Cycling With Functional Electrical Stimulation in an Adult With Spastic Diplegic Cerebral Palsy

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**Background and Purpose.** Adults with cerebral palsy (CP) are at risk for decreased mobility and health complications, and exercise may combat some of these negative changes. Because people with CP have difficulty generating sufficient muscle force, exercise augmented with functional electrical stimulation (FES) is an option for increasing exercise intensity. This mixed-method (quantitative-qualitative) case report describes the effects—across the *International Classification of Functioning, Disability and Health* (ICF) model—of cycling with FES (FES cycling) in an adult with CP.

**Case Description.** An ambulatory 49-year-old man with spastic diplegic CP cycled with FES at home for 30 minutes, 3 times per week, for 12 weeks. Volitional efforts were augmented by FES of the bilateral quadriceps, gastrocnemius, and gluteal muscles. Testing was performed before and after the intervention and 4 weeks after intervention withdrawal.

**Outcomes.** After training, quadriceps muscle strength (force-generating capacity) improved by 22.2%, hamstring muscle strength improved by 18.5%, and the Timed “Up & Go” Test time decreased from 11.9 to 9.0 seconds. The patient reported increased performance and satisfaction for self-identified goals at the ICF level of participation, and his score on the Medical Outcomes Study 36-Item Health Survey questionnaire increased from 62.1 to 77.6. However, he reported increased back pain, which he attributed to positioning while cycling. Qualitative interviews provided context (the patient’s perspective) for some of the quantitative results.

**Discussion.** The patient made gains in body structure and function, activity, and participation (ICF levels) after FES cycling. The mixed-method approach provided insight into his experiences and perceptions about the measures assessed quantitatively. Further investigation on FES cycling in this population as well as positioning during cycling is warranted.
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Fitness levels of people with disabilities have gained attention through Healthy People 2010, a focus of which is secondary conditions of disability and increasing physical fitness. Inactivity in people with disabilities can lead to deconditioning, adversely affecting the cardiovascular system, bone density, and muscle health. This deconditioning often results in a reduction in overall activity level, which can lead to social isolation and decreased self-esteem. Adults with cerebral palsy (CP) are at risk for deconditioning because mobility often declines through the adolescent and young adult years. Common secondary conditions are pain, fatigue, declining physical performance, and musculoskeletal changes. In addition, adults with CP are likely at higher risk of medical complications, including metabolic syndrome and coronary heart disease. Despite these risks, there are limited opportunities for adults with CP—relative to children with CP—to participate in physical activity, and little research has been dedicated to addressing this concern. Because most children with CP live into adulthood, appropriate interventions for this population are critical.

Cycling is an exercise that challenges the muscular and cardiovascular systems, potentially leading to improved health, fitness, and well-being. Cycling with functional electrical stimulation (FES) (FES cycling) has been primarily used by people with spinal cord injury (SCI); improvements have been seen in bone mineral density, muscle strength (force-generating capacity), and cardiorespiratory measures. Recent reports indicated benefits for people after stroke; improvements in strength and motor control were seen when an FES cycling program was added to traditional rehabilitation. However, there have been no reports of FES cycling for adults with CP.

Cycling with FES may be a suitable intervention for adults with CP because the seated position decreases balance demands, and FES can create or augment pedaling forces. Many people with CP may be incapable of generating sufficient forces during cycling to reach the exercise intensity needed for optimal fitness-related outcomes and musculoskeletal benefits. Cycling can be performed at home because home FES cycling units are available, and some insurance companies assist with costs. In addition, many people can perform the activity independently, and others may need assistance only for setup before and breakdown after each session. The combination of decreased balance demands; increased muscle recruitment, exercise intensity, and independence; and the possibility of home intervention makes FES cycling a potential treatment option for addressing aging-related effects reported by adults with CP.

This mixed-method (quantitative-qualitative) case report describes the effects—across the International Classification of Functioning, Disability and Health (ICF) model—of a home FES cycling program in an adult with CP.

Case Description: Patient History and Systems Review
The patient was a 49-year-old male with spastic diplegia classified as Gross Motor Function Classification System level II (walks without an assistive device but has limitations walking outdoors and in the community). He reported first walking at 2 years of age. His past medical history was significant for bilateral hamstring muscle lengthening (at 14 years of age), bilateral hip osteoarthritis, right knee chondromalacia, an L3-L4 disk herniation diagnosed 2 years earlier and leading to back pain and right lower-extremity paresthesia with longer-distance ambulation, a right rotator cuff repair, and a left distal clavicle excision. He had no history of cardiovascular disease, pulmonary disease, or joint instability or dislocation.

