The PEDALS stationary cycling intervention and health-related quality of life in children with cerebral palsy: a randomized controlled trial

SHARON K DEMUTH[1] | LORETTA M KNUTSON[2] | EILEEN G FOWLER[3]

1 Division of Biokinesiology and Physical Therapy, University of Southern California, Los Angeles, CA; **2** Department of Physical Therapy (retired), Missouri State University, Springfield, MO; **3** Department of Orthopaedic Surgery, Tarjan Center, UCLA/Orthopaedic Hospital Center for Cerebral Palsy, University of California, Los Angeles, CA, USA.

Correspondence to Dr Eileen G Fowler at UCLA/Orthopaedic Hospital Center for Cerebral Palsy, 22–70 Rehabilitation Center, 1000 Veteran Avenue, Los Angeles, CA 90095-1795, USA. E-mail: efowler@mednet.ucla.edu

This article is commented on by Taylor on pages 587–588 of this issue.

PUBLICATION DATA

Accepted for publication 4th March 2012.

Published online 14th May 2012.

ABBREVIATIONS

HRQOL	Health-related quality of life
MCID	Minimal clinically important difference
PEDALS	Pediatric Endurance and Limb Strengthening
PedsQL	Pediatric Quality of Life Inventory SF15
PODCI	Pediatric Outcomes Data Collection Instrument

AIM The aim of this study was to assess health-related quality of life (HRQOL) following a stationary cycling intervention in children with cerebral palsy (CP).

METHOD This was a phase I multisite randomized controlled trial with single blinding. HRQOL was evaluated using the Pediatric Quality of Life Inventory SF15 (PedsQL; children) and Pediatric Outcomes Data Collection Instrument (PODCI; parent proxy) before and after a 3-month stationary cycling intervention. Sixty-two children (29 male, 33 female; mean age 11y; range 7–18y) with spastic diplegic CP, classified as levels I to III on the Gross Motor Function Classification System, were enrolled. Paired and independent *t*-tests were used to evaluate within- and between-group differences respectively.

RESULTS Between-group differences, favoring the cycling group, were found for PedsQL emotional functioning (p=0.046) and Parental PODCI treatment expectations scores (p=0.006). Between-group differences were not found for other scales. Within-group improvements were found in the cycling group: PedsQL total score (+5.8; p=0.006), psychosocial health summary (+6.9; p=0.008), and school functioning (+8.0; p=0.038). PODCI satisfaction with symptoms decreased significantly only in the control group (–12.0; p=0.046).

INTERPRETATION A beneficial influence of exercise on pediatric emotional well-being and parental treatment expectations was found. The evidence was not strong for other aspects of HRQOL. Results support the positive relationship between physical fitness and emotional well-being in the general population. A child's perception is important when examining change in his or her emotional well-being due to intervention.

The inclusion of health-related quality of life (HRQOL) outcome measurements in clinical research is currently promoted. However, few studies of children with cerebral palsy (CP) have reported changes in HRQOL over time, and even fewer have studied changes following intervention. When choosing HRQOL instruments to measure outcomes for children with CP, investigators face challenges.[1] HRQOL is a multidimensional construct that includes both physical and psychological dimensions.[2–4] Some assessments take a general health approach while others are disease specific.[5] Additionally, questionnaires may be directed to the child or to parents/guardians as a proxy measure. Different versions are necessary for children of various ages, and some questionnaires exclude younger children because of the complexity of the questions. Although HRQOL instruments can characterize differences across condition severity, their responsiveness to change following intervention has not been well established in CP.[1]

In the general population, there is convincing evidence that regular physical activity improves psychological well-being.[6] A review article on exercise in young people without disabilities concluded that there was a strong positive relationship between physical activity and psychological health based on measures of anxiety and depression symptoms.[7] This relationship has not been as well studied following interventions for individuals with a disabling condition.[6] However, the evidence in individuals without disability gives reason to hypothesize that similar benefit might be found for children with CP. Among the few exercise intervention studies that have evaluated HRQOL in children with CP, there have been mixed results.[8–12] Improved HRQOL is not a consistent finding following exercise programs focused on strength training.[9,10] In fact, a study by Dodd et al.[9] reported a significant decrease in self-concept for scholastic and social competence in child participants after a home strength-training intervention. Engsberg et al.[10] used both parent proxy and child questionnaires to evaluate HRQOL following a strengthening exercise program. A significant improvement was found for the parent/guardian's but not for the child's responses.

 DOI: 10.1111/j.1469-8749.2012.04321.x

Available evidence supports improved psychosocial well-being following aerobic exercise for children with CP. Improved self-concept was reported for child participants, following interventions that either focused on aerobic exercise[11] or included both aerobic and strengthening components.[8] Verschuren et al.[12] studied the effects of an 8-month fitness exercise program using a randomized controlled trial design. During this 8-month study, the intervention group focused on aerobic exercise for the first 4 months, followed by anaerobic exercise for the last 4 months. Following the aerobic exercise period, parental reports of motor, autonomy, and cognition domains, as well as child reports of self-concept for athletic competence, improved. Only autonomy remained significantly higher for the intervention group following the anaerobic exercise period. Collectively, these studies suggest that aerobic exercise may have a positive influence on aspects of HRQOL for children with CP.

