Stationary Cycling and Children with Cerebral Palsy: Case Reports for Two Participants

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ABSTRACT. These case reports describe a stationary cycling intervention and outcomes for two child participants (P1 and P2) with spastic diplegic cerebral palsy. Each child completed a 12-week, 30-session cycling intervention consisting of strengthening and cardiorespiratory fitness phases. P1 exhibited higher training intensities, particularly during the cardiorespiratory phase. Average training heart rates were 59% and 35% of maximum heart rate for P1 and P2, respectively. Lower extremity peak knee flexor and extensor moments, gross motor function (Gross Motor Function Measure (GMFM-66)), preferred walking speed (thirty-second walk test), and walking endurance (600-yard walk-run test) were measured pre- and postintervention. Changes in outcome measurements corresponded with differences in exercise intensity. Greater gains in peak knee extensor moments, GMFM-66 scores (+4.2 versus +0.9), 600-yard walk-run test (-29% versus 0%) occurred for P1 versus P2, respectively. Preferred walking speeds did not increase substantially for P1 and decreased for P2.

KEYWORDS. Cardiorespiratory fitness, cerebral palsy, spastic diplegia, stationary cycling, strengthening

INTRODUCTION

Physical therapists and exercise specialists agree it is important to promote and maintain muscle strength and cardiorespiratory fitness (the ability for the circulatory and respiratory systems to supply oxygen to the body in response to the demands of a specified level of physical activity) in children with cerebral palsy (CP) to maximize health and function (Fowler et al., 2007b). Limited research is available that delineates the type and intensity of exercise required to improve strength and fitness for children with CP. Case reports provide valuable descriptions of individual responses to specific interventions. They allow professionals to understand the impact of patient characteristics on outcomes; thus, enhancing the quality of the physical therapy provided.

Exercise and Cerebral Palsy

Children with CP display low levels of physical fitness (Hoofwijk, Unnithan, & Bar-Or, 1995; Morgan et al., 2005). Impairments associated with CP, particularly reduced balance (Burtner, Woollacott, Craft, & Roncesvalles, 2007), limit the ability

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of a child to play or exercise at a sufficient intensity to improve fitness. Historically, effortful exercise was discouraged for children with CP because of the concern that spasticity would increase; however, research has not supported this premise (Fowler, Ho, Nwigwe, & Dorey, 2001; MacPhail & Kramer, 1995). Available evidence from a recent literature review suggests muscle strength (force-generating capacity) and cardiorespiratory fitness can be improved in children with CP utilizing a wide range of training modes, frequencies, and intensities (Verschuren, Ketelaar, Takken, Helders, & Gorter, 2008).

Stationary Cycling as an Intervention

Stationary cycling is an exercise that minimizes the need for balance and coordination providing children with CP the opportunity to exercise at a higher intensity than is otherwise possible. Adaptations can be made to the stationary cycle to accommodate individual impairments. During cycling, agonists and antagonists of the hip, knee, and ankle may be strengthened simultaneously. Research has shown that children with CP can cycle independently but demonstrate differences in muscle recruitment patterns (Johnston, Barr, & Lee, 2007; Kaplan, 1995), joint kinematics (Johnston et al., 2007), and pedal forces (Johnston, Prosser, & Lee, 2008), compared to children without CP. A recent study showed that stationary cycling could be an effective intervention for children with CP, even those who are non-ambulatory (Williams & Pountney, 2007). After a six-week, three times weekly stationary cycling intervention, these children demonstrated improved supported standing, cruising, and stepping on the Gross Motor Function Measure (GMFM) (p = 0.01). Additionally, improvements in cardiorespiratory fitness have been reported for individuals with CP following a school-based cycling intervention. Berg (1970) trained children and young adults with CP using an adapted stationary bicycle ergometer and modified positions. Exercise frequency was three times weekly for durations ranging between 1.5 to 16 months. In total, 20 of the 22 participants exhibited a 10% to 25% improvement in oxygen uptake.

Case Reports

These case reports describe the response of two children with spastic diplegic CP to a stationary cycling intervention. The children were part of a larger randomized controlled (clinical) study entitled pediatric endurance and limb strengthening (PEDALS) for children with CP (Fowler et al., 2007a). The goals of the intervention were to improve (1) muscle strength and (2) cardiorespiratory fitness. The purpose of these case reports is to illustrate individual differences in performance that are not captured in a large randomized controlled trial report. Personal and environmental factors, outside of the study inclusion and exclusion criteria, may explain these differences.