His goals were to improve strength and endurance through intensive exercise. He had previously, but not recently, exercised routinely. His primary means of exercise had been a stationary recumbent cycle at a local gym. He reported being unemployed and on disability due to "wearing out of my shoulders." Before this time, the patient was employed as a physical therapist for 26 years. He had previously undergone physical therapy for back pain and was self-treated or treated by medical professionals as needed closer to his home. He reported performing Williams flexion and extension exercises for his low back pain. Because he was receiving care at home as needed, a formal assessment of his low back pain was not performed.

Before examination and intervention, the patient signed a consent form approved by the Institutional Review Board at the University of the Sciences in Philadelphia. His primary care physician cleared him medically for FES cycling.

Clinical Impression 1
On the basis of his medical history and current status, the patient was considered a potential candidate for...
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FES cycling. Although he reported back pain due to his herniated disk, he had used a recumbent cycle in the past without any pain exacerbation. On the basis of literature about FES cycling for people with SCI,10 the patient's goals of increased strength and endurance had the potential to be affected by the intervention. Further examination to identify specific limitations across the ICF model was warranted. This approach was chosen to determine the patient's deficits and to allow the examination of potential effects because FES cycling has not been applied to people with CP.

Examination

The patient was independent in functional mobility and activities of daily living. He reported decreased walking endurance with increased energy expenditure compared with when he was younger. His gait was characterized bilaterally by decreased dorsiflexion during swing, a Trendelenburg pattern, decreased base of support, decreased hip and knee flexion during swing, and decreased hip and knee extension during stance.

Measurements at different levels of the ICF (body structure and function, activity, and participation) were obtained. Measures at the level of body structure and function included passive range of motion (PROM), body composition, lower-extremity strength and spasticity (velocity-dependent resistance to stretch), and pain. Lower-extremity PROM decreased during hip extension (Thomas test: left, ~15°; right, ~14°), hip abduction (left, 15°; right, 13°), and dorsiflexion (left, 6°; right, 2°), and hamstrings muscle length decreased (popliteal angle: left, 40°; right, 41°). Increased tone (hypertonicity or resistance to passive stretch) was noted in the hip and knee flexor, hip adductor, and ankle plantar-flexor muscles during passive movements; however, the patient's legs could easily be moved passively through the majority of his PROM. Leg lengths, measured with a tape measure from the anterior superior iliac spine to the medial malleoli, were equal (left, 88.8 cm; right, 89 cm).

Body composition was assessed with a bioimpedance monitor,11 which revealed a fat-free mass of 72.2%; reports of the reliability and validity of this device have been mixed.13 The strength of the right isometric quadriceps, hamstrings, anterior tibialis, and triceps surae muscles was assessed with a Biodex 3 computerized dynamometer,2 and values from the 3 best trials were used (Tab. 1).

The spasticity of the right hamstrings and gastrocnemius muscles was assessed with the Biodex 3 device, which was used to move the extremity passively at 5, 15, 60, 90, and 180°/s to measure resistance to passive movement. Peak values for resistive torque (the highest torque throughout the range) and work (resistance over the full range or area under the curve) were calculated. Moderate test-retest reliability (intraclass correlation coefficients of 0.72 for 180°/s and <.51 for slower movements) has been shown for passive testing via dynamometry in children with CP.14 and measures of torque have been shown to correlate with Ashworth scale scores in people with SCI (r = 0.59-.82).12 Overall, there was a trend for increasing resistance to movement at progressively higher angular velocities (Fig. 1). Pain levels were assessed with the McGill-Melzack Pain Questionnaire, a tool used for people with disabilities (Tab. 1).16

Measures at the level of activity included the Six-Minute Walk Test (6MWT), the Timed "Up & Go" Test (TUG), and gait spatiotemporal parameters (Tab. 1). Perceived exertion during the 6MWT was measured with the OMNI Scale of Perceived Exertion (OMNI).17 Data obtained with the 6MWT have been shown to be reliable in adults with CP (intraclass correlation coefficient = .97-.99). When a practice test is performed, repeatability is ±0.1 m.16 During our treatment program, only 1 test was performed because of the amount of data collected and concern about fatigue. The reliability of TUG scores has been established for people with stroke19 but not for adults with CP.20 Gait spatiotemporal parameters were assessed using an instrumented walkway (~4.3 m [14 ft]; GaitRite Platinum®) with and without shoes. The walkway data have been shown to be reliable for younger and older people (intraclass correlation coefficient = 0.82-0.92).21

