An innovative cycling exergame to promote cardiovascular fitness and health-related quality of life in youth with cerebral palsy

Shannon Knights¹, Nicholas Graham², Lauren Switzer¹, Hamilton Hernandez², Zi Ye², Briar Findlay¹, Wen Xie¹, Virginia Wright¹, Darcy Fehlings¹

¹ Bloorview Research Institute, Holland Bloorview Kids Rehabilitation Hospital, Toronto, Canada
² School of Computing, Queen’s University, Kingston, Canada

Corresponding Author:
Dr. Darcy L Fehlings
Holland Bloorview Kids Rehabilitation Hospital
150 Kilgour Road
Toronto, Ontario, Canada, M4G 1R8
Tel: (416) 425-6220 ext. 3586
Fax: 416-424-3837
dfehlings@hollandbloorview.ca

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Abstract

Objective: To evaluate the effects of an internet-platform exergame cycling programme on cardiovascular fitness and health-related quality of life (HQL) of youth with cerebral palsy (CP).

Methods: In this prospective case series, eight youth with bilateral spastic CP, Gross Motor Functional Classification System (GMFCS) level III, completed a six-week exergame programme. Outcomes were obtained at baseline and post-intervention. Primary outcome measures were the GMFCS III-specific shuttle run test (SRT-III) (for cardiovascular fitness) and the ‘Physical Activities and Health’ domain of the KIDSCREEN-52 questionnaire (for HQL). Secondary outcomes included the 6-Minute Walk Test, Wingate Arm Cranking Test, and anthropomorphic measurements.

Results: There were significant improvements in the SRT-III \( t=-2.5, p=0.04, d=0.88 \) and the ‘Physical Activities and Health’ domain of the KIDSCREEN-52 \( t=-2.8, p=0.03, d=1 \) post-intervention. There were no significant changes in secondary outcomes.

Conclusion: Measures of cardiovascular fitness and physical HQL in youth with CP improved following an exergame cycling programme.
1. Introduction

Children with cerebral palsy (CP) are at risk for a decrease in gross motor function throughout adolescence [1]. This decline is functionally significant for children classified at Gross Motor Function Classification System (GMFCS) [2] level III who may transition from using a walker to a wheelchair as their primary form of mobility. Many factors contribute to the loss of physical capabilities in adolescents with GMFCS III CP, including proximal muscle weakness secondary to disuse, poor cardiovascular fitness, changes in body composition, and limitations in range of motion [1]. Cardiovascular fitness is a potentially modifiable factor that can be enhanced through participation in physical activities. However, adolescents with CP participate in less physical activity than their peers who do not have disabilities and their level of participation often declines further with age[3]. Youth with CP also have lower levels of social interaction than their typically developing peers and can experience a decrease in social participation as they get older [4].

Exergames, a type of interactive computer play, are emerging as a novel approach to address the combined challenges of declining fitness and social isolation. Exergames are videogames that require physical activity and can offer opportunities for social interaction through multiplayer use. The use of in-home gaming units can eliminate accessibility barriers to participation in physical and social activities. Commercially available exergames such as Konami's Dance Dance Revolution have been shown to improve aerobic fitness in overweight children without physical disabilities [5]. Studies have shown that youth with CP can achieve moderate levels of energy expenditure while playing exergames [6,7]. However, there is limited knowledge about whether this translates into changes in cardiovascular fitness. One study showed improvement in the
Bruce Treadmill Test, a measure of cardiac fitness, in youth with ambulatory CP (GMFCS I and II) after a 20-week interactive computer play intervention [8]. There is limited evidence for individuals with GMFCS level III CP. Exergames have been shown to be enjoyable [6] and to increase levels of participation, motivation, and satisfaction [9] in youth with CP. Little is known about the relationship between exergames and health-related quality of life (HQL).