METHOD

Case Descriptions

Following an explanation of the study, participants and their legal guardians signed assent and consent forms. Both participants met the inclusion criteria: (1) spastic diplegic



CP, (2) age between 7 and 18 years, (3) ability to follow simple verbal directions, (4) Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997) Level I-III, and (5) good or fair selective voluntary motor control of at least one lower extremity as tested in sitting. Good selective voluntary motor control was defined as the ability to isolate both knee and ankle movement out of synergy (knee extension with the hip positioned in flexion, ankle dorsiflexion with the knee positioned in extension). Fair selective voluntary motor control was defined as the ability to isolate knee extension but not ankle dorsiflexion. Neither child had medical or surgical procedures within 12 months of enrollment. There were no changes in physical therapy, exercise, or sports participation for 3 months prior to enrollment, nor throughout the study. Neither child was participating in a cardiorespiratory fitness program at the time of this study.

Participant 1

Participant 1 (P1) was a 9.8-year-old female, one of the twins, delivered at 26 weeks gestation with a birth weight of 0.68 kg. Surgical history included patent ductus arteriosus repair, strabismus correction, tonsillectomy, and bilateral heel cord lengthenings. She was fully integrated in the fourth grade at a public elementary school. P1 received resource educational assistance, speech therapy, occupational therapy, and physical therapy. At the time of study enrollment, physical therapy consisted of twice-weekly lower extremity passive range of motion exercises, generalized conditioning exercises, and balance training. She walked independently without assistive devices or orthoses. She used a railing when ascending and descending stairs and had difficulty walking over uneven surfaces placing her at Level II of the GMFCS. Her height was 139.2 cm and weight was 43.8 kg. Spasticity, measured using the modified Ashworth scale (Bohannon & Smith, 1987), was: quadriceps = 1 and hamstrings = 0, bilaterally.

Participant 2

Participant 2 (P2) was an 8.5-year-old male born at 38 weeks gestation. He had a history of seizures controlled with medication and no history of surgery. P2 had a mild cognitive delay (6.5 years per parent report) and attended a school for children with special needs. His school district provided weekly physical therapy, occupational therapy, and speech therapy. Physical therapy focused on higher-level balance skills and gait training. His mother reported a history of attention and behavioral problems but he was not receiving specific treatment at the time of this study. His height was 134 cm and weight was 32.2 kg. P2 was placed at Level I of the GMFCS as he walked independently indoors and outdoors at a variety of speeds without the use of assistive devices or orthoses. He did not require a railing when ascending and descending stairs. P2 exhibited mild lower extremity spasticity: left quadriceps = 1+, right quadriceps and bilateral hamstrings = 1 on the modified Ashworth scale.

Outcome Assessment Measures

Outcome measurements of muscle strength, gross motor function, walking speed, and walking or running endurance were collected before and after the 12-week intervention



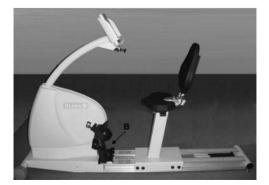
period. Strength measurements were at the body function level, while the other measures were at the activity level of the International Classification of Functioning (ICF) framework (World Health Organization, 2008). Two physical therapists administered the tests in a standardized manner as described in a manual of procedures. All study evaluators were required to demonstrate a 90% competency level for each outcome measure. They were blinded to patient assignment (cycling intervention versus control group). Five repetitions of peak knee extensor and flexor joint moments were performed using a Kin-Com dynamometer version 5.32 (Chattanooga Group, Hixson, TN) at 30, 60, and 120°/s. Verbal coaching and visual feedback provided by the computer monitor encouraged participant maximal effort. Children were assessed using dimension D (standing) and dimension E (walking, running, & jumping) of the Gross Motor Function Measure (GMFM) (Russell, Rosenbaum, Avery, & Lane, 2002). The GMFM-66 Gross Motor Ability Estimator software was used to calculate total scores. Tests of walking speed and endurance were performed in a school gymnasium using the thirty-second walk test (30sWT) (Knutson, Schimmel, & Ruff, 1999) and the 600-yard walk-run test (Fernhall et al., 1998). The 30sWT was designed to evaluate a child's ability to transition between educational activities in school. The distance traveled in the 30 s time period was measured. The 600-yard walk-run test was chosen, as it is reflective of a child's ability to keep up with their peers during sport and play activities. Participants were instructed to walk or run "as fast as you can", and the time to complete the 600-yard distance was recorded.