Measurements at different levels of activity included the Canadian Occupational Performance Measure (COPM) and the Medical Outcomes Study 36-Item Short-Form Health Survey questionnaire (SF-36). With the COPM, self-identified goals were identified and assessed (Tab. 2) through semistructured interviews. The patient rated his performance and satisfaction for each goal on a scale of 1 to 10 (10 being the best). The COPM has been shown to be sensitive to change in children with CP (effect size = 1.44-1.61)22 and valid in various populations.23 The SF-36, a tool that has been shown to be internally consistent (Cronbach alpha = 0.83-0.92) and valid,24 was used to measure health-related quality of life (Tab. 1).

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1 RIL Systems Inc., 35920 Harper Ave, Clinton Township, MI 48035.
3 CIR Systems Inc., 60 Garlock Dr, Havertown, PA 19083.
Table 1.
Quantitative Results

<table>
<thead>
<tr>
<th>Measure</th>
<th>Baseline</th>
<th>Postintervention</th>
<th>% Change Postintervention</th>
<th>Withdrawal</th>
<th>% Change at Withdrawal</th>
</tr>
</thead>
<tbody>
<tr>
<td>Body mass index (kg/m²)</td>
<td>23.02</td>
<td>23.21</td>
<td>0.8</td>
<td>23.69</td>
<td>-2.1</td>
</tr>
<tr>
<td>Fat-free mass (%)</td>
<td>72.2</td>
<td>71.3</td>
<td>0.1</td>
<td>71.4</td>
<td>0.2</td>
</tr>
<tr>
<td>Quadriceps muscle strength (N)</td>
<td>584.11</td>
<td>713.92</td>
<td>22.2</td>
<td>579.18</td>
<td>-18.0</td>
</tr>
<tr>
<td>Hamstring muscle strength (N)</td>
<td>264.12</td>
<td>313.01</td>
<td>18.3</td>
<td>323.78</td>
<td>3.3</td>
</tr>
<tr>
<td>Anterior tibialis muscle strength (N)</td>
<td>102.69</td>
<td>114.02</td>
<td>11.0</td>
<td>125.70</td>
<td>10.2</td>
</tr>
<tr>
<td>Triceps surae muscle strength (N)</td>
<td>204.15</td>
<td>191.01</td>
<td>-6.4</td>
<td>154.45</td>
<td>-19.1</td>
</tr>
<tr>
<td>McGill-Melzack Pain Questionnaire score*</td>
<td>2</td>
<td>2</td>
<td>0.0</td>
<td>1</td>
<td>-50.0</td>
</tr>
<tr>
<td>Pain right now</td>
<td>3</td>
<td>4</td>
<td>33.3</td>
<td>3</td>
<td>-25.0</td>
</tr>
<tr>
<td>Pain at worst</td>
<td>1</td>
<td>1</td>
<td>0.0</td>
<td>1</td>
<td>0.0</td>
</tr>
<tr>
<td>Six-Minute Walk Test score (m)</td>