The primary study objective was to determine the effects of an innovative exergame cycling programme on cardiovascular fitness and physical HQL (physical well-being associated with activity) of youth with GMFCS level III CP. We hypothesized that participation in a six-week exergame intervention would be associated with improvements in both areas. Secondary objectives were to evaluate the effects of the exergame intervention on exercise tolerance, anaerobic power, anthropomorphic measurements, and other domains of HQL.

2. Methods
Participants
Participants were a voluntary convenience sample of eight children with bilateral spastic CP. Inclusion criteria were: age 9 to 18 years; GMFCS level III; high-speed internet in the home; and ability to operate a hand-held videogame controller. Exclusion criteria were: orthopedic surgery in the preceding six months and exercise-induced asthma. Eleven youth were identified through outpatient clinics at a tertiary rehabilitation hospital in Toronto, Canada and consecutively approached about study participation.

Study design
In this prospective case series, participants completed a six-week home-based internet-platform exergame cycling programme. Assessments were performed at baseline and two days post-intervention. To control for external variables, all assessments were performed at the same time of day (mid-day on a weekend) and in the same environment (a gymnasium at a rehabilitation hospital). Ethical approval was granted by the Holland Bloorview and Queen’s University Health Science Research Ethics Boards. All participants and their caregivers provided written informed consent.

Intervention

The participants were fit with customized recumbent bicycle units that had been previously developed through participatory design sessions with youth with CP [10]. Each cycling unit consisted of a PCGamerBike Mini (3D Innovations, Edmundston, Canada) attached to a specialized seat with a seatbelt and lateral supports, connected to a Toshiba DX730 computer (figure 1). Each unit required 2 x 4 feet of floor space and cost approximately $2500. Six different multiplayer exergames were designed for this intervention [10]. Game avatars were powered by the participants’ cycling. Hand-held videogame controllers were programmed using two simple functions to reduce fine motor demands. Participants wore headsets to communicate with each other during game play using TeamSpeak voice-over IP software. Participants wore armband heart rate (HR) monitors with Bluetooth technology (IMPACT Sports Technologies, San Diego, USA). All game play was monitored by a research team member to address technical concerns and to engage in multiplayer play if only one participant was present.

Insert figure 1 about here
Each participant’s HR and number of minutes playing the exergames were monitored. Target HR was defined as 40-60% of HR reserve. HR reserve was calculated as [desired percentage (40-60%) x (HRmax – HRrest)] + HRrest, where HRmax was set at 194 bpm [11] and HRrest was the resting HR of each participant. HR reserve is the preferred method for exercise prescription in children with CP because it includes the resting HR, which reflects an individual’s level of cardiovascular fitness [12]. Participants were asked to play for at least 30 minutes per day a minimum of three days per week and to achieve their target HR for at least 60 minutes per week. These guidelines were based on the American College of Sports Medicine recommendations for cardiorespiratory fitness for individuals with CP [13]. The exergames included incentives for reaching target HR, such as additional ‘power-ups’. Cues on the computer screen (eg. a heart that turned red when in the target HR zone) provided visual feedback. Games were available over an internet platform during specified hour and a half time slots six days per week to encourage multiplayer activity. Participants could choose which days to play. A research assistant provided individual weekly exercise coaching sessions (over the phone or in-person) to review the participants’ performance over the previous week and to encourage them to achieve their goals. The research assistant also screened for adverse events including knee pain, falls, and skin breakdown.

Halfway through the intervention, several participants were noted to be playing the exergames for significantly longer than the recommended playtime. Due to concerns about possible over-exertion or injury, limits were introduced to restrict the amount of playtime. Participants were allowed to play for a maximum of 60 minutes per day, including a maximum of 30 minutes per
day above target HR. The exergames were unavailable for three days in week four while these limits were integrated into the software.

