2020

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Abstract

Purpose: To quantify the impact of motor-assisted elliptical (ICARE) training on cardiorespiratory fitness, balance and walking function of an adolescent with walking limitations due to cerebral palsy.

Materials and methods: A thirteen-year-old boy with hemiplegic cerebral palsy (Gross Motor Function Classification System II) and autism participated. Peak oxygen consumption (peak VO_2, primary outcome measure), oxygen cost of walking, Pediatric Balance Scale (PBS), modified Timed Up and Go (mTUG), 2-Minute Walk Test (2MWT), and gait characteristics (speed, cadence, step length, single support time) were assessed prior to and after completion of 24 sessions

Published in *European Journal of Physiotherapy* 2020, vol. 22, no. 3, pp. 124–132. doi:10.1080/21679169.2018.1536764

Submitted 11 May 2018; revised 8 October 2018; accepted 10 October 2018; published 11 December 2018.

of moderate- to vigorous- intensity ICARE training. The goal was to engage the participant in 3 weekly sessions for 8 weeks with progressively challenging training parameters (speed, time overriding the motor's assistance, step length).

Results: From pre- to post-intervention, improvements were detected for peak VO_2 (27.2 vs. 40.2 ml/ kg/min), oxygen cost (0.24 vs. 0.17 ml/kg/m at 1.52 m/s), PBS (47 vs. 55), mTUG (8.5 vs. 7.1 seconds), 2MWT (76.8 vs. 128.3 meters). Though not all displayed clinically significant changes, self-selected and fast walking speeds improved.

Conclusions: Fitness, balance and walking improvements were achieved by an adolescent with cerebral palsy and autism after participating in a moderate- to vigorous-intensity exercise.

Keywords: peak oxygen consumption, aerobic fitness, gait, balance, cerebral palsy

Introduction

The limited walking ability experienced by many children and adolescents with cerebral palsy (CP) due to balance issues and impaired neuromuscular control can impact the level of engagement in physical activities, leading to lower cardiorespiratory fitness than typically developing peers [1–5]. Physical inactivity-induced decreases in cardiorespiratory fitness contribute to secondary medical conditions [6], such as obesity [7,8], pulmonary [9] and cardiometabolic diseases [7]. In contrast, studies have demonstrated the relationship between enhanced aerobic fitness and changes in walking capacity in individuals with disabilities (e.g. [10,11], including children and adolescents with CP [2,12]. In addition, changes in walking speed positively impact individuals with stroke, allowing them to progress from restricted household ambulation to community ambulation, increasing participation in life situations [13–15]. For children with CP, it is well documented that those who perform at higher levels of gross motor function [e.g. Gross Motor Function Classification System (GMFCS) I] versus lower levels (e.g. GMFCS IV) are able to accomplish more activities of daily living [16] and engage in greater levels of physical activity including an increased number of steps taken in the community [17,18]. Thus, enhancing walking ability and fitness should encourage greater engagement in activities of importance (e.g. family outings, school activities) as children transition from adolescence to adulthood, providing them with the tools to achieve functional independence and health.

Certain gait-training devices have the capacity to challenge the cardiovascular system when used with training protocols that elevate the heart rate to recommended levels [19]. Although stationary cycles potentially offer children the means to achieve fitness and lower extremity muscle strength [20], this approach does not address the standing, dynamic balance issues that can emerge with CP [21,22]. Body weight-supported treadmill training has generated improvements in children with mobility impairments [23], including improvements in walking speed and endurance [24] and balance [25] of children and adolescents with CP. This approach, however, does not generate consistent results [23,26,27]. Additionally, its use is infrequent in smaller rehabilitation centers and schools partially due to the demanding manual labor involved with assisting an individual with spasticity and weakness to sustain the repetitive stepping motion [28–31]. Robotic devices require less clinician effort to advance and stabilize the legs. Children and adolescents with CP have benefited from such approach, with improvements in walking [32] and balance [33,34] measures following LokomatVR interventions. Unfortunately, robotic locomotor training devices are not widely available beyond large hospitals/research environments given the cost of the technology [35,36]. In addition, most robotic devices do not promote sufficient cardiorespiratory stress to improve aerobic fitness [37].

The ICARE (**Figure 1**) is a therapeutic robotic device developed to address walking and fitness of individuals with disabilities. It has recently been used with children with limited walking ability [38,39]. Compared to other elliptical devices, the ICARE promotes movement patterns that more closely simulate the joint motions and muscle demands of overground gait [40,41]. With regards to the resisted training mode, the device can operate similarly to other traditional ellipticals; however, the device also includes a motor-drive system that facilitates pedal advancement. This inclusion enables the repetitive practice of gait-like movement while minimizing the physical demand placed on clinicians when delivering gait-training interventions [28–31]. As a user's capacity improves, alterations to the ICARE's speed and amount of motor assistance can be tailored to produce continued cardiorespiratory and musculoskeletal challenges [38,39,42–44]. Another unique therapeutic feature of the device is the adjustable step length (from 19–71 cm) [45] which can be

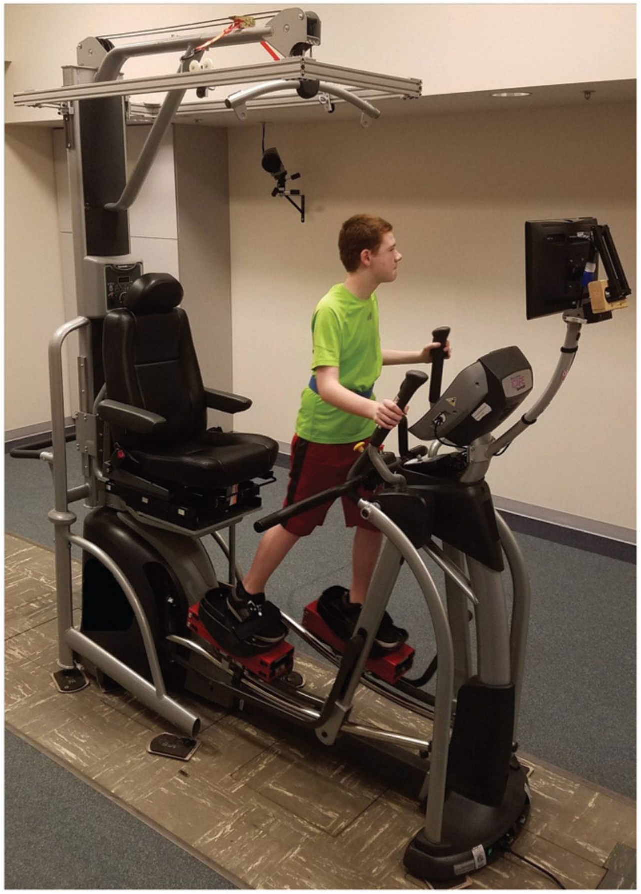

Figure 1. The motor-assisted elliptical device ICARE in use by our participant (photograph obtained with parental consent).

used clinically [46] to progressively increase hip position at terminal stance across training sessions.