The aim of this study was to assess the effect of the Pediatric Endurance and Limb Strengthening (PEDALS) stationary cycling intervention on HRQOL in children with spastic diplegic CP. We previously reported the results of the PEDALS intervention relative to the International Classification of Functioning, Disability and Health (ICF) body structure and function and activity levels.[13] Significant improvements were found in walking/running endurance, gross motor function, and select measures of muscle strength for the cycling group. There was no significant difference between the cycling and control group. This report focuses on HRQOL. Considering the limited research examining stationary cycling in this population, PEDALS was designed as a phase I study.

METHOD

The design for this study was a phase I, multisite randomized controlled trial with single blinding. Power analysis determined that a sample size of 58 participants (29 intervention, 29 control) would have 80% power to detect a moderate effect size of 0.7.[13] The participants were randomly assigned to a control (no intervention) group or an intervention (cycling) group. Randomization was blocked by age (7–11y, 12–18y) and lower extremity selective voluntary motor control ability (good, fair) to minimize the effects of physical impairment and maturation. Participants who exhibited good selective motor control (isolated knee and ankle motion) bilaterally were assigned to the 'good' category for stratification. Participants who had fair voluntary selective motor control (isolated knee but not ankle motion) of at least one leg were assigned to the 'fair' category. Group assignment was determined using a computerized random number generator. Evaluators were blinded to group assignment.

The stationary cycling intervention consisted of 30 sessions over 12 weeks in community-based pediatric physical therapy clinics. Each 60-minute session was divided into strengthening and cardiorespiratory phases. A progressive resistive exercise protocol was used for the strengthening phase. For the cardiorespiratory phase, participants were encouraged to achieve a target heart rate range of between 70% and 80% maximum, calculated using the Karvonen method: target heart rate (HR) =([220–age]– $HR_{resting}$)×0.7 [or 0.8]+$HR_{resting}$.[14] Participants were asked to maintain their current physical activity level, including physical therapy, during the study. Each participant was provided with a physical activity calendar for the 12-week intervention period, so that group activity levels could be quantified. Cycling group participants did not report time spent in the cycling intervention on their calendars. An intervention was not provided for the control group. More detailed descriptions of the PEDALS intervention protocol have been reported.[13–15]

What this paper adds

- This is the first pediatric randomized controlled trial that investigates the effect of a stationary cycling intervention on health-related quality of life.
- The results demonstrate improved emotional well-being in the cycling group compared with the control group.
- A child's perception is important when examining change in his or her emotional well-being due to intervention.

Participants

Potential participants were recruited from southern California and south-west Missouri via brochures, flyers, and postings on disability-related websites. Inclusion criteria were: (1) spastic diplegic CP; (2) age between 7 and 18 years; (3) the ability to comply with simple verbal directions; (4) Gross Motor Function Classification System (GMFCS) levels I to III; and (5) selective motor control rating of good or fair for at least one leg.[13] Exclusion criteria were: (1) neurological surgery, orthopedic surgery, or implantation of a baclofen pump within 12 months preceding enrollment; (2) botulinum injections, new orthotics, serial casting, or alteration in medications acting on the neuromuscular system within 3 months preceding enrollment; (3) initiating exercises, sports, or physical therapy or changing assistive devices for walking within 3 months preceding enrollment; (4) difficulty in maintaining age-appropriate behavior; (5) serious medical conditions such as diabetes, cardiac disease, or uncontrolled seizures; (6) current participation in a fitness program; (7) significant contractures of the hip, knee, or ankle preventing passive movement of the legs through the pedaling cycle; and (8) poor bilateral voluntary selective motor control (unable to isolate knee or ankle joint motion out of synergy).[14] Study procedures were approved by the institutional review board of each institution. Informed consent was obtained from a parent or guardian and participants over 14 years of age. Informed assent was obtained from participants under 14 years of age.

Health-related quality of life instruments

We evaluated the perceptions of the child participant as well as his or her parent or guardian. HRQOL of the child participants was assessed using the Pediatric Quality of Life Inventory 4.0 Generic Core Scales SF15 (PedsQL).[16] More detailed HRQOL questions about physical capacity and satisfaction were directed to the parents using the Pediatric Outcomes Data Collection Instrument (PODCI).[2] Both the PedsQL and the PODCI have been used to measure the perceptions of children with CP and those of their parents/guardians.[17–20]

The PedsQL was designed to measure HRQOL in healthy children and those with a wide range of health conditions.[21] It has been shown to be valid and reliable.[3,16] The PedsQL assesses four dimensions of function: physical, emotional, social, and school.[3] The last three scales combine to create a psychosocial health summary. The 15 questions at baseline and follow-up are identical. The questionnaire has three versions, with selection based on the child's age. Simplified answer choices anchored to pictures of happy, neutral, or sad faces are used for children aged 5 to 7 years. A higher score represents a more positive HRQOL.

The PODCI was developed to assess HRQOL for children undergoing orthopedic treatment for musculoskeletal conditions. Good construct validity and reliability have been reported.[2] The PODCI has separate baseline and follow-up forms that differ slightly in language to capture change due to treatment at follow-up. There are two parent proxy versions based on the age of the child. There are 117 questions. The parent or guardian is asked to select one answer from at least four choices. The PODCI contains four sections: global function and symptoms, happiness, treatment expectations, and satisfaction with symptoms. A higher score represents a more positive HRQOL. After consulting with the developers of the PODCI, we altered the wording of the post intervention treatment expectations section to make it clear that the time period was limited to the previous 3 months during which the child was enrolled in the PEDALS study. A question asking whether a child used assistive devices for sitting and standing was removed as parents were confused about reporting whether orthoses were worn versus whether they were required for these activities.

