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Utilizing Functional Electrical Stimulation for the Treatment of a Pediatric Patient with Cerebral Palsy: A Case Report

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Abstract

Background: Functional electrical stimulation (FES) treatment is becoming an increasingly studied intervention topic. To date, the majority of studies evaluate the use of FES with gait analysis, and there are few studies which utilized FES during cycling. **Intervention:** This case report evaluates FES treatment utilized for a five year old boy with quadriplegia spastic cerebral palsy. FES treatment was initially used in conjunction with the RT300 cycling machine, and we then had the child transition to performing treatment sessions on the RT600 elliptical stepper machine. **Outcome Measures:** We utilized outcome measures directly related to performance during the RT300 and RT600 treatment sessions. Our patient was able to obtain a small degree of power output, 0.11 Watts, by the end of our RT300 treatment, indicating some volitional contribution to the cycling. Average percentage of weightbearing increased from 17% to 31.5% over the RT600 treatment sessions, and average stimulation in microcoulomb was increased over both the cycling and elliptical stepper treatments. **Discussion:** There may be variations in response to FES treatment depending at which Gross Motor Function Classification System (GMFCS) level the patient is classified, as this is indicative of a spectrum of physical impairments. Additionally, future studies may find it beneficial to incorporate both FES-specific outcome measures as well as valid and reliable objective outcome measures. FES may serve as a tool for incorporating physical activity and resultant overall health benefits into the lives of patients with higher mobility impairments.

Keywords: Cerebral palsy; pediatrics; functional electrical stimulation; FES; physical therapy; rehabilitation

Background and Purpose

The complications involved in cerebral palsy (CP) arise due to an insult to the brain that is not progressive[1]. Individuals with CP may experience impairments such as spasticity, decreased strength, and difficulty with ambulation[1]. Functional electrical stimulation (FES) refers to electrical stimulation utilized during functional activities, such as cycling or gait analysis. Based on electrode placement, muscles are stimulated to contract at the correct time in correspondence with the movement patterns during the functional activity. There has been an increasing amount of research dedicated to the topic of FES utilized for the cerebral palsy population in the last several years. However, the majority of this research is focused on FES utilized during gait activities. The cerebral palsy subjects in these studies involving gait analysis are therefore at higher levels of function on the Gross Motor Function Classification System (GMFCS) than the patient in this case report, who we classified at GMFCS level IV. For example, in one literature review involving studies utilizing FES and gait analysis, all of the children with CP were classified at GMFCS levels I or II[1]. The following case report is beneficial in highlighting the application and progression of FES treatment for both a cycling machine and an elliptical stepper for a patient at GMFCS level IV, topics of which there is currently limited research.

Though there is limited research to document effectiveness of FES in improving muscle output or function in individuals with CP, there are several articles, primarily case reports or case series, that support its use[2,3,4,5]. These research articles served as references when assessing components of our case report. One research article utilizing cycling as the FES intervention evaluated power output in Watts as one of their outcome measures, as we do in this case report[2]. Additionally, that same study included cerebral palsy subjects who were unable to perform the FES cycling without assist from the motor in powering the cycling pattern, as was the case with our subject[2].

There are many challenges to progressing physical therapy in patients at higher impairment levels of cerebral palsy, such as decreased ambulation capabilities, spasticity, and limited resources to facilitate gains in muscular strength and neuromuscular control. There is a need for increased awareness of the importance of physical activity in all populations, despite level of impairment. Typically, for patients with higher impairment levels, the focus is on utilization of a power wheelchair and other adaptive resources. The following case report highlights the use of FES to augment muscle function, which is valuable in promoting physical activity and overall health in addition to facilitating improvements in musculature. The purpose of the following case report is to serve as an example of application and progression of FES utilized in conjunction with both cycling and an elliptical stepper for the cerebral palsy population. It will be particularly informative for pediatric cerebral palsy patients with increased impairment, indicated by higher GMFCS level.