Although the device has the potential to address cardiovascular fitness in children [38,39] and walking efficiency in adults [43], no pediatric studies to date have explored a more detailed analysis of cardiorespiratory measures after ICARE training. The current work adds to the existing literature by investigating changes in oxygen uptake, a measure that reflects adaptations in aerobic capacity, after participation in an ICARE intervention. The purpose of this work was to quantify the impact of ICARE training on cardiorespiratory fitness, balance and walking function of an adolescent with limited walking ability and low cardiorespiratory fitness [4] due to CP. We hypothesized that (1) cardiorespiratory fitness would be enhanced after participation in our systematic ICARE exercise protocol, along with improvements in (2) balance and (3) walking function.

Materials and methods

Participant

The participant was recruited from the local community via word of mouth. Prior to participating, parental informed consent and child assent were obtained according to the World Medical Association Declaration of Helsinki and the protocol approved by the Institutional Review Board at Madonna Rehabilitation Hospitals. Physician clearance for participation was obtained and the participant's parent completed the PAR-Q+ (The Physical Activity Readiness Questionnaire for Everyone) to screen for any potential medical conditions that might increase the risk associated with study participation.

A 13-year-old boy with left-sided spastic hemiplegic CP and high-functioning autism spectrum disorder participated. Our participant started walking at the age of 18 months without the use of any assistive devices (left ankle-foot orthosis was introduced at the age of 26 months). Observational gait analysis revealed unilateral gait deviations characteristic of his primary diagnosis of hemiplegic CP. These characteristics included left foot-flat contact (initial contact), left knee flexion from initial contact through stance phase, no apparent push-off, and a contralateral (i.e. right) heel rise at midstance to assist with toe clearance. The characteristic gait deviations expected due to autism spectrum disorder [47,48] were not observed. The participant's parent reported no history of medication use for CP or autism, and the participant was not receiving any therapy during the study. No history of surgeries was reported, but the participant received a full year of chiropractic care for a left hip complaint 18 months before engaging in our study. He did not require physical assistance nor did he use lower limb orthotics to walk (stopped using 6 months prior to study because, per parent report, the ankle-foot orthosis did not fit him properly). The participant's parent reported a history of 5–10 falls in the previous year when attempting to run or walk on uneven surfaces, with no hospitalizations required. His motor function was classified as level II according to the Expanded and Revised GMFCS [49] with the key distinction from level I based on the need for handrail use to go up/down a flight of stairs. Anthropometric measurements were completed during initial assessment including height (159 cm), weight (49 kg), and leg length (42 cm). Scores from manual muscle testing identified bilateral hip extensors as fair (3/5), knee extensors as good (4/5), right ankle plantar flexor as good (4/5), and left ankle plantar flexor as poor (2/5). The range of motion for bilateral popliteal angles (participant supine, 90° of hip flexion) indicated right and left knees lacked 30° and 35° from full knee extension, respectively.

Cardiorespiratory fitness test

Resting heart rate (HR) and blood pressure were taken using standard procedures after the participant entered our lab and rested quietly for five minutes. Since traditional treadmill protocols for aerobic capacity testing (e.g. Bruce protocol) are not appropriate for children with CP, we utilized the protocol presented by Verschuren and colleagues [50]. Specifically, the session started with the participant performing a warm-up session by walking for 3 minutes at 2 km/hr and no treadmill inclination. After the warm-up, he rested for 5 minutes seated on a chair with arm and back rests. During the rest interval, a snug fitting face mask was placed on his face and connected to the metabolic measurement system with a 6-foot flexible tube. After the rest period, the participant began walking on the treadmill at a speed of 2 km/hr with an incline of 2%. The speed was increased by 0.25 km/ hr every minute for the remainder of the test while the incline was maintained at 2%. The protocol ended when the participant indicated he could not continue due to fatigue and could no longer maintain the pace imposed by the treadmill speed. A gait belt was worn for safety and we asked our participant to use only light finger touch on treadmill handrails if needed to maintain balance.

Balance test

During a separate session, the participant performed all 14 items of the Pediatric Balance Scale (PBS). This pediatric-valid scale was used since it has excellent reliability for children with CP [51,52]. PBS items include functional tasks experienced during everyday tasks, involving the control of balance during sitting, standing, transferring, turning, stepping, reaching forward and picking an object from the floor [53].

Walking function

On the same day of the PBS session, the clinical measures Timed Up and Go (TUG) test and 2-Minute Walk Test (2MWT) were administered to describe the participant's walking function, as well as the assessment of his spatiotemporal characteristics of walking. The modified TUG (mTUG) test was performed in which a seat without armrests was used and adjusted to allow for 90° of hip and knee flexion with the adolescent sitting comfortably with feet on the floor [54]. On the command 'ready, set, go', the participant stood up and walked around a cone placed on the floor three meters away. The test was over when he sat back down on the chair. This test was performed three times. The mTUG has been administered for children and adolescents with disabilities, including those with CP, with high reliability [55]. During the 2MWT, a validated test of walking endurance [56,57], the participant walked for 2 minutes at a self-selected comfortable speed on an unobstructed 38.4-meter long hallway. Spatiotemporal characteristics of walking (speed, cadence, step length and single support time) were assessed by having the participant traverse a 6-meter long electronic walkway (GAITRiteVR) at his self-selected comfortable speed and at a fast pace. The participant completed three attempts for each walking speed (self-selected and fast pace).

