9:10 (3:2)	10:10 (2:7)
Males	5 (45)	3 (30)
Anthropometrics		
Mean height (SD), cm	135 (10)	134 (20)
Mean mass (SD), kg	30.39 (9.5)	41.3 (20.3)
Mean BMI (SD)	16.45 (3.4)	22.09 (8.1) ^a
GMFCS level (stratified)		
II	3 (27)	3 (30)
III	3 (27)	3 (30)
IV	5 (45)	4 (40)
Topographical involvement		
Unilateral	3 (27)	2 (20)
Bilateral	8 (72)	8 (80)
Predominant motor type		
Spastic	9 (81)	10 (100)
Other	2 (18)	0
Primary mobility aid		
Nil aid	3 (27)	3 (30)
Walking frame	1 (9)	0 (0)
Manual wheelchair (self-propels)	2 (18)	2 (20)
Manual wheelchair (does not self-propel)	5 (45)	1 (10)
Power wheelchair	0 (0)	4 (40)
Relevant medical history		
Attentional/behavioural problems	5 (45)	1 (10)
Intellectual disability ^b	7 (63)	3 (30)
Non-verbal	4 (36)	0 (0)
Visual impairment	4 (36)	1 (10)
Hearing impairment	0 (0)	1 (10)
Epilepsy (controlled with medication)	4 (36)	1 (10)
Selective dorsal rhizotomy (>12mo ago)	1 (9)	1 (10)
Cycling experience and equipment		
Previous FES cycling experience	1 (9)	4 (40)
Previous cycling experience	9 (81)	9 (90)
Participated in cycling at the time of study enrolment	3 (30)	2 (20)
Owned an appropriate bike at study enrolment	3 (30)	3 (30)

Data are *n* (%) unless otherwise stated. ^aBody mass index (BMI) was significantly higher in the control group compared to the intervention group (mean difference=5.64; 95% CI=0.07–11.21; *p*=0.05). GMFCS, Gross Motor Function Classification System.

^bIntellectual disabilities were determined by parent report or attendance at a special school. No formal IQ testing was performed.

Table 2: Results from the primary outcome measures at baseline and follow-up, including the Gross Motor Function Measure (GMFM) and Canadian Occupational Performance Measure (COPM)

Outcome measure	Intervention group <i>n</i> =11		Control group <i>n</i> =10		Between-group difference at T2 after baseline adjustment Mean difference (95% CI); <i>p</i>
	Mean (SD)		Mean (SD)		
	Baseline (T1)	Follow-up (T2)	Baseline (T1)	Follow-up (T2)	
GMFM					
GMFM-88	52.8 (22.3)	62.6 (24.7)	61.3 (23.6)	63.8 (22.8)	7.4 (2.3, 12.6); 0.007*
GMFM-66	49.9 (9.84)	56.3 (11.60)	55.0 (13.4)	55.8 (13.9)	5.9 (3.1, 8.8); <0.001*
Goal score	46.4 (10.9)	63.0 (14.4)	57.0 (19.0)	60.0 (18.9)	13.5 (5.9, 21.0); <0.001*
COPM					
Performance	2.2 (1.1)	7.0 (1.2)	2.7 (1.6)	2.9 (1.4)	4.4 (3.9, 5.3); <0.001*
Satisfaction	2.4 (1.1)	7.6 (1.6)	3.1 (1.6)	3.0 (1.7)	5.2 (4.0, 6.4); <0.001*

Note: *Statistically significant result ($p<0.05$); There was no missing data for primary outcome measures at baseline or follow-up; **Abbreviations:**SD, standard deviation; CI, confidence interval.