Questionnaire administration and assessment

Both questionnaires were administered following a standardized protocol outlined in the study's manual of procedures.[14] The blinded evaluators were trained in questionnaire administration and passed a videotaped evaluation of competency with a 95% performance rating. The PedsQL was administered in English as all the children spoke English. The youngest children in the study (7y) used the PedsQL young child version; older children (8–12y) completed the child version and adolescents (13–18y) used the teen version. The PODCI parent proxy versions for parents or guardians of children (2–10y) or adolescents (11–18y) were used. Written Spanish and English language versions of the PODCI were administered. An interpreter was available to assist parents or guardians who spoke Spanish, Korean, or Japanese. Each question and all possible responses were read to the participant and the evaluator recorded the selected answer. This verbal administration method was chosen to ensure that all questions were answered and to eliminate the need for participant literacy. Parents or guardians and the child participants were separated during questionnaire administration so that each participant could answer the questions confidentially. The same parent or guardian completed both baseline and follow-up questionnaires.

Data analysis

Statistical analyses were performed using JMP version 6.0 software and SAS version 9 (SAS Institute, Cary, NC, USA). Demographics and characteristics were compared between the cycling and control groups using χ^2 tests for comparison of proportions and one-way analysis of variance for continuous

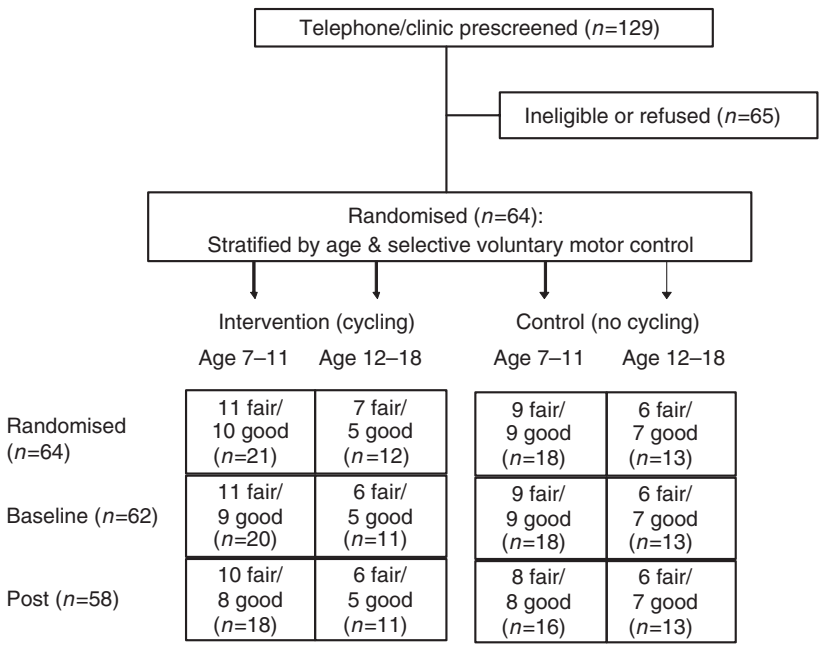

Figure 1: CONSORT trial diagram illustrating the flow of participants through the trial.[13] (Reprinted with permission of the American Physical Therapy Association)

variables. Baseline, post intervention, and change scores were calculated. Independent *t*-tests were used to evaluate the change scores between groups. As this was a phase I study, within-group changes were also of interest. Paired *t*-tests were applied to examine the difference between baseline and post intervention scores within groups. Significance level was set at $p<0.05$, and all *t*-tests were two tailed.

RESULTS

The CONSORT diagram is shown in Figure 1.[13] Of the 129 individuals who responded to recruitment efforts, 64 were randomized, 62 completed baseline testing, and 58 (29 cycling, 29 control) completed the study. There were a greater number of participants in the 7- to 11-year category. Blocking by age group was removed as we had greater difficulty in recruiting to the older age group. The two groups were well matched at baseline for demographics, characteristics, and physical ability (Table I).[13] Significant differences were not found for sex, age, ethnicity, race, selective voluntary motor control, GMFCS level, anthropometrics, medical problems, or gross motor function at baseline ($p>0.05$). Adherence to cycling session attendance was 89.6%. Protocol variations occurred for three cycling group participants who missed one, three, or four of 30 scheduled cycling sessions. Twenty-eight mild adverse events were reported for 18 participants that were potentially related to the study. These were observed falls, complaints of soreness, muscle cramping, mild pain and fatigue, and skin rash related to the heart rate monitor. There were 30 adverse events unrelated to study procedures: illness (colds, flu), tooth loss, headache, stomach ache, tonsillectomy, and skin irritation from orthotic use.[13]