Training protocol

Training sessions (and evaluations) for this study were conducted by both a physiotherapist and an exercise physiologist (with more than 12 years of clinical experience) and occurred at Madonna Rehabilitation Hospitals' Movement and Neurosciences Center located within the Institute for Rehabilitation Science and Engineering. An E872MA ICARE (SportsArt, Mukilteo, WA) motor-assisted elliptical device was used for the intervention, enabling gait-like movement pattern at speeds up to 65 cycles per minute (CPM) [40,41,58].

Baseline training parameters, such as comfortable speed and step length [allowing for gait-like trailing limb posture in late stance (i.e. visually inspected thigh extension)], were identified during the familiarization session with the device and used as initial training parameters. During this session, the participant trained for _5 minutes with motor assistance. Speed was increased gradually until the participant's HR approximated 139 beats per minute (bpm), which corresponds to 70% of his estimated maximum HR (i.e. 199 bpm) according to the predictive equation 208–(0.7*age) [59,60].

The goal of the training protocol was to engage the participant in 8 weeks of periodized exercise (3 times per week) for cardiorespiratory fitness while simultaneously addressing walking ability. The protocol duration (i.e. 24 sessions) was selected based on prior literature work with children and adolescents with CP [61–63] and previous pediatric work from our lab [38,39] that elicited favorable effects in aerobic fitness after 24 sessions.

Our goal was to build the participant's weekly exercise tolerance to 150 minutes of moderate intensity exercise [50–70% of predicted maximum HR (HRmax)], 75 minutes of vigorous intensity (70–85% HRmax), or a combination of intensities [64]. One-minute bouts of higher intensity ICARE exercise (>70% HRmax) were accomplished by having the participant systematically override the device's motor at predetermined intervals within each training session. Across the 24 sessions, protocol parameters including speed, total exercise time, time spent overriding the motor's assistance, and step length, were systematically altered to progressively challenge the participant's cardiorespiratory fitness and walking. Specifically, the planned increments included a 5% increase in training duration every session and in training speed every 4 sessions. The one-minute higher intensity bouts were introduced in the first session (between one to three bouts, as tolerated) and scheduled to increase by one bout every three sessions. The systematic progression of our protocol allows for acclimation to exercise while reducing the risk of injury in individuals with CP and limited walking ability (GMFCS II) [65–67]. Exercise intensity was monitored via the participant's HR and his comments regarding perceived exertion within each session. The parameter step length was scheduled to increase every three sessions; however, this parameter was only manipulated based on the comfort level of the participant. Training step length was re-assessed every session and it was increased as tolerated by the participant to account for his comfort during the exercise. During each session, resting and exercise HRs were recorded, as well as HR during bouts of overriding the motor's assistance.

Evaluation of outcome measures

Assessment of all outcome measures took place one week prior to and one week after the completion of the intervention. Tests involving balance and walking function were performed first with the subsequent cardiorespiratory fitness test (peak VO_2 and oxygen cost) scheduled 48 hours later.

Data analysis

Cardiorespiratory fitness was evaluated sub-maximally as testing HR was limited to 85% predicted of age-appropriate maximum, consistent with the American College of Sports Medicine guidelines for individuals without previous stress testing [68]. Breath-by-breath gas analysis (TrueOne® 2400, ParvoMedics, East Sandy, UT) was utilized to evaluate peak oxygen consumption (peak VO_2 in ml/kg/m, the primary outcome variable) during the incremental exercise and also the oxygen cost of walking (ml/kg/m), defined as the amount of oxygen consumed (ml) per body weight (kg) at a selected treadmill speed (m/min). The fastest treadmill speed achieved during pre-assessment was selected for comparison at post-intervention. An average of 5 breaths was used to minimize the inclusion of potential artifacts (breath-by-breath noise) in the analysis.

The PBS's 14 items, graded from 0 (unable to perform) to 4 (perform without difficulty), were calculated and presented as static (PBS-Static, 6 items, sub-total maximum of 24 points), dynamic (PBS-Dynamic, 8 items, sub-total maximum of 32 points), and total (PBS-Total, maximum of 56 points).

Dynamic stability during gait was tested using the mTUG test and the fastest time was used for comparison. For the participant's gait spatiotemporal characteristics (GAITRiteVR), the average of three trials was used for speed (m/s), cadence (steps/min), step length (cm), and single limb support (% gait cycle, GC) for each walking speed.

Descriptive statistics were calculated and used for comparison of findings.

Results

Training progression

Scheduling conflicts allowed the participant to train an average of 2.4 times/week over a period of 9.7 weeks (68 days). All 24 sessions were completed and 1 session re-scheduled (session 23) since the participant arrived at our lab exhibiting symptoms of an upper respiratory tract illness and expressed feeling fatigued. Adverse events were monitored and no reports generated across the duration of the study.

With regards to the overall goal of the training protocol, the participant achieved a total of 52 minutes of moderate intensity exercise during the first week, in which 11 minutes were performed at a vigorous intensity. By training week eight, the participant exercised for a weekly total of 142 minutes at moderate intensity, with 47 minutes performed at vigorous intensity. A drop in intensity occurred during the last week, with the participant achieving a weekly total of 95 minutes of moderate-intensity exercise, with 14 minutes performed at a vigorous intensity (**Figure 2**).

Considering the training progression, the participant trained for 24 minutes in the first session with the motor's assistance at an average speed of 45 CPM. The combination of exercise duration and training speed promoted 675 strides in this first training session, with the ICARE step length at 49 cm. He was able to accomplish three higher-intensity bouts of overriding the motor's assistance, with the average HR during overriding the motor's assistance of 188 beats per minute (bpm). The number of bouts overriding the motor's assistance achieved its peak on session 21 with the participant overriding for 18 minutes (average HR of 156 bpm) out of the total session duration of 49 minutes. During the last session, the participant trained for 49 minutes with the motor's assistance at an average speed of 56 CPM. This combination of exercise duration and speed promoted 2,720 strides in the last training session, with the ICARE step length at 68 cm. The participant completed nine higher-intensity bouts of overriding the motor's assistance at this last session, with the average HR during overriding the motor's assistance of 148 bpm (Figure 2).