Case Description: Patient History and Systems Review

An initial examination was performed on a young boy diagnosed with quadriplegia spastic cerebral palsy at GMFCS level IV. At this classification level, children generally utilize power mobility and need physical assist for the majority of transfers[6]. Depending on patient presentation, children may be able to ambulate short distances but require assist in this task[6]. Our patient utilizes a manual wheelchair and is able to independently propel himself but is not currently ambulating. In addition to cerebral palsy, this patient had a medical diagnosis of colpocephaly brain malformation. A diagnosis of colphocephaly indicates that the lateral ventricles remain as they were formulated in the fetus[7].

The patient was a twin born at 28 weeks through emergency cesarean section. He experienced intraventricular hemorrhage in his frontal lobe at birth, leading to intracranial bleeds that were classified as Grade 2. He was in the neonatal intensive care unit secondary to these medical complications until 12 weeks of age. Another medical comorbidity that he experienced was necrotizing enterocolitis and ultimately required a bowel resection. He had a history of plagiocephaly and torticollis, but plagiocephaly was successfully treated with helmet use. The patient also had a mild patent ductus arteriosis murmur present.

The patient received botulinum toxin injections to his bilateral hamstrings and calves at age 3 and then again 3 months later. Additionally, after the second round of botulinum toxin injections, bilateral short leg walking casts were applied to facilitate stretching and to increase supination of the feet. He then began physical therapy services at our facility (see more details on his initial evaluation below) and had a selective dorsal rhizotomy procedure performed about four and a half months after initial physical therapy examination. Selective dorsal rhizotomy is a procedure in which a portion of spinal dorsal rootlets are eliminated to decrease the amount of afferent input[8]. This procedure is primarily targeted at improving the amount of spasticity in cerebral palsy[8].

The patient received occupational therapy and speech language pathology services at the same time he underwent our physical therapy interventions outlined in this case report. The physical therapists providing his treatment felt that he would benefit from the use of functional electrical stimulation treatment as a result of his lower extremity weakness, impaired ambulation, and some residual spasticity after his selective dorsal rhizotomy procedure. His family's primary goal was to optimize his physical potential without pushing him too rigorously.

Examination

The initial physical therapy examination was performed when the patient was three years and six months old. In the initial examination, upper and lower extremity strength was assessed from a global and functional perspective. This is secondary to the patient's young age, developing cognition, and behavior interfering with the ability to perform formal manual muscle testing. Throughout the examination, strength appeared to be globally reduced, both in upper and lower extremities, during functional activities.

Gross motor skills were analyzed to determine the patient's level of independence with daily functional positions and transfers. Additionally, this analysis was performed to determine if there were any fundamental delays in progression of gross motor skills. The examination of gross motor skills was performed by subjective interpretation of the amount of assistance required by the physical therapist. The patient required maximal assistance to move from prone to quadruped and then required moderate to maximal assistance to maintain a quadruped position. He was dependent for maintaining both half kneeling and tall kneeling and in transitioning into and out of these positions. He was dependent in transferring from the floor to standing. Once standing, he required maximal assistance. He ascended stars with a step-to-step gait pattern with the right lower extremity leading and required maximal assistance for ascent. The patient was dependent in his ability to descend stairs. At initial examination, he was also dependent in transfers both into and out of his wheelchair.

Subjectively, the patient demonstrated fair static sitting balance but poor dynamic sitting balance. More standardized measures of balance, such as single leg stance and tandem stance, were not applicable for this patient as he was not able to stand without maximal assistance. While utilizing the gait trainer, he demonstrated the ability to ambulate forward, backward, and perform turns with coordination impairments. He demonstrated expression of his extensor tone while ambulating. He was able to independently self-propel his wheelchair in a straight direction and when turning right; he had difficulty with performing a left turn independently in the wheelchair.

For all of the examination findings outlined thus far, objective reliability and validity do not exist. This is due to the subjective nature of determining level of independence during transfers and positional changes and in performing gait and strength analysis through observation. We can hypothesize that there is likely variability between examiners as to the level of assist they feel they are contributing to transfers and positional changes. There also likely assists inter-rater variability in strength analysis. The analysis of gait is likely less subject to variability as ambulating stairs and ambulating with a gait trainer produce more objective results.