The PedsQL results are shown in Table II. One participant's data were removed from the analysis because he did not complete the questionnaire independently, leaving 57 participants (28 cycling, 29 control) in the post study analysis. The only significant between-group difference was for emotional functioning (8.9, 95% confidence interval [CI] 0.2–17.7, $p=0.046$). The responses that improved the most for this section were for questions relating to feeling scared (from 50.0–66.1) and angry (from 44.6–63.4). For both questions, these scores reflect a mean change in the answer from closest to 'sometimes' to one that was closest to 'not at all' (age 7y) or 'almost never' (age ≥8y). Significant within-group improvements were found for the cycling but not for the control group. There was a 5.8-point increase in total score (1.8–9.7; $p=0.006$). In addition, the psychosocial health summary (+6.9, 95% CI 2–12; $p=0.008$) and two of its contributing scales, emotional functioning (+9.1, 95% CI 2.4–15.9; $p=0.01$) and school functioning (+8.0, 95% CI 1.0–15.6; $p=0.038$), improved in the cycling group. The response that improved most for the school section was to a question asking if it was hard to pay attention in school. The score increased from an average of 67.0 to 77.7, reflecting a change in response from one that was closest to 'sometimes' to one that was closest to 'not at all' (7y of age) or 'almost never' (≥8y of age). Significant changes were not found for physical functioning.

Table I: Demographics and characteristics for cycling and control group participants at baseline (n = 62).[a] Adapted from Fowler et al.[13]

	Cycling (n=31)[b]	Control (n=31)[b]	p-value[c]
Demographics			
Sex: Male/Female	18/13	11/20	0.13
Age (y)	10.7 (8.5, 12.3)	11.2 (9.8, 13.3)	0.59
Ethnicity			
Hispanic	12	7	0.57
Race			
African American	5	3	0.32
White	18	15	
Asian	1	5	
Other	7	8	
Parental language			
English	24	26	0.79
Spanish	6	4	
Other	1	1	
Age categories (y)			
7–11	20	18	0.80
12–18	11	13	
Selective voluntary motor control			
Fair	17	15	0.80
Good	14	16	
Mobility			
GMFCS level I	11	8	0.31
GMFCS level II	8	6	
GMFCS level III	12	17	
Anthropometrics			
Height (m)	1.4 (1.3, 1.5)	1.4 (1.3, 1.5)	0.94
Weight (kg)	35.5 (25.4, 48.6)	36.5 (28.0, 47.2)	0.83
Related medical history[d]			
Asthma	11	6	0.25
Attention/behavioral problems	8	8	>0.99
Intellectual disability	4	4	>0.99
Seizure disorder	2	4	0.67
Learning problems	10	16	0.20
Speech problems	11	10	>0.99
Vision problems	15	23	0.07
Hearing problems	1	2	>0.99
GMFM-66 (maximum score=100)	69.2 (64.8, 74.2)	69.2 (62.2, 72.0)	0.96

[a]Values are median (25th, 75th centile) for continuous variables, frequency for categorical variables. [b]n=sample size for cycling/control groups. [c]χ^2 test for categorical variables, one-way analysis of variance for continuous variables. [d]Related medical history was obtained from the parent Pediatric Outcomes Data Collection Instrument (PODCI). GMFCS, Gross Motor Function Classification System; GMFM-66, Gross Motor Function Measure (66 items).

The PODCI results are shown in Table III. Fifty-eight parents or caregivers completed the PODCI at baseline. The response of one caregiver was removed from the analysis owing to her difficulty in comprehending some of the questions. Another was not available to complete the post intervention questionnaire, leaving 56 participants (27 cycling, 29 control) in the post study analysis. The only significant between-group finding was for the treatment expectations section. The baseline version of this section queries the parent about his or her expectations for the child's performance, e.g. 'to be able to do activities at home' or 'to be able to do more sports'. Post intervention, they were asked about changes in their child's ability, and these responses were compared with their expectations at baseline. A significant difference in

Table II: Pediatric Quality of Life Inventory SF15 (PedsQL) child responses[a]

	Cycling group (*n*=28)	Control group (*n*=29)	Between-group difference[b]	*p*-value[c]
Physical functioning				
Baseline	67.2 (57.2–77.3)	65.9 (58.2–73.5)		
Post intervention	70.4 (61.9–79.0)	69.9 (63.7–76.2)		
Within-group change[d]	3.2 (−1.9–8.2)	4.0 (−3.8–11.9)	−0.8 (−10.0–8.3)	0.85
p-value[e]	0.21	0.30		
Psychosocial health summary[f]				
Baseline	62.8 (53.8–71.8)	68.0 (61.8–74.2)		
Post intervention	69.7 (61.8–77.6)	69.5 (63.5–75.5)		
Within-group change[d]	6.9 (2.0–12.0)	1.5 (−2.0 to 5.0)	5.4 (−0.5 to 11.5)	0.07
p-value[e]	0.008[h]	0.40		
Emotional functioning				
Baseline	55.6 (44.7–66.5)	68.1 (60.2–76.0)		
Post intervention	64.7 (56.3–73.2)	68.3 (61.0–75.6)		
Within-group change[d]	9.1 (2.4–15.9)	0.2 (−5.7 to 6.1)	8.9 (0.2–17.7)	0.046[h]
p-value[e]	0.01[h]	0.94		
Social functioning				
Baseline	74.1 (63.2–85.0)	67.8 (60.2–75.5)		
Post intervention	77.1 (66.6–87.6)	71.0 (62.6–79.3)		
Within-group change[d]	3.0 (−5.2 to 11.2)	3.2 (−5.0 to 11.3)	−0.2 (−11.5 to 11.1)	0.97
p-value[e]	0.46	0.43		
School functioning				
Baseline	61.0 (50.3–71.7)	68.1 (59.9–76.3)		
Post intervention	69.0 (58.6–79.4)	69.5 (61.4–77.7)		
Within-group *change*[d]	8.0 (1.0–15.6)	1.4 (−5.9 to 8.7)	6.6 (−3.7 to 16.9)	0.20
p-value[e]	0.038[h]	0.69		
Total score[g]				
Baseline	64.2 (55.6–72.8)	67.3 (62.0–72.6)		
Post intervention	70.0 (62.3–77.6)	69.6 (64.1–75.2)		
Within-group change[d]	5.8 (1.8–9.7)	2.3 (−1.5 to 6.1)	3.5 (−2.0 to 8.8)	0.21
p-value[e]	0.006[h]	0.22		