Stationary Bicycle Intervention

The Biodex Cyclocentric Semi-recumbent Cycle (Biodex Medical Systems, Inc., Shirley, NY) (Figure 1A) was selected for this intervention. An operation panel above the handlebars allowed exercise intensity to be adjusted in either standard

FIGURE 1. (A) The Biodex Cyclocentric Semi-recumbent Cycle chosen for this intervention. The knob located on the front panel could be pulled to release the seat. Once released, the seat was allowed to slide back along the track as the patient pushed forward on the pedal. (B) Ten tensioning cords (10 lb per cord) located at the base of the stationary bicycle provided scaled levels of loading. In this photograph, eight cords are engaged for a total load of 80 lb.

A B







"constant resistance" (level 1 to 10) or isokinetic modes. The bicycle also had a unique limb-loading feature to provide progressive resistance.

The cycling intervention was conducted in community pediatric physical therapy clinics. Each intervention physical therapist received a manual of procedures, at least one training session, and passed a competency assessment. Thirty exercise sessions occurred over a 12-week period. Resting HR was recorded prior to cycling. The stationary bicycle intervention was divided into two phases: lower extremity strengthening and cardiorespiratory fitness training.

Phase 1: Lower Extremity Strengthening

The goal of this phase was to increase hip and knee muscle strength using resistance provided by tensioning cords. The limb-loading mechanism was enabled by pulling on a lever located on the bike's front panel and releasing the seat to slide forward and backward along a linear track (Figure 1B). Once released, the tensioning cords acted to draw the seat forward and the child's push or pull on the pedals kept the seat from moving too far forward. Up to 10 cords, each providing 10 lb of resistance, could be attached. Participants were instructed to (1) hold the handles located at the sides of the seat rather than the handlebars to prevent upper extremity assistance and (2) to maintain the seat in the demarcated forward–backward position (exercise zone) while cycling.

Phase 2: Cardiorespiratory Fitness Training

Exercise intensity was measured objectively and subjectively. The threshold of maximal aerobic power is 70%-80% maximum heart rate (HRmax = 220 – age) for young adults and is described to be at least this high in children (Bar-Or & Rowland, 2004). A target HR (THR) range of 70% to 80% of age predicted HRmax was calculated for each session using the Karvonen formula (THR = $[(HRmax - resting HR) \times 0.70$ to 0.80] + resting HR) (American College of Sports Medicine, 2006). HR was monitored using an electronic pulse monitor placed on the participant's earlobe (Cat Eye Co., LTD., Boulder, CO) or chest (Polar Electro Inc., Lake Success, NY) and recorded periodically during the cycling session on a worksheet. For each session the following objective measurements were documented: "typical exercise HR" (TEHR), TEHR duration, and other representative HRs.

The Children's Effort Rating Table (CERT) (Williams, Eston, & Furlong, 1994) was used to gauge the child's perceived effort. The CERT is a 1–10 point subjective measure, with higher numbers corresponding to greater exercise effort. Children were encouraged to reflect on their breathing effort and muscle fatigue when choosing a CERT level. The most representative rating for each session was recorded.

During phase 2, the bike seat was locked, disengaging the limb-loading feature. Cycling resistance was adjusted according to each child's ability using the constant resistance mode. Therapists encouraged each participant to cycle for a minimum of 15 min. Participants were coached to reach their THR. Therapists used a variety of motivational strategies such as listening to music, imaginary play, and counting revolutions. Initial cycling duration was 15 min and was progressively increased to a maximum of 30 min. At the end of this phase, a cool-down period of pedaling without resistance was performed until the participant's heart rate decreased to within 20 beats above baseline.



P1 Performance

Initially, P1 required considerable physical assistance. Assistance was required to maintain the seat within the "exercise zone" preventing knee hyperextension, to move the foot forward at the top of the pedaling cycle, and to prevent the right hip from rotating internally during limb extension.

Load progression for phase 1 is presented in Figure 2A. During the first session, P1 cycled with one cord (10 lb) for 10 revolutions and progressed to two cords (20 lb) for 2 revolutions. During sessions two and three, training began at 10 lb and the number of revolutions at the 20 lb load was increased. During session four, she progressed to 30 lb and could cycle independently, no longer requiring physical assistance. Occasional verbal cueing was given to prevent knee hyperextension and maintain right lower extremity alignment. Sessions typically began with 20 revolutions at the previously attained load prior to progressing. Her maximum load dropped below that previously attained during sessions 13, 25, 26, and 30. The intervention therapists recorded that P1 reported feeling "tired" during these sessions and noted that she required cueing to stay focused. A maximum load of 90 lb for nine revolutions was attained during the 29th session for a total gain of 80 lb of resistance.