Tone was analyzed to determine the extent of hypertonia and the impact of hypertonia on upper and lower extremity functional movement. Hypertonia was present and graded as a 1 on the Modified

Ashworth Scale, with lower extremities exhibiting greater tone than upper extremities. According to Mutlu et al., interrater reliability of the Modified Ashworth Scale ranges between 0.61-0.87, and test-retest outcomes range between 0.36-0.83 in children with a diagnosis of spastic cerebral palsy, consistent with the patient in this case report[9]. These authors concluded that the Modified Ashworth Scale does not have high reliability in spastic cerebral palsy[9]. In a study evaluating validity of the Modified Ashworth Scale in identifying spasticity in children with cerebral palsy, the authors found this scale to have poor to fair agreement (K=0.24).[10] In this case report, the initial evaluation tests and measures were not re-assessed after the FES intervention was initiated.

Intervention

The physical therapy interventions for the first four months did not involve FES and were focused on improving the patient's ability to perform transfers and other functional activities with a decreased amount of assistance. He practiced bench sitting and reaching outside of his base of support, working to improve his dynamic sitting capabilities without loss of balance. Physical therapy interventions also worked on the patient's ability to step up on a four-inch curb with handrail assist as well as sit-to-stand transfers. Finally, transfers in and out of the wheelchair were included as part of the therapy sessions. The patient had a selective dorsal rhizotomy performed after four months of therapy and underwent extensive physical therapy treatment at another facility post-operatively. A month and a half following his selective dorsal rhizotomy procedure, the patient returned to our facility to resume outpatient physical therapy. He participated in seven months of physical therapy treatment working on similar interventions as outlined above, such as sit-to-stand transfers, tall kneeling, and standing with appropriate alignment of the lower extremities.

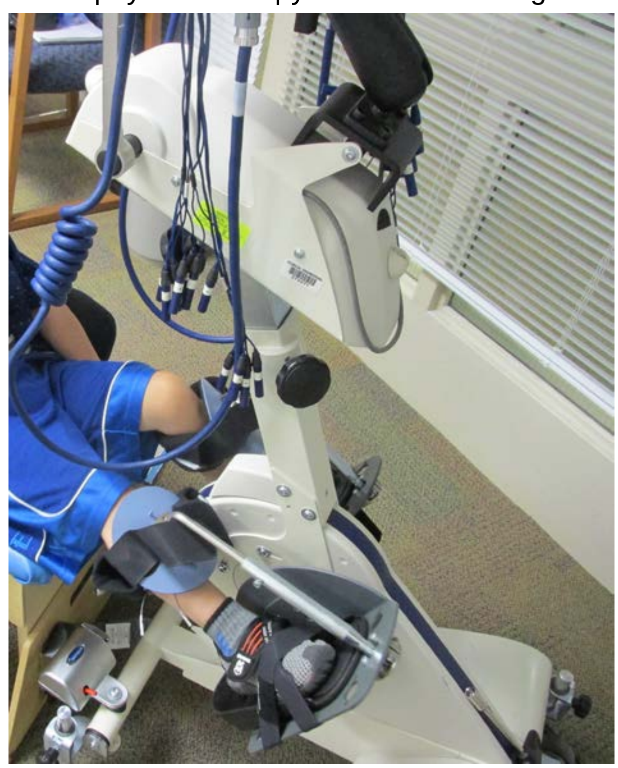

Figure 1: Patient set-up for the RT300 Cycling Machine.

FES treatment was initiated for the patient at four years and seven months of age. It is important to reiterate that the patient has not ambulated since undergoing selective dorsal rhizotomy, and our FES interventions were aimed at increasing strength to target this functional deficit. He had an FES session once per week and participated in his other physical therapy activities (without FES) once per week with another physical therapist. The RT300 cycling FES was utilized for 13 sessions with the patient. The RT300 consists of a stationary bike, and FES was delivered to our patient's lower extremities during the cycling movement. Electrodes were placed bilaterally on the quadricep, hamstring, gluteus maximus, tibialis anterior, and gastrocnemius and soleus musculature. We utilized two 6-channel stimulators to allow for stimulation of bilateral musculature. Testing was initially performed to determine the patient's tolerance to electrical stimulation; each muscle group was gradually and individually stimulated. The level of initial stimulation was set based on the patient's tolerance and response to an increase in amplitude for each muscle group. All of the FES parameters and stimulation levels were managed and saved through a portable electronic device called the SAGE controller.