Table 3: Results from the secondary outcome measures at baseline and follow up, including peak cycling power and resistance, the Pediatric Evaluation of Disability Inventory Computer Adaptive Test (PEDI-CAT), and Participation and Environment Measure – Child and Youth (PEMCIY)

Outcome measure	Intervention group Mean (SD)		Control group Mean (SD)		Between-group difference at T2 after baseline adjustment Mean difference (95% CI); <i>p</i>
	Baseline (T1)	Follow-up (T2)	Baseline (T1)	Follow-up (T2)	
Cycling test^a	<i>n</i> =11	<i>n</i> =11	<i>n</i> =8	<i>n</i> =8	
Peak power ^a	17.3 (19.2)	31.9 (20.1)	26.6 (21.8)	31.9 (21.8)	8.0 (-4.0, 20.0); 0.18
Peak resistance ^a	3.9 (2.8)	7.2 (4.4)	5.7 (2.7)	5.8 (2.4)	3.4 (1.0, 5.8); 0.009*
PEDI-CAT	<i>n</i> =11	<i>n</i> =11	<i>n</i> =10	<i>n</i> =10	
Daily activities	50.6 (4.2)	51.5 (4.7)	54.0 (4.2)	54.4 (5.5)	0.8 (-1.4, 2.9); 0.48
Mobility	57.3 (5.1)	56.6 (5.7)	56.6 (6.7)	56.0 (7.1)	-0.2 (-3.5, 3.2); 0.93
Social	64.8 (5.6)	64.5 (6.3)	67.9 (4.1)	67.8 (3.9)	-0.3 (-2.1, 1.6); 0.77
Responsibility	45.6 (7.1)	45.8 (5.4)	51.1 (4.0)	51.0 (4.6)	-1.2 (-3.9, 1.4); 0.34
PEMCIY home	<i>n</i> =11	<i>n</i> =11	<i>n</i> =10	<i>n</i> =10 ^b	
Participation	4.7 (1.4)	5.1 (0.8)	5.9 (0.6)	5.4 (1.1)	0.5 (-0.3, 1.2); 0.20
Involvement	3.1 (1.2)	3.3 (1.2)	4.1 (0.4)	4.0 (0.4)	0.2 (-0.2, 0.6); 0.35
Environmental support	49.2 (25.1)	53.8 (24.5)	60.0 (20.3)	55.0 (19.7)	5.8 (-10.3, 22.0); 0.46
Environmental barriers	12.1 (15.5)	9.1 (12.6)	15.0 (13.5)	10.8 (10.4)	0.2 (-5.3, 5.8); 0.93
PEMCIY school					
Participation	3.7 (1.4)	3.4 (0.9)	3.2 (1.4)	3.4 (1.6)	-0.4 (-1.3, 0.5); 0.33
Involvement	3.0 (1.0)	3.1 (1.4)	3.9 (1.0)	4.0 (1.1)	-0.1 (-0.8, 0.6); 0.73
Environmental support	57.2 (20.3)	63.1 (22.9)	65.9 (20.3)	55.9 (21.3)	13.7 (-0.3, 27.7); 0.06
Environmental barriers	12.8 (10.1)	8.0 (11.8)	8.8 (10.1)	11.2 (12.8)	-6.5 (-14.3, 1.3); 0.10
PEMCIY community					
Participation	2.1 (1.1)	2.4 (0.7)	2.4 (1.1)	2.2 (1.3)	0.5 (-0.2, 1.1); 0.13
Involvement	3.0 (0.8)	3.5 (0.8)	3.8 (0.8)	4.1 (0.8)	-0.4 (-1.1, 0.3); 0.27
Environmental support	36.4 (19.6)	43.8 (29.4)	35.6 (19.6)	50.0 (22.8)	-6.9 (-25.4, 11.7); 0.45
Environmental barriers	27.3 (18.2)	15.9 (20.0)	20.6 (18.2)	18.7 (20.6)	-8.3 (-21.5, 4.8); 0.20

*Statistically significant result ($p < 0.05$); ^aCycling test data is missing for two participants in the control group because of technical issues at baseline. ^bA PEMCIY baseline score was carried forward for one participant in the intervention group as a questionnaire was not returned at follow up. SD, standard deviation; CI, confidence interval.