Co-primary outcome measures

The primary outcome measure for cardiovascular fitness was the GMFCS level III-specific shuttle run test (SRT-III) [14]. Using their regular assistive devices, participants walked or ran a distance of 7.5m at increasingly faster speeds as determined by an auditory signal. The starting speed was 1.5km/hour with a graded increase of 0.19km/hour per minute. Participants wore heart rate monitors during the SRT-III. A peak HR of at least 180 bpm during the SRT-III is recommended as an indicator of maximal effort [14]. The test was stopped when the participants did not reach the 7.5m marker on two consecutive paced signals, regardless of the peak HR achieved. The highest half-level completed in the allotted time was recorded, with higher levels indicating better cardiovascular fitness. The SRT-III has been shown to be a reliable measure of cardiorespiratory fitness in youth with CP classified as GMFCS level III. The standard error of measurement is 0.48 levels and the smallest detectable change is 1.32 levels [14].

The primary outcome measure for physical HQL was the ‘Physical Activities and Health’ domain of the KIDSCREEN-52 questionnaire. The KIDSCREEN-52 is a self-report generic HQL measure for youth ages 8-18 years. Fifty-two items are divided into 10 domains. The ‘Physical Activities and Health’ domain includes five items about physical well-being, energy and fitness. Higher scores on the KIDSCREEN-52 indicate better HQL. The reliability of the KIDSCREEN-52 has been established in a large cross-cultural study [15]. Participants completed
the KIDSCREEN-52 independently or with a research assistant to transcribe the participant’s answers. All participants were able to perform all outcome measures.

Secondary outcome measures

The 6-Minute Walk Test is a reliable measure of exercise tolerance for ambulation in the community, with a minimum detectable change of 47.4m for children with CP in GMFCS level III [16].

The Wingate Arm Cranking Test is a reliable measure of anaerobic power for children with CP [17]. The test was performed while seated with the ergometer at shoulder height. Load was determined based on each participant’s height, weight, and a torque factor of 0.26. Participants completed a two-minute habituation phase, followed immediately by a 30-second maximal effort phase. Mean power was the outcome.

Anthropomorphic measurements were obtained to assess adiposity. Waist circumference was measured between the iliac crests and inferior rib cage using a flexible tape measure [18]. Triceps skinfold thickness was measured at the midpoint between the acromion and olecranon using a caliper [18].

Five other relevant domains (‘Feelings’, ‘Mood and Emotions’, ‘Self-Perception’, ‘Autonomy’ and ‘Peers’) of the KIDSCREEN-52 Questionnaire were also included as secondary outcome measures.
Statistical analyses

Descriptive statistics were calculated for all outcome variables. Paired t-tests were performed to assess pre- and post-intervention changes in outcome measures. A $p$-value of 0.05 was set as the level of statistical significance for both primary outcome measures. Bonferroni corrections were applied to the multiple secondary outcomes with an adjusted $p$-value of 0.006. Cohen’s $d$ was calculated to measure effect size, with $d$ values of $\leq 0.20$, $0.50$, and $\geq 0.80$ considered as small, medium, and large respectively [19].

3. Results

Demographic information

Three of 11 eligible youth declined study participation, giving a recruitment rate of 73%. Eight youth (six males, two females; mean age 14.3 SD 2.5 years; range 9-18 years) with bilateral spastic CP classified at GMFCS level III participated in the study. Body Mass Index (BMI), Manual Ability Classification System (MACS) [20] and Gross Motor Function Measure (GMFM-66) scores [21] were assessed at baseline (table 1). Mean waist circumference and triceps skinfold thickness were 83.1 SD 11.9 cm and 27.8 SD 14.2 mm at baseline, respectively. One participant left on a family vacation after four weeks and four days and her post-intervention outcomes were obtained at that time. The other participants all completed the six-week exergame cycling programme. One participant had mild skin breakdown from his knees rubbing together while cycling. His bicycle was modified with a pummel to address this problem. None of the participants reported knee pain or falls from the equipment.

*Insert table 1 about here*
Heart rate and playtime

The mean weekly amount of playtime per participant was 202 SD 95 minutes, with 79 SD 48 minutes (39% of playtime) above target HR. Individual and group results are shown in table 1. Seven individual participants achieved the goal for mean weekly playtime and five achieved the goal for mean playtime above target HR. Mean weekly playtime and time above target HR graphed across the six weeks are shown in figure 2. Apart from week four, the goal of 60 minutes of mean weekly playtime above target HR was exceeded each week. Percentage of total playtime at different levels of HR reserve is shown in figure 3.