For the first three RT300 FES sessions, the pulse width was set to 250 μsec. For the remainder of the FES sessions, the pulse width was decreased to 150 μsec. By decreasing the pulse width, the patient tolerated an increase in the stimulation of the musculature, measured in mA or microcoulomb. After decreasing the pulse width from 250 to 150, the amplitude of stimulation provided was increased at each treatment session except for one. In one of the sessions in which an increase in current amplitude occurred, the patient only tolerated an increase in stimulation to the quadriceps and hamstrings. For the remainder of the sessions in which stimulation was increased, he tolerated these adjustments at all electrode sites. The objective quantity of stimulation provided was not fully symmetrical between the right and left lower extremities or between muscle groups. A narrow pulse width utilized with an increased amplitude of stimulation recruits a greater number of motor neurons compared to sensory neurons[11]. For the RT300 treatment, the following parameters were consistent between sessions: Control speed set to 30 rpm and frequency set to 40 Hz. Resistance was set to 0.50 Nm for the first 11 sessions, 0.51 Nm for the 12th session, and 0.53 Nm for the 13th session (see Table 1). For tetanic contraction to occur in the musculature, a frequency of 30-40 Hz is necessary[11].

Additionally, a feature of the SAGE stimulator is that it is able to be used independently for electrical stimulation. With the same electrode placement we utilized on the RT300, we used the SAGE stimulator to provide stimulation for the patient as he performed sit-to-stand transfers. During these sit-to-stand transfers, the cables were still hooked up to the RT300 bike. With maximal assistance at bilateral buttocks and knees and stimulation at all electrode locations, the following parameters were utilized: 5 seconds of ramp-up time for the stimulation during which he performed sit-to-stand, 30 seconds standing, 5 second ramp-down of stimulation during which he transitioned from standing to sitting, and 10 seconds off to rest. The number of repetitions for sit-to-stand transfers varied between sessions.

After 13 sessions on the RT300, we had the patient transition to utilizing the RT600 elliptical stepper, which allowed for practice of the reciprocal gait pattern with a decreased percentage of weight-bearing compared to that which occurs during true ambulation. He has participated in 8 RT600 elliptical stepper treatments thus far (as of November 7, 2018) but is presently undergoing additional sessions. As in the RT300, two 6-channel stimulators were utilized for bilateral muscle groups. We again placed electrodes on bilateral quadricep, hamstring, gluteus maximus, tibialis anterior, and gastrocnemius and soleus musculature. Bilateral lower extremities were unweighted to a degree, though the patient was able to bear more weight through the lower extremities with continual practice on the RT600. The following treatment parameters were set consistently throughout the 8 treatment sessions: Frequency at 50 Hz, speed at 0.35 feet/sec, and pulse width at 150 μsec (see Table 2).

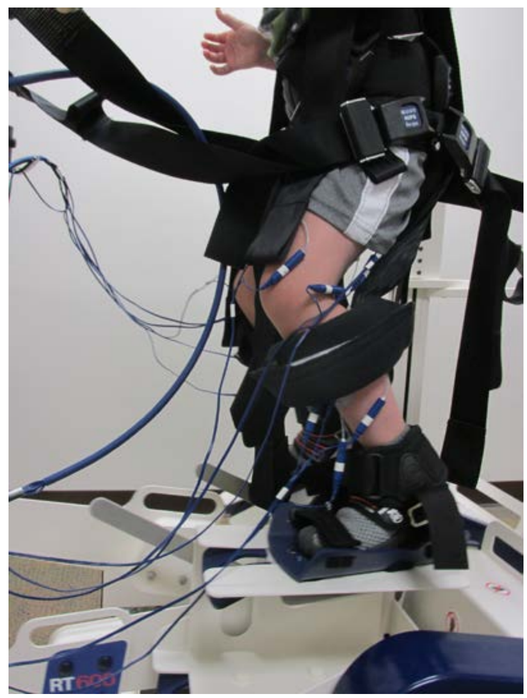

Figure 2: Patient set-up using the RT600 Elliptical Stepper.