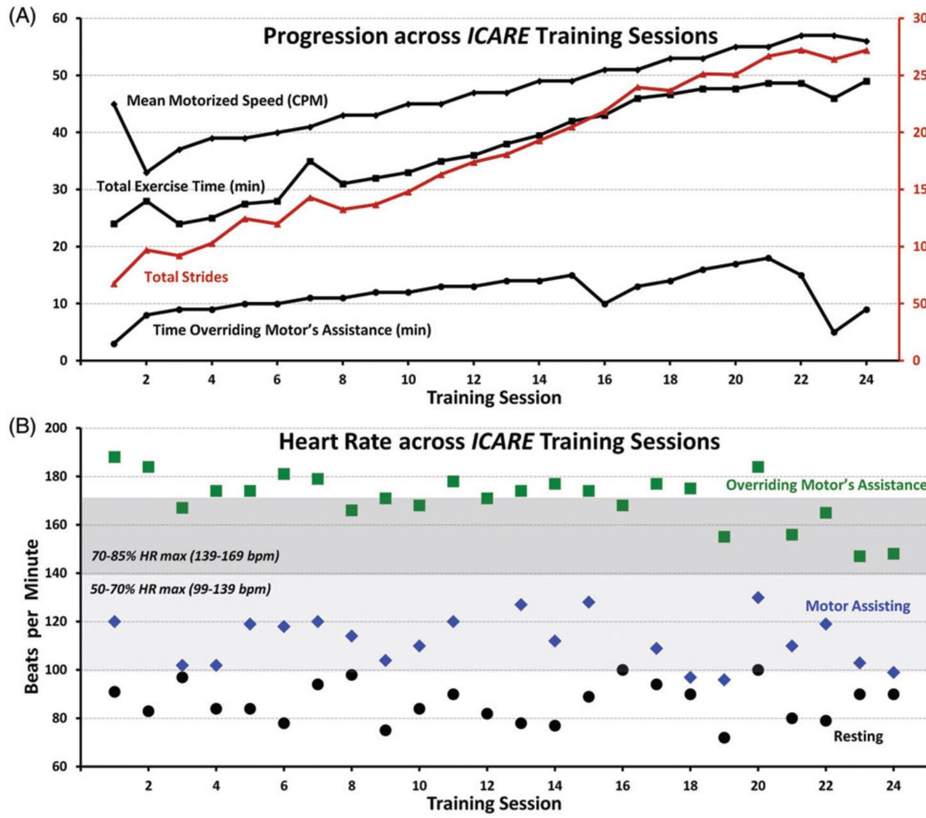

Figure 2. (A) Graph displays the ICARE training progression across 24 sessions including the parameters speed, exercise time, number of strides, and total time (1-minute bouts) overriding the motor's assistance. All parameters besides total strides (red) are represented on the left y-axis. (B) Graph displays the average heart rate during bouts of overriding the motor's assistance (green square), while exercising with the motor assistance (blue diamond), and at rest prior to exercising (black dot). The regions bounded by light and dark grey bands on the bottom graph represent the expected age-appropriate heart rate range for moderate and vigorous intensities, respectively, of an adolescent with predicted maximum heart rate of 199 beats per minute.

Cardiorespiratory fitness

Peak VO_2, the primary outcome measure, improved 48% after intervention (**Table 1**). The 13 ml/kg/min improvement surpassed the small detectable change of 5.7 ml/kg/min (captured through upright cycle ergometer) for children with CP, GMFCS I-III [69]. This improved aerobic capacity was achieved with a faster pre-post testing treadmill speed (1.52 vs. 1.97 m/s) and exercise testing time (11:49 vs. 16:12

Table 1. Cardiorespiratory fitness, balance, walking function and spatiotemporal characteristics.

	PRE	*POST*
Peak VO_2 (ml/kg/min)	27.2	40.2
Oxygen Cost (ml/kg/m) at 1.52 m/s	0.24	0.17
Pediatric Balance Scale (Total)	47	55*§
Static	20	23*§
Dynamic	27	32*§
Modified Timed Up & Go (s)	8.5	7.1§
2-Minute Walk Test (m)	76.8	128.3§
Self-selected comfortable speed (m/s)	0.63	1.0§
Cadence (steps/min)	79	107
Step length, right (cm)	42.4 (6.5)	60.7 (4.3)
Step length, left (cm)	52.5 (9.2)	53.2 (11.9)
Single support, right (% Gait cycle)	39.4 (10.6)	37.2 (8.7)
Single support, left (% Gait cycle)	30.7 (11.2)	33.0 (9.0)
Fast Walking Pace (m/s)	1.20	1.75
Cadence (steps/min)	121	142
Step length, right (cm)	52.4 (10.1)	73.1 (4.5)
Step length, left (cm)	66.2 (7.7)	74.4 (5.1)
Single support, right (% Gait cycle)	43.3 (14.5)	43.1 (6.6)
Single support, left (%Gait cycle)	34.6 (10)	36.9 (9.0)

* Value surpassing the minimal detectable change (MDC).

§ Value surpassing the minimal clinically important difference (MCID). 1.52 m/s: fastest speed achieved during pre-assessment.

min). Our participant's walking efficiency (i.e. oxygen cost of walking) improved 29% with the decrease in 0.07 ml/kg/m after intervention (Table 1). The change, however, did not reach the minimal detectable change (MDC) in oxygen cost of 0.11 ml O_2/kg/m for children with CP (GMFCS II) [70].

Balance

A 17% (eight points) improvement was recorded for the total score of PBS, surpassing the minimal clinically important difference (MCID) of 5.8 points and the MDC of 1.6 points for children with CP [51]. When breaking down the components of PBS, both static (3-point increase) and dynamic (5-point increase) components also surpassed the MCID and MDC scores [51], suggesting a clinically robust increase in balance performance (Table 1).

Walking function

The mTUG was accomplished 16% faster from pre- to-post assessment, achieving the large effect minimal clinically important difference (MCID) score of 1.2s for children with CP (GMFCS II) [71]. The participant's walking endurance (2MWT) improved 67%, also surpassing the MCID score (16.6 m) for children with disabilities who walk independently [57] (Table 1).

Walking speed during self-selected comfortable and fast paces improved 59% and 46%, respectively, surpassing the large effect size MCID of 0.13 m/s [72,73]. Additionally, single support time became more symmetrical during self-selected comfortable walking speed owing to increases in left (involved) side single limb support time and decreases in the right (uninvolved) single limb support time. This change was also observed during the fast walking speed, although the change in the non-involved side was not substantial (Table 1).