During phase 2, P1 was motivated to cycle at a fairly high intensity throughout the intervention. She required physical guidance to pedal and maintain right hip alignment as described for phase 1. She cycled continuously for 15 min during the first five sessions, increasing cycling duration by 1 min per session from session six through eight. A maximum duration of 23 min was achieved during session nine. Remaining session durations varied between 15 to 23 min.

HR data for P1 are presented in Figure 3A. Resting HR averaged 75 beats/min (bpm) over the 30 sessions yielding an average THR range of 170 to 184 bpm. TEHR is plotted for each session. The highest HR that exceeded TEHR for at least 1 min during a particular session, was plotted as the peak session HR. TEHR increased from 110 bpm during session one to 160 bpm by session six. Excluding the first two weeks, the intervention acclimation period, P1 exercised at an average TEHR of 58.5% of her HRmax. TEHR was within the THR range for 14 of the 30 sessions. During sessions

FIGURE 2. Load progression for phase 1 during each of the 30 sessions for (A) P1 and (B) P2. The lines represent the minimum and maximum loads at which the participant was able to cycle for at least 10 revolutions.

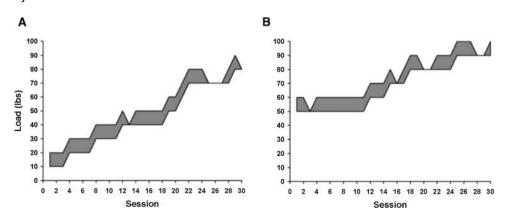
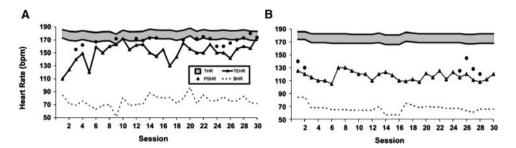




FIGURE 3. Heart rate data for phase 2 during each of the 30 sessions for (A) P1 and (B) P2. Baseline heart rate (BHR) was measured at the beginning of each session in beats/min (bpm). Using BHR, a target heart rate (THR) range of between 70% and 80% maximum was calculated. Typical exercise heart rate (TEHR) was the most representative heart rate attained during each session. Peak session heart rate (PSHR) was recorded if the subject's heart rate exceeded TEHR for at least 1 min.



5, 11, 17, and 26, when her HR dropped below previously attained levels, she reported feelings of overall fatigue. Her best performance was on the last day of the intervention when her TEHR was well within the THR range. CERT ratings generally ranged from 4 (just feeling a strain) to 6 (getting quite hard). Increased TEHR was associated with both increased and decreased CERT ratings.

P2 Performance

P2 cycled independently with good lower extremity alignment and without physical assistance. Load progression during phase 1 is presented in Figure 2B. Within the first session, he progressed to a maximum load of six cords (60 lb). He did not progress above 60 lb until session 12 despite considerable encouragement by the intervention therapist. In order to progress to a higher load, he was required to perform a minimum of 10 repetitions at 60 lb. He progressed by 10 lb during sessions 15, 18, and 25 achieving the maximum capability of the bicycle (100 lb) during session 25, for a total gain of 40 lb.

P2 did not appear motivated for the cardiorespiratory fitness phase of the intervention. Considerable positive reinforcement was required to increase exercise duration, intensity, and HR. He cycled at an inconsistent cadence, took frequent breaks, and could not be encouraged to elevate his HR near his THR range. The minimum expectation of 15 min cycling duration was not achieved until session five, increasing to 20 min for session 19. A maximum duration of 22 min was achieved during session 27. His HR data are presented in Figure 3B. TEHR decreased over the first six sessions. The highest recorded TEHR was 130 bpm during session seven. A maximum peak session HR of 145 bpm was recorded for session 26. Average TEHR, excluding the first two weeks, was 117 bpm (35% of age predicted HRmax). He did not reach his THR at any time during the intervention. CERT ratings ranged from 4 (just feeling a strain) to 7 (hard) but a level of 6 (getting quite hard) was reported for most sessions. P2 stated that he enjoyed coming to physical therapy but did not like the way "it feels" to cycle at a high rate.