*Insert tables 2 and 3 about here*

Co-primary outcomes

Following the exergame intervention, there was a significant improvement in the mean level achieved on the SRT-III, from 4.8 SD 4.6 to 6.4 SD 5.0 ($t=-2.5$, $p=0.04$, $d=0.88$). The mean change in level was 1.7 SD 1.9. On an individual level, one participant showed no change on the SRT-III and the remaining participants showed improvement ranging from 0.5 to 5 levels (table 1). There were no significant correlations between changes in SRT-III performance and weekly playtime, playtime above target HR, BMI percentile, MACS level, or GMFM-66 score. Peak HR during the SRT-III is shown in table 1. GMFM-66 scores were correlated with peak HR during the baseline SRT-III (Pearson’s $r=0.74$, $p=0.04$). The mean score on the ‘Physical Activities and Health’ domain of the KIDSCREEN-52 questionnaire increased from 48.5 SD 5.8 to 58.1 SD
11.6 ($t=-2.8$, $p=0.03$, $d=1$). The mean change in score was 9.6 SD 9.6 points, ranging from -5.4 to 26.1.

Secondary outcomes
There were no significant changes in secondary outcomes following the intervention: 6-Minute Walk Test ($p=0.52$), Wingate Arm Cranking Test ($p=0.21$), ‘Feelings’ domain of the KIDSCREEN-52 ($p=0.31$), ‘Mood and Emotions’ domain ($p=0.20$), ‘Self-Perception’ domain ($p=0.12$), ‘Autonomy’ domain ($p=0.36$), ‘Peers’ domain ($p=0.77$), and triceps skinfold thickness ($p=0.89$). There was a decrease in waist circumference with a mean change of -3.9 SD 3.9 cm, but the reduction was not statistically significant ($p=0.03$).

Enjoyment
In rating their enjoyment of the exergames on a scale from one (lowest) to ten (highest), the participants’ mean scores ranged from 6.3 to 8.8 for the six individual games. Informal comments during exergame sessions included: ‘I haven't had this [much] fun in years’ and ‘I can't wait until after school so I can play’. Several participants expressed interest in the social aspect and in continuing with the exergame program at the completion of the study.

4. Discussion
This study demonstrates that a six-week exergame cycling intervention is associated with improvements in measures of cardiovascular fitness and physical HQL in youth with CP classified at GMFCS level III. This has important treatment implications. Exergames may be an
important component to add to the rehabilitation tool kit. They have the potential to improve fitness in a manner that is fun and enhances adherence.

The mean level achieved on the SRT-III increased by 1.7 levels, which represents a large effect size and exceeds the minimum detectable change of 1.32 levels. While this change is clinically significant at a group level, there was wide individual variability in changes on the SRT-III post-intervention. Improvements in SRT-III performance were not explained by weekly playtime, playtime above target HR, BMI percentile, MACS level, or GMFM-66 score. It is notable that the improvement was observed after a short six-week exergame cycling intervention, compared with other interventions lasting up to 20 weeks [8].

Many of the participants did not achieve a peak HR of 180 bpm during the SRT-III, as recommended in the test protocol [14]. Lower peak HR during the SRT-III was highly correlated with lower GMFM-66 score, suggesting that the SRT-III performance of some participants may have been limited more by gross motor ability and less by cardiovascular exertion. Given these limitations in peak HR, improvement in SRT-III performance may be interpreted as either an improvement in cardiovascular fitness or better exercise tolerance. The SRT-III has been shown to have excellent test-retest reliability with intra-class correlation coefficients of 0.98[14]; therefore, it is unlikely that the significant change in this measure was due to a learning effect. Inclusion of additional objective measurements of cardiovascular fitness such as maximal oxygen consumption will be helpful in future research.
On average, participants exceeded the goals for weekly playtime and target HR. To improve aerobic capacity, the American College of Sports Medicine recommends exercise at 40-85% of HR reserve for 20-40 minutes three to five days per week. This corresponds to moderate to vigorous intensity exercise with metabolic equivalents for task (METs) of 5-10 [22]. Our innovative exergame cycling system allowed participants to achieve the recommended elevations in HR. This level of physical activity has been shown to promote cardiovascular fitness when sustained over time [22]. As seen in figure 3, most of the participants’ time above target HR occurred between 40-60% of HR reserve, with very little playtime above 60% HR reserve. The exergame intervention yielded moderate intensity aerobic activity but minimal vigorous intensity activity.