Discussion

Many adolescents with disabilities lack the cardiorespiratory fitness to sustain meaningful activities with peers and family [3,74]. Engagement in regular exercising is usually diminished and walking endurance is limited as fatigue hinders ambulatory capacity [6,75]. The gradual increased participation in a moderate-intensity exercise, including greater weekly minutes of high-intensity exercise across the 9.6 weeks of ICARE training, allowed our participant to achieved enhanced aerobic fitness and walking endurance. This case report supports the use of the motor-assisted elliptical device to improve the fitness of those with impaired walking ability, as exhibited by our adolescent with CP and autism.

Consistent with our first hypothesis, the structured ICARE exercise stimulated positive changes in our participant's cardiorespiratory measures, building his tolerance to aerobic exercise and enhancing his walking efficiency. When compared with other gait training devices, the 48% increase in peak VO_2 exhibited by our participant well exceeded the improvements previously documented in the literature, such as 20%–22% after cycle ergometer training [76,77]. No studies to date have explored the impact on aerobic capacity in children with CP and autism after training with robotic technologies. However, a prior study conducted with adults with spinal cord injury indicated no changes in peak VO_2 after 24 training sessions [37], suggesting that the technology used in our study is promising with regards to promoting challenges to the users' aerobic capacity.

In agreement with our second hypothesis, balance improvement was recorded after the completion of the gait-training exercise protocol. The increase in total PBS score placed our participant with matched scores of a large sample of typically developing boys ages 7–13 years old [78]. Additionally, the larger increase in the dynamic score of PBS compared with the static score was in alignment with the faster time accomplished during the mTUG, which requires the control of balance during walking and turning. Although slower than the time accomplished by typically developing age-matched individuals (5.6 seconds) [55], our participant improved his mTUG time, accomplishing the task 21% faster than 14-year-old adolescents with CP (GMFCS II) [71]. Moreover, the unilateral increase in single limb support time on the involved side (i.e. left) during walking indicates improved stability and thus enhanced balance control. Together, these findings suggest the high repetition of gaitlike movement promoted by the ICARE enhanced dynamic stability during walking. Consistent with our balance results, our participant walked up/down a set of stairs without using the handrail once the intervention was completed. Additionally, the participant's parent reported he fell only once at the beginning of the intervention (between week one and two) and that his movements when walking and running seemed more stable and controlled compared with before the intervention. These accomplishments suggest a positive impact on his overall gross motor function, likely placing him at the GMFCS level I.

Consistent with our third hypothesis, our participant enhanced his walking ability after the intervention. His walking endurance improved (i.e. walked further during the 2MWT at post-intervention), corroborating anecdotal parent reports of improved walking outside the lab environment (i.e. participant walking at the same pace with siblings from school to home without fatiguing, whereas previously siblings had to wait for him to catch up). Supporting this report is the improved oxygen cost of walking achieved by our participant. Although not reaching the minimal detectable change for children with CP (GMFCS II) [70], the change placed the participant at similar level of typically developing children [79,80], and can partially explain the enhanced walking ability post-intervention.

At pre-assessment, the participant walked at a speed that was 38% slower than age-matched adolescents with CP (GMFCS II) [81]. This walking speed was confirmed by the participant's parent as his usual pace outside the lab environment, which may have been adopted to counteract fatigue when trying to walk at similar paces of siblings and peers. After the intervention, our participant's walking speed fell only 3% slower than the range achieved by age-matched boys without known disabilities (1.17 ± 0.14 m/s) [82]. The walking speed increase at post-intervention emerged due to improvements in both step length and cadence. Considering the ICARE's training step length, the participant tolerated a total of 19-cm increase throughout the 24 sessions. Specifically, the initial ICARE step length was 49 cm based on the participant's report of comfortable step length on the device. This initial ICARE step length also accommodated the participant's overground step length for both involved and non-involved limbs (bilateral average of 47 cm during self-selected comfortable speed). After the first three sessions, a progression of the ICARE step length occurred until session 9 (62 cm), with the participant indicating that the new training step length was comfortable but further progression would not be tolerated at the time. Following this, the ICARE step length increased every fifth session to the final length of 68 cm (at session 21), which was similar to the participant's step length used during overground fast walking speed at post-intervention (73 cm right, 74 cm left). The increase in overground step length along with a more symmetrical step length could have emerged from the mass repetition of the progressively increasing ICARE step length, providing the adolescent with a 'new template' of step length. This finding is consistent with the neurorehabilitation literature that highlights the value of high-intensity repetition (mass repetition) of gait-like movements (task specificity) to promote positive behavioral changes [83,84].

Training intensity increased systematically and, despite the drop in the time overriding the motor's assistance at later sessions, our participant exhibited improvements across sessions. Training times were initially short (_24 minutes) to allow him to adapt to the exercise intensity, and bouts of overriding the motor's assistance increased gradually across training sessions. Although tolerated by the participant, we noticed that when overriding the motor's assistance (Figure 2), his HR was above the upper limit for his age-expected vigorous intensity. The fact that our participant was able to increase the number of minutes overriding the motor's assistance (while exercising for longer periods of time) is of great value, suggesting our protocol promoted the gradual improvement in his tolerance to exercise at higher intensities. However, this high intensity level may explain the drop in the number of bouts of overriding the motor's assistance towards the end of the intervention (i.e. sessions 22–24) as the participant may have reached the exhaustion stage of the general adaptation syndrome [85] and was no longer able to adapt to the increased training intensity. Future work should consider the monitoring of individual responses [86], explore different numbers of sessions, and/or the addition of an active rest phase to enhance physiological adaptation. Moreover, inclusion of interactive systems, such as virtual reality, may enhance motivation [87] to maintain engagement in exercising at high-intensity levels.

Although promising, generalization of our findings to other children and adolescents with CP and autism is limited since the study was conducted with only one participant. We are aware of the diverse physical constraints and physiological responses to exercise an individual with CP may present. The structured ICARE intervention, however, has been applied to individuals with other neurologic and chronic conditions [38,39,42,43]. Future work should investigate the impact of ICARE interventions on cardiorespiratory health and walking function in large cohorts of children and adolescents with CP and other chronic conditions using higher research designs (e.g. randomized control trial). When sufficient participants cannot be secured, single case experimental designs could also provide significant contributions to scientific/clinical knowledge. Another limitation of our findings is the lack of an automated quantifiable measure for balance control; future studies should implement analyses such as computerized dynamic posturography to strengthen findings from our dynamic balance measures. Additionally, follow-up assessments will be crucial in detecting the longevity of the encouraging changes reported in this study, along with the inclusion of investigations of the relationship between changes in aerobic capacity and walking abilities in real-world scenarios, such as participation in community and life situations.