TABLE 1. Peak Knee Joint Moments

		P1			P2		
	Speed (°/s)	Baseline	Follow-up	Change	Baseline	Follow-up	Change
Extensor moments (Nm)	30	37.6	45.9	8.3	31.8	38.4	6.6
	60	23.0	31.7	8.7	29.9	35.1	5.2
	120	10.9	19.5	8.6	21.7	28.5	6.8
Flexor moments (Nm)	30	16.8	12.2	-4.6	12.1	10.0	-2.1
	60	10.2	11.7	1.5	9.0	7.7	-1.3
	120	6.5	6.0	-0.5	5.1	11.0	5.9

Nm = Newton meter

RESULTS

Baseline and follow-up peak joint moments for each speed (30,60,120°/s) are shown in Table 1. Peak knee moments, identified from each group of five repetitions, were averaged bilaterally. Peak knee extensor moments increased for both children at all speeds. Improvement was greater for P1 than P2. Results for the knee flexor moments varied. P1 demonstrated a gain at 60°/s but not 30° or 120°/s. P2 exhibited an increase at 120° /s but not 30° or 60° /s.

GMFM-66 scores improved for both participants (Table 2). P1 demonstrated improvement in six items, a 5.6% increase over her baseline score. For dimension D, she obtained the maximum score at baseline for 7 of the 13 items; therefore, improvement potential existed for 6 items. Improvement was observed in three of these items: right unilateral standing with arms free, left unilateral standing with arms free, and transitioning from right high kneeling to standing. For dimension E, improvement was possible in 7 of the 24 items. She improved in three: stepping over a stick with her left foot, vertical jumping, and hopping on her right foot.

P2 improved his performance on the GMFM-66 for three items, a 1.3% increase over his baseline score. The improvement potential was the same as for P1 in dimension D (six items); however, this dimension score was unchanged. P2 had the opportunity to improve the score of nine items in dimension E. Improvement occurred for three items: stepping over a stick with the left foot, stepping over a stick with the right foot, and stair locomotion (alternating his lower extremities without use of a railing).

TABLE 2. Gross Motor Function, Walking and Running Endurance Tests

	F	21	P2		
	Baseline	Follow-up	Baseline	Follow-up	
GMFM-66	72.6	76.8	71.7	72.6	
30sWT (feet)	127.7	127.9	146.7	127.5	
600-yard walk run (min)	6.8	4.8	4.8	4.8	

GMFM-66 = Gross Motor Function Measure (66 items), 30sWT = thirty-second walk test.



Results of the timed walking and running tests are shown in Table 2. Distance covered in the 30sWT was essentially the same following the intervention for P1 and decreased for P2. Both participants were able to complete the full 600-yard distance during preand post-evaluations. P1 decreased her time by 2.1 min, a 29% improvement while P2 completed this distance in the same time.

DISCUSSION

The mechanisms connected with response to therapy are multifaceted and driven by factors associated with the individual and the therapeutic intervention. Exercise intensity is an important parameter governing intervention effectiveness. Intensity measures are not always reported for rehabilitation research (Fowler et al., 2007b). Using the case report format, we were able to quantify individual differences in exercise intensity, as reflected by load progression and HRmax.

Of the two participants, P1 demonstrated a higher training intensity, shown by a greater improvement in load progression. This corresponded with a greater increase in peak knee extensor moments across all speeds (30°, 60°, and 120°/s). In contrast, a relationship between load progression and peak knee flexor moments was not found for either participant. Changes in knee flexor strength were less than those found for the knee extensors and were inconsistent with both increases and decreases occurring. In general, PEDALS participants exhibited greater difficulty performing knee flexion as compared to knee extension during testing, particularly at higher speeds of movement. Others have reported greater variability (Ayalon, Ben-Sira, Hutzler, & Gilad, 2000) and less reliability (Berry, Giuliani, & Damiano, 2004) for testing of knee flexors as compared to extensors.

Functional strength, reflected by the GMFM-66, improved in both children. A ceiling effect was not observed. Consistent with increased gains in training load and knee extensor strength, more improvement (4.2 points) was seen in the GMFM for P1 compared to P2 (0.9 points). These improvements represent clinically relevant changes as an increase of 1.25 points on the GMFM is considered a large effect size, and an increase of 0.78 points is considered a medium effect size (Oeffinger et al., 2008). Test items within both dimensions (transitioning from high kneeling to standing, unilateral stance, stepping, jumping, and hopping) improved and were consistent with gains in knee extensor strength. Williams and Pountney (2007) found a greater increase on the GMFM (mean = 7.3% increase, p < .05) following a stationary cycling intervention for 11 non-ambulatory children with CP. They attributed the large gain to pre-existing low levels of functional ability and few opportunities to participate in physical activity.