<td>403.6</td>
<td>423.7</td>
<td>5.0</td>
<td>435.3</td>
<td>2.7</td>
</tr>
<tr>
<td>Timed &quot;Up &amp; Go&quot; Test time, s, X (SD)</td>
<td>11.9 (0.4)</td>
<td>9.0 (0.8)</td>
<td>-24.4</td>
<td>9.4 (0.4)</td>
<td>4.4</td>
</tr>
<tr>
<td>Gait speed BF, m/s, X (SD)</td>
<td>1.12 (0.08)</td>
<td>1.21 (0.04)</td>
<td>8.0</td>
<td>1.20 (0.06)</td>
<td>-0.8</td>
</tr>
<tr>
<td>Cadence BF, steps/min, X (SD)</td>
<td>133.5 (4.7)</td>
<td>133.8 (4.3)</td>
<td>0.6</td>
<td>135.2 (2.9)</td>
<td>1.0</td>
</tr>
<tr>
<td>Left step length BF, m, X (SD)</td>
<td>0.47 (0.02)</td>
<td>0.50 (0.01)</td>
<td>6.4</td>
<td>0.50 (0.01)</td>
<td>0.0</td>
</tr>
<tr>
<td>Right step length BF, m, X (SD)</td>
<td>0.54 (0.03)</td>
<td>0.59 (0.01)</td>
<td>9.3</td>
<td>0.56 (0.02)</td>
<td>-3.1</td>
</tr>
<tr>
<td>Gait speed SO, m/s, X (SD)</td>
<td>1.27 (0.06)</td>
<td>1.18 (0.08)</td>
<td>-7.1</td>
<td>1.18 (0.11)</td>
<td>0.0</td>
</tr>
<tr>
<td>Cadence SO, steps/min, X (SD)</td>
<td>137.5 (5.8)</td>
<td>129.8 (1.7)</td>
<td>-5.6</td>
<td>134.4 (2.0)</td>
<td>2.6</td>
</tr>
<tr>
<td>Left step length SO, m, X (SO)</td>
<td>0.51 (0.02)</td>
<td>0.52 (0.02)</td>
<td>2.0</td>
<td>0.51 (0.04)</td>
<td>-1.9</td>
</tr>
<tr>
<td>Right step length SO, m, X (SO)</td>
<td>0.61 (0.02)</td>
<td>0.57 (0.04)</td>
<td>-6.6</td>
<td>0.35 (0.03)</td>
<td>-3.5</td>
</tr>
<tr>
<td>SF-36 score*</td>
<td>62.1</td>
<td>77.6</td>
<td>25.0</td>
<td>77.6</td>
<td>0.0</td>
</tr>
<tr>
<td>Total</td>
<td>18.75</td>
<td>23.3</td>
<td>23.3</td>
<td>68.8</td>
<td>10.1</td>
</tr>
<tr>
<td>Physical function</td>
<td>75</td>
<td>62.5</td>
<td>-16.7</td>
<td>100</td>
<td>60.0</td>
</tr>
<tr>
<td>Role limits: physical</td>
<td>100</td>
<td>100</td>
<td>0.0</td>
<td>100</td>
<td>0.0</td>
</tr>
<tr>
<td>Role limits: emotional</td>
<td>56.25</td>
<td>81.25</td>
<td>44.4</td>
<td>75</td>
<td>-7.7</td>
</tr>
<tr>
<td>Energy/fatigue</td>
<td>90</td>
<td>75</td>
<td>-16.7</td>
<td>80</td>
<td>6.7</td>
</tr>
<tr>
<td>Emotional well-being</td>
<td>87.5</td>
<td>87.5</td>
<td>0.0</td>
<td>100</td>
<td>14.3</td>
</tr>
<tr>
<td>Social function</td>
<td>67.5</td>
<td>45</td>
<td>-33.3</td>
<td>57.5</td>
<td>27.8</td>
</tr>
</tbody>
</table>