[a]Values are mean (95% confidence interval). [b]Difference in post intervention change between the cycling and control groups. [c]*p*-value for between-group difference calculated using independent *t*-tests. [d]Post intervention change calculated by subtracting baseline value from post session value. [e]*p*-value for within-group change calculated using paired *t*-tests. [f]Psychosocial health summary is the combined scores from emotional, social and school scales. [g]Total score is the total of all scales. [h]Statistically significant at $p<0.05$.

Table III: Pediatric Outcomes Data Collection Instrument (PODCI) parent responses[a]

	Cycling group (*n*=27)	Control group (*n*=29)	Between-group difference[b]	*p*-value[c]
Global function and symptoms				
Baseline	74.8 (70.3–79.4)	75.1 (70.4–79.9)		
Post intervention	75.2 (70.4–80.1)	75.4 (70.9–79.9)		
Within-group change[d]	0.4 (−2.5 to 3.3)	0.3 (−2.8 to 3.4)	0.1 (−4.1 to 4.3)	0.96
p-value[e]	0.78	0.86		
Happiness				
Baseline	82.9 (78.0–87.8)	76.7 (69.4–84.1)		
Post intervention	86.1 (80.9–91.3)	77.4 (71.9–82.9)		
Within-group change[d]	3.2 (−2.4 to 8.8)	0.7 (−4.8 to 6.1)	2.5 (−5.1 to 10.2)	0.51
p-value[e]	0.25	0.80		
Satisfaction with symptoms				
Baseline	50.0 (36.3–63.7)	44.8 (32.5–57.1)		
Post intervention	51.9 (39.6–64.1)	32.8 (21.9–43.6)		
Within-group change[d]	1.9 (−11.3 to 15.0)	−12.0 (−23.9 to −0.3)	13.9 (−3.3 to 31.2)	0.11
p-value[e]	0.77	0.046[f]		
Treatment expectations				
Baseline	64.2 (56.4–72.0)	61.8 (55.3–68.3)		
Post intervention	62.5 (53.2–71.7)	42.4 (33.8–51.1)		
Within-group change[d]	−1.7 (−10.3 to 6.8)	−19.4 (−28.8 to −9.9)	17.7 (5.2–30.0)	0.006[f]
p-value[e]	0.68	0.0002[f]		

[a]Values are mean (95% confidence interval). [b]Difference in post intervention change between the cycling and control groups. [c]*p*-value for between-group difference calculated using independent *t*-tests. [d]Post intervention change calculated by subtracting baseline value from post session value. [e]*p*-value for within-group change calculated using paired *t*-tests. [f]Statistically significant at $p<0.05$.

change scores was found between the cycling and control groups (p=0.006) in this section. There was a significant decrease for the control group score (−19.4, 95% CI −28.8 to −9.9, p=0.002). There was little change in the cycling group (−1.7, 95% CI −10.3–6.8, p=0.68), indicating that parents' treatment expectations were not significantly different from

their child's post intervention abilities. A significant within-group difference was found for the satisfaction with symptoms section. This section contained one question: 'If your child had to spend the rest of his/her life with his/her bone and muscle condition as it is right now, how would you feel about it?' A significant before versus after decrease in the change score was found in the control group (−12.0, 95% CI −23.9 to −0.3, p=0.046). Their mean scores decreased from 44.8 to 32.8, reflecting a satisfaction level that moved further away from 'neutral' and closer to 'somewhat dissatisfied'. Significant changes were not found between or within groups for the PODCI global function and symptoms or happiness scores.

DISCUSSION

The strongest finding from this study was the significant between-group difference in the PedsQL emotional functioning scale. A more positive emotional outlook was expressed by a reduced frequency of negative feelings, especially fear and anger. An overall improvement in HRQOL was not found, as there were no between-group differences in change scores for the PedsQL total score and most subscales. As few studies have examined a cycling intervention for children with CP, and the level of evidence was low, this was designed as a phase I study. As such, we felt that it was important to critically examine within-group changes that might provide additional information and assist with the design of future research. There were significant increases in the PedsQL psychosocial health summary and the contributing emotional and school functioning scores in the cycling group. The improvement in psychosocial score of 6.9 exceeded the minimal clinically important difference (MCID) of 5.3 reported by Varni et al.[3] for the longer version of the PedsQL. The MCID has not been reported for the short form of the PedsQL. While a between-group difference was not found for the psychosocial health summary, the relatively low p-value (p=0.07) and the small overlap in the confidence intervals of the change scores for the two groups suggests that significance might have been found with a larger sample size.