Improvements in walking and running endurance may be linked to exercise intensity during phase 2 on the basis of HR data. While P1 did not consistently reach her THR, her average TEHR was fairly high (59% of maximum HR). Post-intervention, P1 improved her walking/running endurance substantially during the 600-yard walk run (a 29%) decrease in time). At baseline, P1 was considerably slower compared to P2 (80 versus 114 m/min, respectively); however, their speed was identical at follow-up (116 m/min). This post-intervention speed was half of that reported for children without disability of similar age (232 m/min) (Vodak & Wilmore, 1975).

Preferred walking speed (30sWT) did not improve substantially for P1 and declined for P2. Baseline distance (and concomitantly, walking speeds) for the test were already quite high at 79% (P1) and 90% (P2) of that reported for children without disability of



similar ages (Knutson et al., 1999). Failure to increase the distance covered in 30 s may indicate the intervention was not task specific for preferred walking speed or that this speed is intrinsic to the individual and somewhat resistant to change.

CERT levels reported by both participants were similar despite differences in exercise intensity based on HR and varied little across the 30 sessions. Few studies have examined the use of perceived exertion ratings during exercise or testing for children with CP. Johnston et al. (2007) used a similar measure of perceived effort and found no differences between adolescents with and without CP during recumbent stationary cycling despite dissimilarity of muscle activity patterns and cycling efficiency. Perceived exertion ratings may be more useful for monitoring individual responses rather than as a proxy measure for HR data.

Although both children willingly attended exercise sessions and appeared to enjoy their contact with the intervention physical therapists, motivation to work at a high intensity differed considerably. While we did not include a formal measure of motivation in this study, subjective feedback from the patients and the intervention therapists suggests that differences in motivation impacted individual performance. Bartlett and Palisano (2000) proposed a multivariate model of determinants of motor change for children with CP that included primary impairments, secondary impairments, inherent child characteristics, family ecology, and health care services. This model includes factors of cognitive impairment and motivation that may play an important role, particularly for interventions calling for a high level of focus and motivation. "Mastery motivation" (Bartlett & Palisano, 2000; Hauser-Cram et al., 2001) is when a child attempts a task independently in a focused and persistent manner to solve a problem or master a skill or task that is at least moderately challenging. This type of motivation is based on an intrinsic feeling of efficacy rather than an extrinsic reward. Research has shown that children (10 months to 12 years) with motor impairment and developmental delay who exhibit higher mastery motivation scores also demonstrated higher mental age scores, social skills, and daily living skills (Hauser-Cram et al., 2001). Children with behavioral problems demonstrated lower mental age scores, social skills, and daily living skills. In contrast to adults, a child's motivation to fully participate in physical therapy cannot be assumed, as a parent or healthcare professional typically initiates or refers the child to physical therapy.

Initially, P2 cycled independently with greater force-generating capacity during phase 1. He did not advance at an expected rate of load progression given his initial physical competence. This portion of the intervention was more challenging for P1 than P2. P1 initially required physical assistance for cycling and began phase 1 with a lower level of resistance. Despite these limitations, she steadily progressed in her ability to cycle independently and increase repetitions and load. At the intervention's end, her maximum load was only 10 lbs less than that of P2.

During phase 2, differences in exercise intensity were more pronounced. P1 increased her ability to cycle at higher TEHRs while P2 maintained a low TEHR throughout the 30 sessions. Attention and behavioral problems, reported for P2, may have contributed to his apparent poor motivation. Extrinsic motivation was provided in the form of adapted overground large tricycles, offered to all who completed the study. These tricycles were commercially available and modified for P1 by adding strapped pedals. P1 expressed enthusiasm about the prospect of having her own tricycle. P2 had the skill level necessary to cycle independently at the beginning of the study; therefore, the incentive of a new tricycle may not have been as rewarding to him.



An alternative mode of exercise may have been more successful in producing a physiological benefit for P2. For example, high intensity exercise of short duration (anaerobic) is more typical of daily childhood physical activity (Bar-Or & Rowland, 2004). Two recent studies have examined protocols that include interval training or anaerobic exercise for children with CP. Unnithan et al. (2007) performed a 12-week, uphill walking, interval training intervention for seven children and found improvements in aerobic capacity and gross motor function. Verschuren et al. (2007) included anaerobic exercise as part of an eight-month school-based exercise RCT. A significant improvement in aerobic/anaerobic capacity and gross motor function was found for the intervention as compared to the control group. Modifying the PEDALS' protocol to include anaerobic intervals might have been a better match for P2's motivation and attention.