* BF = barefoot, SO = shoes on, SF-36 = Medical Outcomes Study 36-Item Short-Form Health Survey questionnaire.
* Four weeks after intervention withdrawal.
* Measured on a scale from 1 to 5, with 1 representing mild pain and 5 representing exacerbating pain.
* Measured on a scale from 0 to 100, with 100 representing the highest health-related quality of life.

Clinical Impression 2

On the basis of the examination findings (Tab. 1), the patient had several issues that might be addressed through FES cycling. At the level of body structure and function, he had increased body fat and decreased muscle strength. His PROM was adequate for safe FES cycling. Measures at the level of activity indicated decreased gait speed, increased TUG time, and perceived exertion half-way between "somewhat easy" and "somewhat hard" during the 6MWT. Measures at the level of participation indicated decreased satisfaction with performance of some activities and decreased health-related quality of life.

Cycling with FES was chosen because it could be performed at home without assistance and might increase exercise intensity and overall endurance and address strength deficits. It was not known whether gains would lead to improvements in activity and participation. Reexamination after 12 weeks was conducted on the basis of literature.
Figure 1.
Peak resistive torque and work findings from spasticity testing at 4 tested angular velocities (20, 60, 90, and 120°/s). Post=postintervention.

Table 2.
Canadian Occupational Performance Measure (COPM) Goals and Ratings of Performance and Satisfaction*

<table>
<thead>
<tr>
<th>Self Identified Goal</th>
<th>Score for:</th>
<th>Performance</th>
<th>Satisfaction</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>B</td>
<td>P</td>
</tr>
<tr>
<td>Donning socks</td>
<td>3</td>
<td>6</td>
<td>7</td>
</tr>
<tr>
<td>Bending forward to pick up objects</td>
<td>2</td>
<td>5</td>
<td>6</td>
</tr>
<tr>
<td>Going up or down stairs without railing</td>
<td>1</td>
<td>8</td>
<td>4</td>
</tr>
<tr>
<td>House cleaning</td>
<td>4</td>
<td>3</td>
<td>4</td>
</tr>
<tr>
<td>Increasing exercise frequency</td>
<td>1</td>
<td>6</td>
<td>3</td>
</tr>
<tr>
<td>Overall average score</td>
<td>2.2</td>
<td>5.6</td>
<td>4.8</td>
</tr>
</tbody>
</table>

*Scores on the COPM range from 1 to 10 (10 being the best). B=baseline, P=postintervention, W=4 week after intervention withdrawal.

Intervention

The RT3000, a compact, freestanding stationary FES cycle with a computerized controller (Fig. 2), was loaned to the patient. The cycle does not have a seat, so a chair or wheelchair can be used. The cycle was set to allow volitional cycling with augmentation by FES to increase intensity about SCI. Cycling was then discontinued, and another reexamination was completed 4 weeks later. In addition to quantitative measures, 2 semistructured interviews were conducted at follow-up visits to gain insight from the patient’s perspective about his experiences and expectations for outcomes of the intervention.

3 Restorative Therapies Inc, 907 S Lakewood Ave, Baltimore, MD 21224.
sity. The patient sat in a standard chair secured to the cycle using tie-down hooks. During all sessions, heart rate was measured through the integrated monitor.

The intervention involved three 30-minute FES-assisted cycling sessions each week for 12 weeks at home after 2 days of on-site supervised cycling. The patient cycled volitionally at a targeted cadence of 40 rpm, and FES maintained the cadence of 40 rpm with increasing resistance levels. Functional electrical stimulation was delivered to the bilateral gluteus maximus, quadriceps, and gastrocnemius muscles through self-adhesive electrodes with the following stimulation parameters: 250 microseconds, 33 Hz, and maximum currents of 80 mA (quadriceps muscle) and 60 mA (gluteus maximus and gastrocnemius muscles). The gastrocnemius muscle current was decreased to 40 mA after 3 weeks because of discomfort. These muscles were chosen on the basis of biomechanical work that indicated deficits in the extension phase of cycling in adolescents with CP.28

Resistance started at 1 Nm (lowest setting) and was increased in increments of 0.14 Nm so that volitional effort plus FES would enable cycling against higher loads. The initial resistance level was set so that the combination of volitional effort and FES would result in a 30-minute session. This level was determined by increasing the resistance to that which the patient could achieve without maximal effort so that cycling could be maintained for 30 minutes. Minor adjustments were made to this level while the patient was on site. For home sessions, the patient was instructed to increase the resistance level at the next session if he could complete 30 minutes at the set level. However, if effort decreased, the stimulation current would ramp up by 1% per minute until effort increased or the maximum current was reached. If the patient was unable to maintain 40 rpm with volitional effort plus FES for more than 30 seconds, the cycle would go into cooldown. If this scenario occurred, the patient was instructed to decrease resistance and resume cycling after 5 minutes of rest.

Before the intervention, the patient received training on the use of the cycle. The stimulation amplitude initially was set low and then was increased to tolerable levels. After each session, the patient was instructed to stretch his bilateral hip flexor, hamstring, and gastrocnemius muscles at a minimum of 1 time each for 30 seconds. Throughout the 12-week intervention, the patient received telephone calls from a physical therapist once per week for the first 3 weeks and then every other week to determine whether there were any problems with the intervention or whether the patient had any questions or needs.

**Outcome**

**Quantitative Data**

The patient increased cycling resistance and exercising heart rate throughout the 12-week intervention (Fig. 5). Average stimulation delivered varied but did not increase with increasing resistance during the final 6 weeks.