Improved emotional functioning supports the positive relationship between physical fitness and psychological well-being in the general population and in children with CP following fitness exercise programs.[7–9,12] Improved emotional well-being in the present study, especially feeling less scared or angry, is consistent with the positive relationship between physical activity and reduced anxiety and depression reported for children without disability.[7] Available evidence also supports a beneficial relationship between aerobic exercise and cognition in children without disability.[22] A recent randomized controlled trial reported a positive effect of aerobic exercise on academic achievement.[23] Improved executive function, mathematics achievement, and brain activation patterns (via functional magnetic resonance imaging) were found following a 3-month aerobic exercise intervention for sedentary and overweight children (aged 7–11y). These findings are consistent with improved school functioning in cycling group participants in the present study.

Only two studies could be found that examined the PedsQL 4.0 Generic Core Scales following exercise interventions for a total of 15 children with spastic CP.[10,19] These studies used the 23-item version. Dieruf et al.[19] assessed the effect of body weight-supported treadmill training for six participants with spastic hemiplegia. While this protocol was not designed as a focused aerobic intervention, treadmill walking duration was progressively increased over a 2-week period. Four of the participants had improvements in the psychosocial health summary that exceeded the MCID of 5.3 as reported by Varni et al.[3] The results were not reported for the contributing subscale scores. Engsberg et al.[10] examined an ankle strength training intervention for nine children but also only reported the total PedsQL score. They found an increase in the total score for the parent proxy but not for the child report. It is difficult to compare the results of our study with this limited research, but one possibility is that aspects of HRQOL may improve selectively with aerobic exercise. As the PEDALS intervention incorporated strengthening and cardiorespiratory exercise components, we are unable to determine which component was critical to the observed improvement in emotional well-being.

A significant improvement was not found for the PedsQL physical functioning section. It may be that the children did not perceive or attain an increase in strength or function. While significant within-group improvements were reported for strength, gross motor function, and walking and running endurance in the cycling group, these changes were insufficient to produce a between-group difference.[13] Another possible explanation is a low level of sensitivity of these questions for children with physical disabilities. For example, the youngest children were asked how much of a problem a particular physical activity had been over the last few weeks. Their choices were 'not at all', 'sometimes', or 'a lot'. As many of the physical activities listed (walking, running, sports/exercise, picking up big things, and chores) are challenging for children with CP, an activity may remain 'sometimes' hard rather than 'not at all' hard despite an improvement. Given the chronic nature of CP, a HRQOL assessment with smaller units of change for physical health might have exhibited greater sensitivity. This issue was not problematic for psychosocial health questions as having a physical disability does not limit the range of responses possible for questions about emotional health, school, or social functioning.

While medical interventions are directed toward the child, it is primarily the parents or guardians who make the treatment decisions;[24] therefore, their perception of treatment outcomes is important. In addition, there is evidence that parents' perceptions can contribute positively or negatively to their child's well-being.[25] The similarity of the treatment expectations scores at baseline indicates that both groups had comparable positive expectations initially. The cycling group had similar scores before and after the intervention, indicating that the intervention met their expectations for improvement in function, self-image, and pain level. It is logical that control group scores decreased post intervention as their children were assigned to the 'no cycling' group. The control group

change score of −19.4 met the criteria for an MCID (13.3 medium; 21.2 large) as defined by Oeffinger et al.[26] This divergence in post intervention scores resulted in a significant difference between the two groups.

Despite an overall parental perception that there was an improvement, the cycling group did not improve significantly in the PODCI global function and symptoms scale. For this scale, parents were asked to rank their child's current physical abilities, feelings, and pain using a list of descriptors. This finding may indicate a lack of functional improvement or inadequate questionnaire sensitivity for children with physical disabilities. As previously described for the PedsQL, it is possible that, despite modest improvement, physical activities remained 'a little hard' and not 'easy' to perform. Additionally, the PODCI was designed to capture outcomes following orthopedic intervention, which is often surgical. A child's physical health may improve following a fitness intervention such as cycling yet not match the impact of surgery, which often incudes a prolonged rehabilitation period and intensive physical therapy. Therefore, the PODCI may not be the best outcome measure to examine change due to a short-term fitness intervention. Surprisingly, the PODCI satisfaction with symptoms score decreased significantly for the control group. This finding may reflect the parents' disappointment that their child was not selected for the exercise intervention. As parents did not perceive positive changes following the intervention period, their overall satisfaction with the level of their child's 'condition' may have decreased. The decrease in the satisfaction with symptoms change score (−12.0) for the control group was slightly less than a reported MCID value of 14.4.[26]

No additional studies were found that used the parent proxy version of the PODCI as an outcome measure following an exercise intervention. Two studies reported significant increases ($p<0.05$) in the global function and symptoms section following lower limb soft-tissue or bony surgery in children with CP (GMFCS level I–III).[20,27] Damiano et al.[28] reported a significant improvement in the global function and symptoms scale after musculotendinous surgery but not after selective dorsal rhizotomy or baclofen pump implantation. None of these studies reported results from the satisfaction with symptoms or treatment expectation sections, limiting comparisons with the present study.

The definition of HRQOL and how best to measure it remains unclear.[4] A wide range of questionnaires has been used for exercise research focusing on various aspects including functional ability, self-perception, feelings, and perceived competence. While many HRQOL instruments are successful in describing differences between children with and without disability, they may not be sufficiently sensitive to detect change attributable to treatment. It appears that interviewing a child directly about emotional functioning using the PedsQL SF-15 was sensitive to change following exercise in the present study. The PODCI parent proxy does not include an 'emotional functioning' section. The questions in the happiness section were worded to enquire about satisfaction rather than feelings. For example, the parent was asked about how happy his or her child has been with their physical abilities, appearance, or health in general. This section was not sensitive to change following exercise in the present study.