Conclusion

Improvements in cardiorespiratory measures were recorded in an adolescent with limited walking ability due to CP and autism after participating in a moderate- to vigorous-intensity ICARE exercise protocol. Balance and walking were also enhanced as the ICARE device enabled sustained gait-like activity while allowing the adolescent to build exercise tolerance towards recommended levels of aerobic exercising.

Acknowledgments We would like to acknowledge Sonya L. Irons, PT, DPT, CCS for her contribution to the assessment design, and William Frederick Garvey for his assistance with data collection.

Disclosure Guilherme M. Cesar has no conflict of interest. Judith M. Burnfield and Thad W. Buster are the inventors of the patented motor-assisted elliptical technology. The patented technology has been licensed to Sports Art for commercial distribution. The inventors receive royalties.

Funding The contents of this work were developed under a grant initially received from the National Institute on Disability and Rehabilitation Research, Department of Education (Grant Number H133G130274; Principal Investigator: Burnfield JM), and subsequently funded through a grant from the National Institute on Disability, Independent Living, and Rehabilitation Research, Administration for Community Living (Grant Number 90IF0060; Principal Investigator: Burnfield JM). The contents of the manuscript do not necessarily represent the policy of the Department of Education or the Administration for Community Living, and endorsement by the federal government should not be assumed

References

[1] Satonaka A, Suzuki N. Aerobic fitness and lifestyle with non-exercise physical activity in adults with cerebral palsy. JPFSM. 2018;7: 1–7.

[2] Verschuren O, Ketelaar M, Gorter JW, et al. Exercise training program in children and adolescents with cerebral palsy. Arch Pediatr Adolesc Med. 2007;161:1075–1081.

[3] Fowler EG, Kolobe TH, Damiano DL, et al. Promotion of physical fitness and prevention of secondary conditions for children with cerebral palsy: section on pediatrics research summit proceedings. Phys Ther. 2007;87:1495–1510.

[4] Balemans AC, Van Wely L, De Heer SJ, et al. Maximal aerobic and anaerobic exercise responses in children with cerebral palsy. Med Sci Sports Exerc. 2013;45:561–568.

[5] Mitchell LE, Ziviani J, Boyd RN. Habitual physical activity of independently ambulant children and adolescents with cerebral palsy: are they doing enough?. Phys Ther. 2015;95:202–211.

[6] Maltais DB, Wiart L, Fowler E, et al. Health-related physical fitness for children with cerebral palsy. J Child Neurol. 2014;29: 1091–1100.

[7] Peterson MD, Gordon PM, Hurvitz EA. Chronic disease risk among adults with cerebral palsy: the role of premature sarcopoenia, obesity and sedentary behaviour. Obes Rev. 2013;14:171–182.

[8] Bandini L, Danielson M, Esposito LE, et al. Obesity in children with developmental and/or physical disabilities. Disabil Health J. 2015;8:309–316.

[9] Choi JY, Rha DW, Park ES. Change in pulmonary function after incentive spirometer exercise in children with spastic cerebral palsy: a randomized controlled study. Yonsei Med J. 2016;57: 769–775.

[10] Macko RF, Ivey FM, Forrester LW, et al. Treadmill exercise rehabilitation improves ambulatory function and cardiovascular fitness in patients with chronic stroke: a randomized, controlled trial. Stroke. 2005;36:2206–2211.

[11] Pang MYC, Eng JJ, Dawson AS, et al. The use of aerobic exercise training in improving aerobic capacity in individuals with stroke: a meta-analysis. Clin Rehabil. 2006;20:97–111.

[12] Ryan JM, Cassidy EE, Noorduyn SG, et al. Exercise interventions for cerebral palsy. Cochrane Database Syst Rev. 2017;6:CD011660.

[13] Schmid A, Duncan PW, Studenski S, et al. Improvements in speed-based gait classifications are meaningful. Stroke 2007;38: 2096–2100.

[14] Lord SE, Rochester L. Measurement of community ambulation after stroke: current status and future developments. Stroke. 2005;36:1457–1461.

[15] Perry J, Garrett M, Gronley JK, et al. Classification of walking handicap in the stroke population. Stroke. 1995;26:982–989.

[16] Lepage C, Noreau L, Bernard PM. Association between characteristics of locomotion and accomplishment of life habits in children with cerebral palsy. Phys Ther. 1998;78:458–469.

[17] Keawutan P, Bell K, Davies PS, et al. Systematic review of the relationship between habitual physical activity and motor capacity in children with cerebral palsy. Res Dev Disabil. 2014;35:1301–1309.

[18] Mitchell LE, Ziviani J, Boyd RN. Characteristics associated with physical activity among independently ambulant children and adolescents with unilateral cerebral palsy. Dev Med Child Neurol. 2015;57:167–174.

[19] United States Department of Health and Human Services. Physical activity guidelines for Americans. Hyattsville, MD: US Department of Health and Human Services; 2008.

[20] 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;90:367–381.

[21] Jantakat C, Ramrit S, Emasithi A, et al. Capacity of adolescents with cerebral palsy on paediatric balance scale and Berg balance scale. Res Dev Disabil. 2015;36C:72–77.

[22] Panibatla S, Kumar V, Narayan A. Relationship between trunk control and balance in children with spastic cerebral palsy: a cross-sectional study. J Clin Diagn Res. 2017;11:YC05–YC08.

[23] Zwicker JG, Mayson TA. Effectiveness of treadmill training in children with motor impairments: an overview of systematic reviews. Pediatr Phys Ther. 2010;22:361–377.

[24] Booth ATC, Buizer AI, Meyns P, et al. The efficacy of functional gait training in children and young adults with cerebral palsy: a systematic review and meta-analysis. Dev Med Child Neurol. 2018;60:866–883.

[25] Grecco LA, Tomita SM, Christovao TC, et al. Effect of treadmill gait training on static and functional balance in children with cerebral palsy: a randomized controlled trial. Braz J Phys Ther. 2013;17:17–23.

[26] Damiano D, DeJong S. A systematic review of the effectiveness of treadmill training and body weight support in pediatric rehabilitation. J Neurol Phys Ther. 2009;33:27–44.