There was an improvement in the ‘Physical Activities and Health’ domain of the KIDSCREEN-52 questionnaire following the exergame programme, with a large effect size. This highlights the established relationship between physical activity and HQL [23]. The relationship between physical activity and psychosocial functioning is less well understood. One randomized controlled trial of a stationary cycling intervention for youth with CP showed a positive influence on emotional functioning in the intervention group [24]. Despite reported enjoyment and opportunities for social interaction, our study did not yield significant changes in the KIDSCREEN-52 domains related to emotional or social HQL.

This study shows that exergames can be successfully and safely adapted for use in children with GMFCS level III CP. Since youth with GMFCS III CP have more difficulty playing commercial
exergames (e.g. Wii Sports) that require prolonged standing or balance, recumbent cycling-based exergames may be better suited to this group.

Previous studies of exergames have shown a substantial decline in interest over time [25,26]. In contrast, our study showed sustained participation over the course of the six-week intervention. This may be explained by several factors. The six exergames were intentionally introduced at different times during the intervention to maintain interest and novelty. Furthermore, the youth were engaged in social interaction through multiplayer game play. Group play has been shown to increase motivation to participate in exergames [27]. The participants also had the support of an exercise coach who contacted them weekly to encourage them to achieve their playtime and HR targets. During weeks four to six, participants were asked to limit their total playtime due to concerns about possible over-exertion. This likely contributed to the lower mean amount of playtime in week four when the playtime limits were being modified and the games were unavailable for three days (see figure 2).

There were no significant changes in secondary outcome measures. There was a mean decrease in waist circumference of 3.9 cm but this did not reach statistical significance, likely due to our small sample size. Waist circumference is associated with obesity in children [18], suggesting that exergames may play a role in helping to prevent or manage weight gain. The 6-minute walk test, a measure of exercise tolerance, did not change. This outcome measure has been studied in other exergame interventions with both positive [28] and negative [8] results. Anaerobic power, as measured by the Wingate arm cranking test, did not improve. In future, adding anaerobic training to the exergame programme may be helpful in eliciting improvements in this measure.
In conclusion, this study demonstrated improvement in measures of cardiovascular fitness and physical HQL in youth with GMFCS level III CP following a six-week internet-platform interactive exergame cycling programme. Limitations of the study include the lack of a comparison group and a small sample size. A randomized controlled crossover trial is currently being planned. Future research involving youth with other types of disability and typically developing peers will be undertaken, including developing algorithms to enable competitive play between players of different abilities. This will increase the generalizability and appeal of the exergame programme.
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Declaration of interest

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References


Table 1. Participant characteristics (n = 8) and results

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BMI, Body mass index; MACS, Manual ability classification system; GMFM, Gross motor function measure; THR, Target heart rate; HR, Heart rate; SRT-III, Shuttle run test; SD, Standard deviation.
Figure 1. The exergame cycling unit.
Figure 2. Mean weekly playtime and mean weekly playtime above target heart rate per participant over the six-week intervention. Error bars represent standard deviations. The horizontal lines denote the recommended minimum weekly playtime of 90 minutes and the recommended minimum weekly playtime above target heart rate of 60 minutes.
Figure 3. Percentage of total playtime at different levels of heart rate reserve. Error bars represent standard deviations.