In a previous report, significant improvements for PEDALS study primary outcomes (walking and running endurance, gross motor function, and measures of muscle strength) were reported for the cycling but not the control group.[13] As the effects on physical functioning were not strong, this may explain limited improvements in overall HRQOL. While significant between-group differences were found for PedsQL emotional functioning and the PODCI parental treatment expectations, other sections were either not significantly different or improvements were limited to the cycling group. As previously discussed,[13] between-group differences are most easily detected when the interindividual variability is low, control group outcomes are stable, and there is a large treatment effect. Treatment dose is an important variable. Overall, the treatment adherence and intensity appeared sufficient. Adherence was high at 89.6%, protocol variations were minimal, and the mild adverse events reported did not limit cycling participation. Cycling group participants obtained an average heart rate of 147.2 beats per minute during the cardiorespiratory phase of the cycling intervention. This value represents an average of 52.2% of their maximum heart rate, calculated using the Karvonen method (correcting for differences in resting heart rate) and is within the American College of Sports Medicine threshold of 40 to 60% recommended for unfit individuals.[29] Average strength gains during the strengthening phase of each session improved from 12.2 to 29.8kg (30–74% of body weight). Exercise intensity for individual participants, however, was variable. As illustrated in case reports for two PEDALS participants,[15] motivation was not always optimal, particularly for the cardiorespiratory component.

Limitations of this study include a small sample size, a short intervention duration, intraindividual variability, and the lack of an intervention for the control group. It is difficult to recruit participants for an after-school exercise program when families of children with disabilities may already have an increased burden of care. A longer duration program might have further reduced interest in participation. Although this study was small, the Verschuren et al. study[12] is the only exercise study for children with CP to date that recruited more participants ($n=86$). That study, performed in special education schools in the Netherlands, found a between-group difference in aspects of HRQOL after 4 months of aerobic exercise. In the USA, children attend school in the 'least restrictive environment',[30] making implementation in the school system more challenging but certainly feasible. Newer technologies, such as game-based Internet programs, can be monitored remotely and could be implemented as a home exercise program to promote long-term exercise adherence. Intraindividual variability could have been addressed by using a multiple baseline design to identify participants with inconsistent responses. Control group PedsQL scores improved for all components, and the variability was large, as evidenced by the large confidence intervals for the change scores. Children with attention/behavioral problems, intellectual disabilities, and learning problems

may be less reliable for self-report outcome measurements (see Table I). The use of a no-intervention control group does not allow us to rule out alternative explanations for improvement, such as positive attention from adults.

In summary, the results of this study demonstrate the positive effects of a short-term fitness intervention on emotional well-being in children with CP but the evidence was not as strong for other aspects of HRQOL. Cycling is an exercise that can be safely performed in the clinic, community, or home. Future research may clarify the characteristics of individuals with CP who will benefit most from a cycling intervention.

ACKNOWLEDGMENTS

This study was supported by a grant from the Foundation for Physical Therapy to establish PTClinResNet: a clinical research network to evaluate the efficacy of physical therapist practice. The authors acknowledge the families and participants for their time and effort, Kara Siebert DPT, MEd, Mia Sugi DPT and Victoria Simms MPT for contributions to data collection, and the following individuals and organizations for funding, donations, or discounts: the Shapiro Family, Sidney Stern Memorial Trust, Steinmetz Foundation, Sykes Family Foundation, United Cerebral Palsy Research and Education Foundation, Biodex Inc., Freedom Concepts, Helen's Cycles, Santa Monica, National AMBUCS Inc., and Sam's Club.
Clinical Trials.gov Identifier: NCT00401154