With the RT300 and RT600, there is both a warm-up and a cool-down phase in which electrical stimulation is not applied. During both the warm-up and cool-down periods, the motor is responsible for moving the patient's legs through the range of motion at the set control speed (rpm) on the RT300 and at the target weight-bearing percentage on the RT600. An advantage of the warm-up period is that any potential spasticity can be resolved prior to the application of stimulation[11]. After the warm-up period, the stimulation intensity gradually increases to 100%, generally at a rate of 1% per second[11]. For the RT300 machine, our patient relied on motor support throughout the entirety of the FES session secondary to his inability to independently generate the necessary power output. Stimulation level is automatically adjusted by the machine throughout the FES session to ensure control speed and target weight-bearing are maintained in the RT300 and RT600, respectively[11].

Outcomes

The primary outcome measures that we assessed were directly related to parameters on the RT300 and RT600 machines (see Tables 1 and 2). We evaluated asymmetry on both machines, which refers to the extent to which the right lower extremity compared to the left lower extremity was contributing to the cycling or stepper movement. On the RT300, our patient's asymmetry was consistently at 0% because the motor was contributing significantly to the pedaling, as opposed to the patient utilizing volitional movement of the legs. The motor ensured that symmetrical pedaling occurred between the right and left lower extremities. Both right and left lower extremity are assigned a percentage after the cycling session to indicate what percentage each lower extremity was contributing to the pedaling. When utilizing the RT600 elliptical stepper, a positive asymmetry at the end of the session is indicative of greater contribution from the right lower extremity, whereas a negative asymmetry indicates greater contribution from the left lower extremity. The number quantifies what percentage the right or left lower extremity is contributing to the overall weight-bearing and movement pattern. In six of the sessions, our patient's left lower extremity was contributing more significantly, and in two of the sessions, the right lower extremity was dominant. At session three, our patient had the lowest degree of asymmetry, with the left lower extremity contributing only 2% more than the right lower extremity. Overall, for the RT600, asymmetry varied greatly between sessions and did not demonstrate consistent improvement with experience on the machine.

For the RT300, power measured in Watts was indicative of how much volitional movement the patient is providing into the cycling session, or how much power the patient can contribute to the crank[11]. The motor is designed to provide additional power output based on the objective power the patient is able to provide. Of note, it is difficult for young children to attain power output independently (pedaling the cycle on their own with decreased motor support) due to their small size and the heavy weight of the foot pedals. A study found that one of their subjects with cerebral palsy was unable to pedal without motor assistance and noted that this may be due to factors such as residual muscle tone and co-contraction of leg musculature while pedaling[2]. In that same study, they were able to analyze negative power output (Watts) to determine how much the motor was contributing and how much the participant was contributing when the power output was still not greater than 0 Watts[2]. Our patient's power output remained at 0.01 Watts for the first eight sessions. After this point and for the remaining cycling sessions, the average power trended upwards to a peak of 0.11 Watts at the last session. Of note, due to the inefficiency of RT300 cycling, the quantity of physiological power is 20 times the power output reading received after each session[11]. Though our patient's power output remained low for all 13 treatment sessions, our outcomes indicate that he was able to contribute slightly to the RT300 cycling movement with increased training. Therefore, we can hypothesize that he may have had improvements in muscle recruitment or force generation.