Quantitative outcome measures were compared with minimal detectable changes (MDCs) and minimal clinically important differences (MCIDs) when available from the literature. The MDC is the smallest amount of change that is not likely to occur by chance; thus, the MDC takes measurement error into account. The MCID indicates a change that is meaningful for the patient.30 Although the MDC and the MCID represent different concepts, they assist in the interpretation of the results. For adults with CP, MDCs and MCIDs are lacking in the literature, so values for other populations were used for comparison.

For measures at the level of body structure and function (Tab. 1), only
Functional Electrical Stimulation Cycling in Cerebral Palsy

Heart Rate

![Heart Rate Graph](image)

Resistance and Stimulation

![Resistance and Stimulation Graph](image)

Figure 3.
Heart rate (HR), resistance, and stimulation levels across the intervention period. The patient decreased the resistance level during the final 5 sessions because of back discomfort during cycling against higher loads. bpm=beats per minute.

Changes in quadriceps and hamstring muscle strength were meaningful. A change of 8.2% to 12.9% is considered a clinically meaningful change in isokinetic knee strength following exercise after an anterior cruciate ligament injury (standardized response mean=0.49-0.60). The patient in this report gained 22.2% in the quadriceps muscle and 18.5% in the hamstring muscle. Quadriceps muscle strength declined to the baseline after withdrawal (i.e., 4 weeks after intervention withdrawal), but hamstring muscle strength continued to increase. The patient’s body mass index and percentage of lean tissue remained stable throughout the program. The results of hamstring muscle spasticity testing (Fig. 1) were mixed, with trends for increased peak resistive torque and work at lower angular velocities but decreased peak resistive torque and work at higher velocities. Mixed results also were seen for the plantarflexor muscles: however, torque and work both showed trends for decreases at the lowest and highest velocities. No MCID or MDC is available for spasticity testing assessed by resistance to passive motion. For the McGill-Melzack Pain Questionnaire, a change of 24% in present pain intensity has been considered statistically significant after feedback, hypotonic training, or both. Our patient showed no change in pain intensity after the intervention but showed a 50% decrease after withdrawal (from 2 to 1).

For measures at the level of activity (Tab. 1), gains in the TUG were greater than the reported MDC of 2.49 seconds in people with Alzheimer disease. Our patient’s improvement of 2.9 seconds declined slightly to 2.5 seconds after withdrawal. However, the score was lower than the MCID of 4.0 seconds reported for older African American people. It is not known whether this change is clinically meaningful for adults with CP. For the other activity measures, no changes were seen. The MDC for the 6MWT in adults with CP is 66 m when only 1 test is performed, so our patient’s changes of 20.1 m after the intervention and 11.6 m after withdrawal were not true changes. Scores on the OMNI during the 6MWT decreased from 5 to 4 and then increased to 6, but an MDC or an MCID for the OMNI was not available for comparison. Although our patient showed gains in barefoot gait speed of 0.09 m/s and in step length of 0.03 to 0.05 m, these changes likely are not clinically meaningful because the MDCs for gait speed and step length in older people are 0.126 m/s and 0.07 m, respectively. It is not known whether these values apply to adults with CP or whether the walkway is reliable for adults with CP.

For measures at the level of participation, changes were seen in COPM and SF-36 scores. Our patient rated...
his satisfaction and performance on the COPM as improved for 4 of 5 goals (Tab. 2). Changes of 2.5 to 2.7 points in the overall average score on the COPM are detectable in adults with various diagnoses, and our patient's gains in performance scores were greater than these values (Tab. 2). For the SF-36, the MCID is at least 10 points after knee arthroplasty, and our patient's score improved by 15.5 points after training and remained stable.

Qualitative Data
Semistructured interviews were conducted and audiorecorded after the intervention and again after withdrawal to gain insight from the patient's perspective about his experiences and expectations for intervention outcomes (the interview questions are shown in the Appendix). Each interview was transcribed and reviewed by the patient to ensure that his perspectives were accurately captured. No additional information was added through this member check. A second member check of the completed article provided insight into the paradoxical qualitative findings and quantitative outcome measures. These qualitative interviews allowed the identification, from the patient's perspective, of outcomes not assessed through quantitative measures.