We chose stationary cycling with extrinsic motivation provided by the physical therapist. Alternative strategies, such as interactive video screens operated by increasing exercise intensity, may provide intrinsic motivation or distract a child from feelings of exertion and might be more effective. Overground cycling with variant terrain may be more motivating and uphill cycling would provide a functional reason to increase physical exertion.

Both children were provided with overground tricycles at the end of this study for continued exercise and recreation. Following the study, P1 reported riding her bicycle on a regular basis and her entire family joined a community gym program. Additionally, her mother reported that, for the first time, P1 spent all day at an amusement park without requiring a wheelchair. P2's mother reported that he did not ride his bicycle regularly after the study's completion due to parental time constraints.

CONCLUSION

The information gleaned from these case reports suggests that a stationary cycling intervention has the potential to improve lower extremity muscle strength, gross motor function, and walking endurance in children with CP. The relationship between training intensity and improvement in relevant outcomes was strong in these two participants. The detailed description of the participants' individual characteristics and their different responses to training illustrates the complex nature of therapeutic interventions and may assist therapists in identifying factors to improve outcomes for individual children. Intellectual ability, attention deficits, and behavior problems appeared to be barriers for performance, particularly for the cardiorespiratory fitness phase, which required considerable effort and motivation.

The role of a pediatric physical therapist includes promoting lifelong fitness for children with CP and other disabilities (Campbell, 1997; Fowler et al., 2007b). A goal of the PEDALS intervention was instructing children with CP in a fitness program that could be maintained independently using the bicycle provided at the study's end. We prepared the children to transition from the therapy intervention to a home program by teaching them about appropriate exercise elements such as intensity progression, duration, warm up, and cool down. These strategies could be incorporated into a variety of other exercise modes, sports and settings throughout their lifetime. Further research is necessary to identify successful strategies for individuals with diagnoses such as CP where the population is extremely heterogeneous.



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REFERENCES

- American College of Sports Medicine (ACSM). (2006). *Guidelines for exercise testing and prescription* (7th ed.). Philadelphia: Lippincott, Williams & Wilkins.
- Ayalon, M., Ben-Sira, D., Hutzler, Y., & Gilad, T. (2000). Reliability of isokinetic strength measurements of the knee in children with cerebral palsy. *Developmental Medicine & Child Neurology*, 42, 398–402.
- Bar-Or, O., & Rowland, T. W. (2004). Pediatric exercise medicine: From physiologic principles to health care applications. Champaign, IL: Human Kinetics.
- Bartlett, D. J., & Palisano, R. J. (2000). A multivariate model of determinants of motor change for children with cerebral palsy. *Physical Therapy*, 80, 598–614.
- Berg, K. (1970). Effect of physical training of school children with cerebral palsy. *Acta Paediatrica Scandinavica Supplement*, 204, 27–33.
- Berry, E. T., Giuliani, C. A., & Damiano, D. L. (2004). Intrasession and intersession reliability of handheld dynamometry in children with cerebral palsy. *Pediatric Physical Therapy*, 16, 191– 198.
- Bohannon, R. W., & Smith, M. B. (1987). Interrater reliability of a modified Ashworth scale of muscle spasticity. *Physical Therapy*, 67, 206–207.
- Burtner, P. A., Woollacott, M. H., Craft, G. L., & Roncesvalles, M. N. (2007). The capacity to adapt to changing balance threats: A comparison of children with cerebral palsy and typically developing children. *Developmental Neurorehabilitation*, 10, 249–260.
- Campbell, S. K. (1997). Therapy programs for children that last a lifetime. *Physical & Occupational Therapy in Pediatrics*, 17, 1014.
- Fernhall, B., Pitetti, K. H., Vukovich, M. D., Stubbs, N., Hensen, T., Winnick, J. P., et al. (1998). Validation of cardiovascular fitness field tests in children with mental retardation. *American Journal of Mental Retardation*, 102, 602–612.
- Fowler, E. G., Ho, T. W., Nwigwe, A. I., & Dorey, F. J. (2001). The effect of quadriceps femoris muscle strengthening exercises on spasticity in children with cerebral palsy. *Physical Therapy*, 81, 1215–1223.
- Fowler, E. G., Knutson, L. M., DeMuth, S. K., Sugi, M., Siebert, K., Simms, V., et al. (2007a). Pediatric endurance and limb strengthening for children with cerebral palsy (PEDALS)-A randomized controlled trial protocol for a stationary cycling intervention. *BioMed Central Pediatrics*, 7. doi: 10.1186/1471-2431-7-14.
- Fowler, E. G., Kolobe, T. H., Damiano, D. L., Thorpe, D. E., Morgan, D. W., Brunstrom, J. E., et al. (2007b). Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: Section on Pediatrics research summit proceedings. *Physical Therapy*, 87, 1495–1510.
- Hauser-Cram, P., Warfield, M. E., Shonkoff, J. P., Krauss, M. W., Sayer, A., & Upshur, C. C. (2001). Children with disabilities: A longitudinal study of child development and parent well-being. *Monographs of the Society for Research in Child Development*, 66, i–viii, 1–114; discussion 115–126.
- Hoofwijk, M., Unnithan, V., & Bar-Or, O. (1995). Maximal treadmill performance in children with cerebral palsy. *Pediatric Exercise Science*, 7, 305–313.
- Johnston, T. E., Barr, A. E., & Lee, S. C. (2007). Biomechanics of submaximal recumbent cycling in adolescents with and without cerebral palsy. *Physical Therapy*, 87, 572–585.
- Johnston, T. E., Prosser, L. A., & Lee, S. C. (2008). Differences in pedal forces during recumbent cycling in adolescents with and without cerebral palsy. *Clinical Biomechanics (Bristol, Avon, UK)*, 23, 248–251.
- Kaplan, S. L. (1995). Cycling patterns in children with cerebral palsy. Developmental Medicine & Child Neurology, 37, 620–630.