The stimulation measurement in μC on the RT300 and RT600 refers to the "charge per phase"[12]. Average stimulation intensity measured in microcoulomb increased from 2.30 μC to 4.46 μC over the 13 RT300 treatment sessions. For the RT600 intervention, the average stimulation increased from 2.30 μC to 4.85 μC over the 8 treatment sessions. These findings suggest that our patient was able to tolerate additional stimulation intensity at the musculature as he adapted to the treatment. The increase in average stimulation allowed him to experience greater resultant musculature contractions, facilitating more functional activation at each electrode site.

We were able to analyze distance traveled in miles for both the RT300 and RT600 interventions. For the RT300 and RT600, the distance traveled trended upward over the treatment sessions, although there was not a linear increase in mileage. This was likely a result of confounding factors, such as time restrictions during treatment sessions, as opposed to true reflection of the patient's tolerance to the treatment or endurance. Finally, for the RT600, we were able to analyze the amount of weight the patient was able to bear through bilateral lower extremities. Average weightbearing increased from 17% to 31.5% over 8 sessions, with a peak of 35% at session 6. This outcome is indicative of the patient having increased neuromuscular control to utilize lower extremities through a gait pattern, and if weightbearing continues to increase, it will likely translate to functional improvements in daily mobility.

One of the primary limitations of the outcome measures we utilized is that they were specific to either the RT300 or RT600 machine; therefore, there does not yet exist reliability or validity measures for these specific outcome measures. Though we did not formally re-assess the patient's initial examination findings after a given number of FES sessions, we were able to assess observed function based on sit-to-stand transfers and ambulation. For sit-to-stand transfers utilizing electrical stimulation, he required maximal assistance. The patient never ambulated after the dorsal rhizotomy, so we can hypothesize that he would be dependent in this task. An initial examination measure with published validity and reliability, such as the Gross Motor Function Measure (GMFM), has been utilized in other FES studies and would be beneficial in objective analysis of FES intervention[5].

Table 1: RT300 Intervention Parameters and Outcomes.

Session	Frequency (Hz)	Speed (feet/sec)	Average stimulation (μcoulomb)	Average asymmetry (%)	Distance traveled (miles)	Average weightbearing (%)
1	50	0.35	2.31	-25	0.04	17
2	50	0.35	2.13	-10	0.03	19
3	50	0.35	2.91	-2	0.07	25
4	50	0.35	3.33	-7	0.06	15
5	50	0.35	3.63	-15	0.08	25
6	50	0.35	3.93	46	0.09	35
7	50	0.35	4.41	-4	0.06	33
8	50	0.35	4.86	31	0.07	31.5

Table 2: RT600 Intervention Parameters and Outcomes.

Session	Pulse width (μsec)	Speed (RPM)	Resistance (Nm)	Frequency (Hz)	Average stimulation (μcoulomb)	Average asymmetry (%)	Distance traveled (miles)	Average power (Watts)
1	250	30	0.50	40	2.30	0	0.86	0.01
2	250	30	0.50	40	2.25	0	1.11	0.01
3	250	30	0.50	40	2.75	0	1.39	0.01
4	150	30	0.50	40	2.72	0	1.62	0.01
5	150	30	0.50	40	2.86	0	1.03	0.01
6	150	30	0.50	40	3.24	0	1.47	0.01
7	150	30	0.50	40	3.68	0	1.36	0.01
8	150	30	0.50	40	3.53	0	1.84	0.01
9	150	30	0.50	40	3.71	0	1.20	0.01
10	150	30	0.50	40	3.86	0	1.59	0.02
11	150	30	0.50	40	4.16	0	1.42	0.08
12	150	30	0.51	40	4.46	0	1.94	0.04
13	150	30	0.53	40	4.46	0	1.53	0.11

Discussion

The purpose of this case report was to present an example of the use of FES during cycling and elliptical exercise in a pediatric patient with cerebral palsy over a number of treatment sessions. Similar to Harrington et al.'s findings, our analysis of outcome measures focused on FES-specific improvements in performance while utilizing FES in conjunction with the cycling or stepper machines[2]. Our patient exhibited FES-specific improvements over time, such as ability to tolerate a continual increase in stimulation intensity, increased weight-bearing capability while utilizing the RT600, and progression to a small amount of power output on the RT300.