Although definitive themes cannot be determined from 1 patient, preliminary factors were identified and used to develop a conceptual framework (Fig. 4) to guide future research. This patient's perspectives were categorized into 3 primary factors: prior health and functional status, anticipated outcomes, and motivating factors to participate. The levels of the ICF model were represented in his comments for all 3 factors. The interaction of these factors (represented by his perceived psychologic and physiologic benefits,
improved strength, endurance, or both during cycling. However, any increased endurance or strength did not appear to translate to walking distance on the 6MWT, even though perceived exertion during walking showed a trend toward a decrease. In addition, heart rate during cycling showed a trend toward an increase over time, and the patient was able to maintain a heart rate of more than 120 bpm for most sessions during the final 6 weeks.

The patient’s reports of increased pain with increasing resistance levels were concerning. He was instructed to decrease resistance, but he decreased it only slightly, and his back pain persisted. He later decreased it more significantly and reported relief but believed that he was not exercising to his capacity at the lower level. From his descriptions, modifications to the cycle and cycle setup to minimize cycle movement at higher resistance levels may be warranted. The cycle attaches to the chair with tie-down straps similar to those used for securing a wheelchair during transport. At the highest resistance, these straps were insufficient to minimize motion between the cycle and the chair and the motion of the cycle itself on the floor, making it necessary for the patient to hold the handlebars tightly. Bracing the cycle against a wall helped but did not eliminate the problem. These issues did not occur when the patient previously used a recumbent cycle at his local gym because the seat and the cycle were attached with rigid material. Therefore, cycle design should be considered when the FES cycle is used by people who can achieve higher resistance levels. In addition, the patient reported that the 90-degree angle of the chair was unlike that of the recumbent cycle at his gym and was an issue. The 90-degree angle would have increased the amount of flexion throughout the spine, likely placing more pressure on the patient’s lumbar spine and herniation. Therefore, alternate seating designs should be investigated.

In addition to alternate seating designs, a more thorough evaluation of the patient’s low back pain before the intervention might have provided information suggesting that cycling might increase his pain. Therefore, it is recommended that an in-depth evaluation of areas of the body that may be subjected to increased stress during cycling be performed. Although the patient was a physical therapist, closer and more frequent monitoring might have been needed.

Some gains achieved during the intervention were maintained, and some improvements continued to increase after withdrawal. Quadriceps muscle strength declined; therefore, strength alone cannot explain improvements in activity and participation. Mazzum et al.51 reported that strength is not a predictor of 6MWT distance in adults with CP and that increased strength is more important for more-demanding activities. Individual factors, such as self-perception of ability, interpretation of exercise intensity and outcomes, focus of concentration, and perception of control, influence affective responses to exercise.42 Such affective responses and beliefs should be explored to gain insight into performance on outcome measures and satisfaction with achieved outcomes. Therefore, the strength gains made by our patient may not have had an impact on the other outcome measures.

The application of a mixed-method approach expanded the scope of the program and provided insight into the patient’s perspectives and experiences.43 Although collecting both quantitative and qualitative data currently was effective,44 it was apparent that using quantitative measures such as SF-36 items to inform the qualitative interviews would have provided contextually rich follow-up questions. Merging the quantitative and qualitative data provided insight into the patient’s experiences, which provided a context for understanding the outcome measures. Embedding the qualitative data in the quantitative data clarified and supported the quantitative findings, as in issues of spasticity.28 Alternatively, comparison of the qualitative and quantitative data revealed a contradiction in reports of pain between the quantitative questionnaire and the qualitative interviews. This contradiction identified the need to explore outcomes within the context of a patient’s beliefs and attitudes across the levels of the ICF model. Completing the second member check allowed the patient an opportunity to provide insight into contradictory findings:

Donning my socks; before (cycling) I rated it 3 and after a 6, so that would be better. Four weeks later, I rated it a 7. As I told you . . . it’s very difficult to quantify—it depends on the day, and how tight the lumbar spine is. If I looked back, I might change a few numbers.

Consistent with the iterative nature of a qualitative approach, this case report has provided results indicating the effectiveness of the intervention in a patient and has afforded us experience to refine the mixed-method approach to achieve a broader and contextual understanding of clinically relevant factors.

It is not known whether the gains made by our patient would have occurred if he had cycled without FES. Functional electrical stimulation might have allowed him to cycle more vigorously than he would have voluntarily by providing greater muscle activation, sensory cues
about the correct timing for muscle activity, or both. Compared with adolescents with typical development, adolescents with CP have been shown to have increased muscle cocontraction,29 prolonged muscle activity,29 altered kinematics,29 and altered force application66 during volitional cycling. Functional electrical stimulation might have provided a more typical cycling pattern for our patient. However, research is needed to determine how the pattern may be changed and what benefits FES cycling may offer over volitional cycling for adults with CP.