[27] Mutlu A, Krosschell K, Spira D. Treadmill training with partial body-weight support in children with cerebral palsy: a systematic review. Dev Med Child Neurol. 2009;51:268–275.

[28] Corbridge LM, Goldman AJ, Shu Y, et al. Clinician's muscle effort during partial body weight support treadmill training: Is it hard work? Online Proceedings, American Physical Therapy Association's 2009 Annual Conference and Exposition. 2009.

[29] Buster TW, Goldman AJ, Corbridge LM, et al. Partial body weight support treadmill training: clinician's upper extremity muscle activation during facilitation of hemiparetic limb movement. Proceedings, Gait and Clinical Movement Analysis Society Annual Meeting. March 13, 2009, Denver, CO.

[30] Shu Y, Taylor AP, Buster TW, et al. editors. Clinicians' motion and muscle activation patterns during body weight support treadmill training. Proceedings, Gait and Clinical Movement Analysis Society Annual Meeting. May 12, 2012, Grand Rapids, MI.

[31] Burnfield JM, Buster TW, Goldman AJ, et al. Partial body weight support treadmill training speed influences paretic and non-paretic leg muscle activation, stride characteristics, and ratings of perceived exertion during acute stroke rehabilitation. Hum Mov Sci. 2016;47:16–28.

[32] Borggraefe I, Schaefer JS, Klaiber M, et al. Robotic-assisted treadmill therapy improves walking and standing performance in children and adolescents with cerebral palsy. Eur J Paediatr Neurol. 2010;14:496–502.

[33] Druzbicki M, Rusek W, Szczepanik M, et al. Assessment of the impact of orthotic gait training on balance in children with cerebral palsy. Acta Bioeng Biomech. 2010;12:53–58.

[34] Wallard L, Dietrich G, Kerlirzin Y, et al. Effect of robotic-assisted gait rehabilitation on dynamic equilibrium control in the gait of children with cerebral palsy. Gait Posture. 2018;60:55–60.

[35] Hidler J, Hamm LF, Lichy A, et al. Automating activity-based interventions: the role of robotics. J Rehabil Res Dev. 2008;45: 337–344.

[36] Morrison SA. Financial feasibility of robotics in neurorehabilitation. Top Spinal Cord Inj Rehabil. 2011;17:77–81.

[37] Hoekstra F, van Nunen MP, Gerrits KH, et al. Effect of robotic gait training on cardiorespiratory system in incomplete spinal cord injury. J Rehabil Res Dev. 2013;50:1411–1422.

[38] Cesar GM, Irons SL, Garbin A, et al. Child with traumatic brain injury improved gait abilities following intervention with pediatric motor-assisted elliptical training: a case report. J Neurol Phys Ther. 2017;41:84.

[39] Burnfield JM, Cesar GM, Buster TW, et al. Walking and fitness improvements in child with diplegic cerebral palsy following motor-assisted elliptical training intervention. Pediatr Phys Ther. 2018;30:E1–E7

[40] Burnfield JM, Cesar GM, Buster TW, et al. Kinematic and muscle demand similarities between motor-assisted elliptical training and walking: implications for pediatric gait rehabilitation. Gait Posture. 2017;51:194–200.

[41] Burnfield JM, Shu Y, Buster T, et al. Similarity of joint kinematics and muscle demands between elliptical training and walking: implications for practice. Phys Ther. 2010;90:289–305.

[42] Irons SL, Buster TW, Karkowski-Schelar E, et al. Individuals with multiple sclerosis improved walking endurance and decreased fatigue following motor-assisted elliptical training intervention. Arch Phys Med Rehabil. 2016;97:e34.

[43] Irons SL, Brusola GA, Buster TW, et al. Novel motor-assisted elliptical training intervention improves Six-Minute Walk Test and oxygen cost for an individual with progressive supranuclear palsy. Cardiopulm Phys Ther J. 2015;26:36–41.

[44] Burnfield JM, Yeseta M, Buster TW, et al. Individuals with physical limitations can benefit from training on a motorized elliptical for community-based exercise. Med Sci Sports Exerc. 2012;45:S360.

[45] Burnfield JM, Buster TW, Pfeifer CM, et al. Adapted motor-assisted elliptical for rehabilitation of children with physical disabilities. J Med Device. 2018; Published online ahead of print.

[46] Pang M, Yang J. The initiation of the swing phase in human infant stepping: importance of hip position and leg loading. J Physiol. 2000;528:389–404.

[47] Kindregan D, Gallagher L, Gormley J. Gait deviations in children with autism spectrum disorders: a review. Autism Res Treat. 2015; 2015:741480.

[48] Pauk J, Zawadzka N, Wasilewska A, et al. Gait deviations in children with classic high-functioning autism and low-functioning autism. J Mech Med Biol. 2017;17:1750042.

[49] Palisano RJ, Rosenbaum P, Bartlett D, et al. Gross Motor Function Classification System: Expanded and revised. 2007. Available at https://canchild.ca/system/tenon/assets/attachments/000/000/058/original/GMFCS-ER_English.pdf (accessed 23 August, 2018).

[50] Verschuren O, Takken T, Ketelaar M, et al. Reliability and validity of data for 2 newly developed shuttle run tests in children with cerebral palsy. Phys Ther. 2006;86:1107–1117.

[51] Chen CL, Shen IH, Chen CY, et al. Validity, responsiveness, minimal detectable change, and minimal clinically important change of Pediatric Balance Scale in children with cerebral palsy. Res Dev Disabil. 2013;34:916–922.

[52] Gan SM, Tung LC, Tang YH, et al. Psychometric properties of functional balance assessment in children with cerebral palsy. Neurorehabil Neural Repair. 2008;22:745–753.

[53] Franjoine MR, Gunther JS, Taylor MJ. Pediatric Balance Scale: a modified version of the Berg Balance Scale for the school-age child with mild to moderate motor impairment. Pediatr Phys Ther. 2003;15:114–128.

[54] Butz SM, Sweeney JK, Roberts PL, et al. Relationships among age, gender, anthropometric characteristics, and dynamic balance in children 5 to 12 years old. Pediatr Phys Ther. 2015;27:126–133.

[55] Nicolini-Panisson RD, Donadio MV. Normative values for the Timed 'Up and Go' test in children and adolescents and validation for individuals with Down syndrome. Dev Med Child Neurol. 2014;56:490–497.