A limitation of this case report is that we did not utilize objective outcome measures with specific reliability and validity. Future cases may benefit from evaluating both FES machine-specific and reliable and valid outcome measures. For example, other studies have utilized outcome measures such as the Canadian Occupational Performance Measure and strength assessment through the use of dynamometry[3,13]. The difficulty of utilizing objective outcome measures in this case report was compounded by our patient being classified at GMFCS level IV, indicating more significant impairment. As previously mentioned, this patient had not ambulated since undergoing his selective dorsal rhizotomy procedure, so outcome measures which involve gait parameters would not be appropriate. Additionally, outcome measures such as dynamometry and ROM through the use of goniometry would have been difficult to obtain secondary to barriers, such as the patient's young age and developing cognition.

This case report serves as an example of application of specific FES parameters and settings. There is not yet research regarding which magnitude of parameters are most beneficial, but we were able to compare our intervention with other FES interventions discussed in literature. Harrington et al. utilized a pulse width range from 90-200 μsec during cycling, which is consistent with the majority (10 out of 13) of our RT300 treatment sessions[2]. In Johnston et al.'s case report utilizing FES during cycling, they set target speed at 40 rpm, pulse width at 250 μsec, and frequency at 33 Hz[3]. Some variability exists between our FES parameters and those used by Johnston et al. The main difference between Johnston et al.'s case report and our case report is that our patient was not able to contribute volitionally to movement of the bike and required motor support at all times[3].

There are several studies which outline positive outcomes of the utilization of FES interventions. Research for FES use during cycling primarily consists of case reports and studies with fewer subjects[2,3,4]. One case report on 12 weeks of FES cycling found improvement in strength of both the quadriceps and hamstring musculature post-intervention[3]. Additionally, the subject's Timed Up and Go (TUG) score decreased by 2.9 seconds, and his score on the SF-36 increased by 15.5 points, indicating improved quality of life[3]. Another case report found that, after FES and cycling, co-contractions of musculature present during cycling decreased[4]. In a study utilizing FES during gait in addition to traditional physiotherapy treatment in children with cerebral palsy, the authors found that GMFM-66 scores improved[5]. They also found an increase in strength generation of the tibialis anterior muscle, which was the only muscle stimulated in this study[5]. Our case report outlined similar support for FES intervention in that FES-specific improvements were noted.

Many of the studies done analyzing FES in the cerebral palsy population have found varying treatment responses with regard to differences in GMFCS levels. Trevisi et al. found that their subject at GMFCS-Expanded and Revised (GMFCS-E&R) level II had more numerous improvements, such as oxygen expenditure reduction during cycling, than the subject at GMFCS-E&R level III[4.] Therefore, for the cerebral palsy population, it is likely necessary to individualize FES treatment parameters and duration based upon initial degree of mobility impairment and level of functional independence. This is further supported by Harrington et al.'s study in which two of their participants required assistance from the motor for propulsion while cycling; these participants were both quadriplegic and at GMFCS levels III or IV[2]. The patient presented in this case report was also classified at a higher GMFCS level (level IV). He thus exhibited more significant mobility impairments at baseline, evidenced by his inability to produce power output on the FES cycling machine without motor assist. In order to receive optimal treatment benefits and enhance function in daily activities, our patient would likely benefit from increased frequency and duration of FES sessions.

This case report highlights that FES can be used even in a young patient with advanced impairments from CP. FES can augment other forms of physical therapy and occupational therapy and may provide a means to improve muscle activity during cycling and elliptical exercise. While clear functional gains were not necessarily documented, this case provides support for the use of FES to promote enhanced muscle function. Future studies are needed to determine if these improvements are sufficient to improve overall health and functional status over time. However, studies are now indicating that even small increases in physical activity are important for health benefits. Current physical activity guidelines recommend that children ages 3-5 should engage in physical activity throughout their day[14]. Additionally, children 6-17 years old should engage in 1 hour of physical activity each day[14]. FES may serve as a tool that assists children with limited mobility in achieving these physical activity requirements to optimize their health.

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