Both authors provided concept/idea/project design, writing, and data collection and analysis. Dr Johnston provided project management, fund procurement, the patient, and facilities/equipment.

This case report was based on 1 patient in a pilot research study conducted to examine the effects of cycling with functional electrical stimulation in adults with cerebral palsy. Approval for that study was obtained through the Institutional Review Board at the University of the Sciences in Philadelphia.

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References


pain, and time needed to train) throughout the training process may provide contextual meaning to postintervention physical performance measures.

As the patient reflected upon the training experience, he described outcomes most commonly within the context of body structure and function. Performance of activities was second in frequency, and participation was third. Although the patient identified his goals at the activity and participation levels of the ICF model (Tab. 2), he made comments about impairments in body structure and function 3 times more often than comments about activity and participation in the interviews. In the factor sort, he identified goals at the participation level of the ICF model as most important:

Number 1 is exercise. Number 2 is social integration. Just the chance to do stuff more. Professionally, I still do stuff politically.

Although the patient did not identify maintaining a professional role as a goal before the intervention, he cited ongoing participation in professional activities as important in both interviews.

The patient indicated that exacerbation of impairments at the level of body structure and function were limiting factors in his ability to fully engage in the intervention and meet his goals:

It is because of my back pain, ... I think the main problem was that the chair was at a 90-degree angle, and every time I pedaled I was having so much pain. It was all the way down to L3, L4, numb at night, heaviness in the leg. I didn’t want to make it worse.

At withdrawal, the patient identified the positive effect of training and the subsequent negative effect of not being able to participate in the intervention:

At the beginning, I was able to get good peak heart rates going at about 160 to 170. ... I don’t believe my cardiovascular function is better (now), because I haven’t really worked out. I’ve gained weight.

In addition, the patient indicated that exacerbation of back pain resulted in a decline in activity level, as evidenced by comments about balance and frequency of falls:

Yeah, I would say I fall less. I usually (fall) about once a day. The question I have is: Have I been walking as much? And I’m not walking as much. I’m tired.

Discussion

The patient made gains at the levels of body structure and function, activity, and participation after the intervention. Gains in quadriceps and hamstring muscle strength and functional mobility (TUG) are encouraging because aging adults with CP tend to show declines in these areas. Qualitatively, the patient reported at withdrawal: “...lifting my leg up better. I'm not dragging my legs as much while walking.” Although there may not have been a clinically meaningful change in gait, improvement in a factor that the patient identified as contributing to a decrease in falls is meaningful. For measures at the level of participation, the patient's performance and satisfaction improved for self-identified goals (COPM) and the total SF-36 score improved, indicating that he believed that gains were made in the performance of common daily activities and his overall health-related quality of life. Increases in pain were inconsistent between the McGill-Melzack Pain Questionnaire and the semistructured interviews; the reasons for the increases in pain must be considered.

Two studies (n=10 in each) examined progressive resistive strength training performed twice per week for 10 weeks by nonathlete adults with CP. Gains were reported in lower-extremity strength, performance of the sit-to-stand task, gross motor function, walking speed, and TUG time, suggesting that increased strength can improve function. It is not known whether our patient made functional gains because of increased strength or whether other factors, such as endurance, were involved. Research on strength and endurance training of adults with CP is needed because of the lack of knowledge in this area. Research on improving walking performance through upright activity also is lacking.

Across data collection sessions, inconsistencies in the results of spasticity testing were seen. Typically, in spasticity testing, resistance to movement increases at higher angular velocities; this finding was not consistently obtained. Lower angular velocities test joint stiffness, whereas higher angular velocities test spasticity. The patient reported increased planter-flexor muscle spasticity: “My spasticity has been bad. Yes, especially in my calves;” however, the plantar flexor muscles did not show increased resistance to the fastest movement during testing. There are 2 potential reasons for this discrepancy: the patient's definition of spasticity differed from the medical definition, and the patient reported functionally how spasticity affected his life (whereas the dynamometer test is a static open kinetic chain measure).

Throughout the intervention, the patient increased cycling resistance to as high as 13.9 Nm, which is challenging. Even though stimulation levels varied, there was no concomitant increase as resistance was increased. This finding suggests that the patient...