[56] Bohannon RW, Bubela D, Magasi S, et al. Comparison of walking performance over the first 2 minutes and the full 6 minutes of the Six-Minute Walk Test. BMC Res Notes. 2014;7:269.

[57] Pin TW, Choi HL. Reliability, validity, and norms of the 2-min walk test in children with and without neuromuscular disorders aged 6-12. Disabil Rehabil. 2018;40:1266–1272.

[58] Nelson CA, Burnfield JM, Shu Y, et al. Modified elliptical machine motor-drive design for assistive gait rehabilitation. J Med Devices. 2011;5:021001–021007.

[59] Mahon AD, Marjerrison AD, Lee JD, et al. Evaluating the prediction of maximal heart rate in children and adolescents. Res Q Exerc Sport. 2010;81:466–471.

[60] Machado FA, Denadai BS. Validity of maximum heart rate prediction equations for children and adolescents. Arq Bras Cardiol. 2011;97:136–140.

[61] Verschuren O, Peterson MD, Balemans AC, et al. Exercise and physical activity recommendations for people with cerebral palsy. Dev Med Child Neurol. 2016;58:798–808.

[62] Unnithan VB, Katsimanis G, Evangelinou C, et al. Effect of strength and aerobic training in children with cerebral palsy. Med Sci Sports Exerc. 2007;39:1902–1909.

[63] Lauglo R, Vik T, Lamvik T, et al. High-intensity interval training to improve fitness in children with cerebral palsy. BMJ Open Sport Exerc Med. 2016;2:e000111.

[64] Centers for Disease Control and Prevention. Target heart rate and estimated maximum heart rate. Division of Nutrition, Physical Activity, and Obesity, National Center for Chronic Disease Prevention and Health Promotion 2015; Available from: https://www.cdc.gov/physicalactivity/basics/measuring/heartrate.htm (accessed 23 August, 2018).

[65] Jacobs PL, Svoboda SM, Lepeley A. Chapter 8: neuromuscular conditions and disorders. In: Jacobs PL, editor. NSCA's essentials of training special populations. Champaign, IL: Human Kinetics; 2018.

[66] Maltais DB. Chapter 27: cerebral palsy. In: Moore GE, Durstine JL, Painter PL, editors. ACSM's exercise management for persons with chronic diseases and disabilities. 3rd ed. Champaign, IL: Human Kinetics; 2016.

[67] Reuter BH, Dawes JJ. Chapter 20: program design and technique for aerobic endurance training. In: Haff GG, Triplett NT, editors. Essentials of strength training and conditioning. 4th ed. Champaign, IL: Human Kinetics; 2016.

[68] Riebe D, Ehrman JK, Liguori G, et al. ACSM's guidelines for exercise testing and prescription. 10th edition. Philadelphia, PA: Wolters Kluwer; 2018.

[69] Brehm MA, Balemans AC, Becher JG, et al. Reliability of a progressive maximal cycle ergometer test to assess peak oxygen uptake in children with mild to moderate cerebral palsy. Phys Ther. 2014;94:121–128.

[70] Thomas SS, Buckon CE, Schwartz MH, et al. Variability and minimum detectable change for walking energy efficiency variables in children with cerebral palsy. Dev Med Child Neurol. 2009;51: 615–621.

[71] Hassani S, Krzak JJ, Johnson B, et al. One-Minute Walk and modified Timed Up and Go tests in children with cerebral palsy: performance and minimum clinically important differences. Dev Med Child Neurol. 2014;56:482–489.

[72] Pathokinesiology Service and Physical Therapy Department. Observational gait analysis. 4th ed. Downey, CA: Los Amigos Research and Education Institute, Inc., Rancho Los Amigos National Rehabilitation Center; 2001.

[73] 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;50:918–925.

[74] King G, Law M, King S, et al. A conceptual model of the factors affecting the recreation and leisure participation of children with disabilities. Phys Occup Ther Pediatr. 2003;23:63–90.

[75] Verschuren O, Takken T. Aerobic capacity in children and adolescents with cerebral palsy. Res Dev Disabil. 2010;31:1352–1357.

[76] Nsenga AL, Shephard RJ, Ahmaidi S. Aerobic training in children with cerebral palsy. Int J Sports Med. 2013;34:533–537.

[77] Shinohara TA, Suzuki N, Oba M, et al. Effect of exercise at the AT point for children with cerebral palsy. Bull Hosp Jt Dis. 2002;61: 63–67.

[78] Franjoine MR, Darr N, Held SL, et al. The performance of children developing typically on the pediatric balance scale. Pediatr Phys Ther. 2010;22:350–359.

[79] Johnston TE, Moore SE, Quinn LT, et al. Energy cost of walking in children with cerebral palsy: relation to the Gross Motor Function Classification System. Dev Med Child Neurol. 2004;46:34–38.

[80] Rose J, Gamble J, Burgos A, et al. Energy expenditure index of walking for normal children and for children with cerebral palsy. Dev Med Child Neurol. 1990;32:333–340.

[81] Bolster EAM, Balemans ACJ, Brehm MA, et al. Energy cost during walking in association with age and body height in children and young adults with cerebral palsy. Gait Posture. 2017;54:119–126.

[82] Thevenon A, Gabrielli F, Lepvrier J, et al. Collection of normative data for spatial and temporal gait parameters in a sample of French children aged between 6 and 12. Ann Phys Rehabil Med. 2015;58:139–144.

[83] Zbogar D, Eng JJ, Miller WC, et al. Movement repetitions in physical and occupational therapy during spinal cord injury rehabilitation. Spinal Cord. 2017;55:172–179.

[84] Lang CE, Macdonald JR, Reisman DS, et al. Observation of amounts of movement practice provided during stroke rehabilitation. Arch Phys Med Rehabil. 2009;90:1692–1698.

[85] Selye H. Stress and the general adaptation syndrome. Br Med J. 1950;1:1383–1392.

[86] Cunanan AJ, DeWeese BH, Wagle JP, et al. The General Adaptation Syndrome: a foundation for the concept of periodization. Sports Med. 2018;48:787–797.

[87] Siebert KL, DeMuth SK, Knutson LM, et al. Stationary cycling and children with cerebral palsy: case reports for two participants. Phys Occup Ther Pediatr. 2010;30:125–138.