REFERENCES

1. Vargus-Adams J. Longitudinal use of the Child Health Questionnaire in childhood cerebral palsy. *Dev Med Child Neurol* 2006; **48:** 343–7.
2. Daltroy LH, Liang MH, Fossel AH, Goldberg MJ. The POSNA pediatric musculoskeletal functional health questionnaire: report on reliability, validity, and sensitivity to change. Pediatric Outcomes Instrument Development Group. Pediatric Orthopaedic Society of North America. *J Pediatr Ortbop* 1998; **18:** 561–71.
3. Varni JW, Burwinkle TM, Seid M, Skarr D. The PedsQL 4.0 as a pediatric population health measure: feasibility, reliability, and validity. *Ambul Pediatr* 2003; **3:** 329–41.
4. Waters E, Davis E, Ronen GM, Rosenbaum P, Livingston M, Saigal S. Quality of life instruments for children and adolescents with neurodisabilities: how to choose the appropriate instrument. *Dev Med Child Neurol* 2009; **51:** 660–9.
5. Clarke SA, Eiser C. The measurement of health-related quality of life (QOL) in paediatric clinical trials: a systematic review. *Health Qual Life Outcomes* 2004; **2:** 66.
6. Rimmer JH, Chen MD, McCubbin JA, Drum C, Peterson J. Exercise intervention research on persons with disabilities: what we know and where we need to go. *Am J Phys Med Rehabil* 2010; **89:** 249–63.
7. Strong WB, Malina RM, Blimkie CJ, et al. Evidence based physical activity for school-age youth. *J Pediatr* 2005; **146:** 732–7.
8. Darrah J, Wessel J, Nearingburg P, O'Connor M. Evaluation of a community fitness program for adolescents with cerebral palsy. *Pediatr Phys Ther* 1999; **11:** 18–23.
9. Dodd KJ, Taylor NF, Graham HK. Strength training can have unexpected effects on the self-concept of children with cerebral palsy. *Pediatr Phys Ther* 2004; **16:** 99–105.
10. Engsberg JR, Ross SA, Collins DR. Increasing ankle strength to improve gait and function in children with cerebral palsy: a pilot study. *Pediatr Phys Ther* 2006; **18:** 266–75.
11. Schlough K, Nawoczenski D, Case LE, Nolan K, Wigglesworth JK. The effects of aerobic exercise on endurance, strength, function and self-perception in adolescents with spastic cerebral palsy: a report of three case studies. *Pediatr Phys Ther* 2005; **17:** 234–50.
12. Verschuren O, Ketelaar M, Gorter JW, Helders PJ, Uiterwaal CS, Takken T. Exercise training program in children and adolescents with cerebral palsy: a randomized controlled trial. *Arch Pediatr Adolesc Med* 2007; **161:** 1075–81.
13. Fowler EG, Knutson LM, DeMuth SK, et al. Pediatric endurance and limb strengthening (PEDALS) for children with cerebral palsy using stationary cycling: a randomized controlled trial. *Phys Ther* 2010; **90:** 367–81.
14. Fowler EG, Knutson LM, DeMuth SK, et al. Pediatric endurance and limb strengthening for children with cerebral palsy (PEDALS) –a randomized controlled trial protocol for a stationary cycling intervention. *BMC Pediatr* 2007; **7:** 14.
15. Siebert KL, DeMuth SK, Knutson LM, Fowler EG. Stationary cycling and children with cerebral palsy: case reports for two participants. *Phys Occup Ther Pediatr* 2010; **30:** 125–38.
16. Chan KS, Mangione-Smith R, Burwinkle TM, Rosen M, Varni JW. The PedsQL: reliability and validity of the short-form generic core scales and Asthma Module. *Med Care* 2005; **43:** 256–65.
17. Cuomo AV, Gamradt SC, Kim CO, et al. Health-related quality of life outcomes improve after multilevel surgery in ambulatory children with cerebral palsy. *J Pediatr Orthop* 2007; **27:** 653–7.
18. Davis E, Davies B, Wolfe R, et al. A randomized controlled trial of the impact of therapeutic horse riding on the quality of life, health, and function of children with cerebral palsy. *Dev Med Child Neurol* 2009; **51:** 111–9; discussion 88.
19. Dieruf K, Burtner PA, Provost B, Phillips J, Bernitsky-Beddingfield A, Sullivan KJ. A pilot study of quality of life in children with cerebral palsy after intensive body weight-supported treadmill training. *Pediatr Phys Ther* 2009; **21:** 45–52.
20. McMulkin ML, Baird GO, Gordon AB, Caskey PM, Ferguson RL. The pediatric outcomes data collection instrument detects improvements for children with ambulatory cerebral palsy after orthopaedic intervention. *J Pediatr Orthop* 2007; **27:** 1–6.
21. Varni JW, Seid M, Rode CA. The PedsQL: measurement model for the pediatric quality of life inventory. *Med Care* 1999; **37:** 126–39.
22. Hillman CH, Erickson KI, Kramer AF. Be smart, exercise your heart: exercise effects on brain and cognition. *Nat Rev Neurosci* 2008; **9:** 58–65.
23. Davis CL, Tomporowski PD, McDowell JE, et al. Exercise improves executive function and achievement and alters brain activation in overweight children: a randomized, controlled trial. *Health Psychol* 2011; **30:** 91–8.
24. Coyne I. Children's participation in consultations and decision-making at health service level: a review of the literature. *Int J Nurs Stud* 2008; **45:** 1682–9.
25. Orrell-Valente JK, Cabana MD. 'The apple doesn't fall far from the tree': the role of parents in chronic disease self-management. *Curr Opin Pediatr* 2008; **20:** 703–4.
26. Oeffinger D, Bagley A, Rogers S, et al. Outcome tools used for ambulatory children with cerebral palsy: responsiveness and minimum clinically important differences. *Dev Med Child Neurol* 2008; **50:** 918–25.
27. Lee KM, Chung CY, Park MS, et al. Level of improvement determined by PODCI is related to parental satisfaction after single-event multilevel surgery in children with cerebral palsy. *J Pediatr Orthop* 2010; **30:** 396–402.
28. Damiano DL, Gilgannon MD, Abel MF. Responsiveness and uniqueness of the pediatric outcomes data collection instrument compared to the gross motor function measure for measuring orthopaedic and neurosurgical outcomes in cerebral palsy. *J Pediatr Orthop* 2005; **25:** 641–5.
29. Mazzeo RS, Cavanagh P, Evans W, et al. ACSM Position Stand: Exercise and physical activity for older adults. *Med Sci Sports Exercise* 1998; **30:** 992.
30. Individuals With Disabilities Education Improvement Act of 2004, Pub L No. 108–446, 118 Stat. 2647. Sect. 612 (a) (5) (A) (2004).