- Knutson, L. M., Schimmel, P. A., & Ruff, A. (1999). Standard task measurement for mobility: Thirty-second walk test. Pediatric Physical Therapy, 11, 183-190.
- MacPhail, H. E., & Kramer, J. F. (1995). Effect of isokinetic strength-training on functional ability and walking efficiency in adolescents with cerebral palsy. Developmental Medicine & Child Neurology, *37*, 763–775.
- Morgan, D., Keefer, D. J., Tseh, W., Caputo, J. L., Craig, I., Griffith, G., et al. (2005). Walking energy use in children with spastic hemiplegia. *Pediatric Exercise Science*, 17, 91–92.
- Oeffinger, D., Bagley, A., Rogers, S., Gorton, G., Kryscio, R., Abel, M., et al. (2008). Outcome tools used for ambulatory children with cerebral palsy: Responsiveness and minimum clinically important differences. Developmental Medicine & Child Neurology, 50, 918–925.
- Palisano, R., Rosenbaum, P., Walter, S., Russell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. Developmental Medicine & Child Neurology, 39, 214–223.
- Russell, D. J., Rosenbaum, P. L., Avery, L. M., & Lane, M. (2002). Gross motor function measure (GMFM-66 & GMFM-88) user's manual. London: Mac Keith Press.
- Unnithan, V. B., Katsimanis, G., Evangelinou, C., Kosmas, C., Kandrali, I., & Kellis, E. (2007). Effect of strength and aerobic training in children with cerebral palsy. Medicine & Science in Sports & Exercise, 39, 1902–1909.
- Verschuren, O., Ketelaar, M., Gorter, J. W., Helders, P. J., Uiterwaal, C. S., & Takken, T. (2007). Exercise training program in children and adolescents with cerebral palsy: A randomized controlled trial. Archives of Pediatrics & Adolescent Medicine, 161, 1075–1081.
- Verschuren, O., Ketelaar, M., Takken, T., Helders, P. J., & Gorter, J. W. (2008). Exercise programs for children with cerebral palsy: A systematic review of the literature. American Journal of Physical Medicine & Rehabilitation, 87, 404–417.
- Vodak, P. A., & Wilmore, J. H. (1975). Validity of the 6-minute jog-walk and the 600-yard run-walk in estimating endurance capacity in boys, 9–12 years of age. Research Quarterly, 46, 230–234.
- Williams, J. G., Eston, R., & Furlong, B. (1994). CERT: A perceived exertion scale for young children. Perception & Motor Skills, 79, 1451–1458.
- Williams, H., & Pountney, T. (2007). Effects of a static bicycling programme on the functional ability of young people with cerebral palsy who are non-ambulant. Developmental Medicine & Child Neurology, 49, 522-527.
- World Health Organization. (2008). International classification of functioning, disability and health (ICF). Geneva, Switzerland: WHO. Retrieved October 28, 2008, from http://www.who.int/ classification/icf

