Volitional Cycling Augmented by Functional Electrical stimulation in Hemiparetic Adolescents: a Case Series Study

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Abstract—The aim of this work was to assess the feasibility of a treatment based on volitional cycling augmented by Functional Electrical Stimulation (FES) on hemiparetic adolescents.

Six chronic hemiparetic adolescents were included in a case series study. Patients underwent FES-cycling training combined with voluntary pedaling. The intervention consisted of 21 sessions lasting 30 minutes each. Patients were evaluated before, after training, and at a 3-month follow-up visit through clinical scales (Winter scale, observational gait scale, gross motor function measurement, Boyd test and Ashworth scale), a standard gait analysis and a voluntary pedaling test. Results were compared with an age-matched healthy control group (N=6).

Two subjects withdrew the study before the completion of the intervention. Concerning the four remaining subjects, the clinical scales showed a slight level of disability already at baseline and no changes were observed after the intervention. In terms of walking ability, some significant improvements (Kruskal-Wallis test, p-value<0.05) were obtained after training in two out of four subjects: an increase of about 16% and 41% of the ankle range of motion and of about 18% and 33% of the ankle propelling power were achieved for two subjects, respectively. During pedaling, the work produced by the paretic leg while pulling the pedal significantly increased in 3 out of 4 subjects. In one subject a more symmetrical cycling movement was observed, whilst for another subject a significant improvement in terms of co-contracture between rectus femoris and biceps femoris was achieved.

In conclusion, this study assessed the feasibility of FES-cycling training on hemiparetic adolescents, but did not provide evidences about the effectiveness of this intervention in improving motor recovery and walking ability. However, since only a small group of patients with a low level of disability was involved in the study, further investigations are needed to provide conclusive results.

Index Terms—Cycling, functional electrical stimulation, hemiparetic adolescents, rehabilitation.

I. INTRODUCTION

HEMIPARESIS is a one-side partial loss of motor function caused by a number of neurological diseases, such as stroke, traumatic brain injury, or cerebral palsy (CP).

People with hemiparesis are affected by muscle weakness and spasticity, resulting in an impaired motor and postural control. Gait is an essential motor function that requires highly integrated sensorimotor control systems. These systems are compromised in people affected by hemiparesis, causing long-term walking impairment. [1]. Thus, the restoration of walking ability is considered the main goal of neuro-motor lower limb rehabilitation [2].

Since the 1990s, Functional Electrical Stimulation (FES) has been used in the rehabilitation of hemiparetic subjects: the FES-induced afferent-efferent stimulation together with cutaneous and proprioceptive inputs seem to enhance the synaptic controls needed to produce a well-organized movement [3], [4].

Different studies demonstrated the effectiveness of FES in improving walking ability [5]-[7].

The kinematic pattern of walking is very similar to the one of cycling [8]: both are cyclical, require reciprocal flexion and extension movements and have alternating muscle activations of agonist/antagonist muscles in a well-timed and coordinated manner. This observation supports the hypothesis that the use of FES-induced cycling training in hemiparetic patients can improve walking ability as well as muscle strength and motor recovery in general. FES-cycling also guarantees repetitive and goal-oriented tasks that are recognized as fundamental elements of the motor relearning process [9]. Thus, FES-induced cycling training represents a functional, safe and widely accessible alternative to the FES-induced gait training for subjects with lower limb impairments.

Recent neuroplasticity studies have shown that the combination of FES and volitional effort might further enhance the therapeutic effects of FES as it can increase the activity in the cortical [10] and spinal [11] circuits enhancing both short and long term neuroplasticity.

As many studies have shown the effectiveness of FES to improve muscular strength and motor control for stroke...
adults patients [6], [12], little evidence is given for younger subjects [13], although the benefits shown in adults could be further enhanced in a younger population because of the greater plasticity and flexibility of their central nervous system [14]. Some studies have recently proposed the use of FES in children affected by cerebral palsy [13], [15], but up to now clear evidence has not been provided yet. A feasibility study of FES-induced cycling training was performed on five cerebral palsy adolescents [14]; the results of the study gave some evidence that the stimulation was well tolerated. A case series study on two hemiparetic children was also carried out [15]: the participants underwent 21 sessions of FES-cycling treatment and some improvements in terms of activation timing, symmetry of voluntary pedaling and gait strategy were obtained. However, up to our knowledge, no studies have investigated the effectiveness of FES-augmented voluntary cycling.

The present study aimed at investigating the feasibility and the effectiveness of a volitional cycling training augmented by FES in improving gait and lower limb motor function in four hemiparetic adolescents.

II. MATERIAL AND METHODS

A. Participants

Patients (Table I) have been recruited from the Neurological Institute “Carlo Besta” in Milan. Inclusion criteria were: age between 10 and 18 years, diagnosis of acquired hemiparesis, possibility of autonomous walk, lack of articular limitation, lack of pharmacological treatment with botulinum toxin or of surgery. No other rehabilitation programs were allowed during the study. All patients received an information sheet and family compliance was required. The research protocol was approved by the ethical committee of the Besta Neurological Institute.

<table>
<thead>
<tr>
<th>TABLE I. PARTICIPANTS' DETAILS AT BASELINE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, Gender (M/F), Height [cm], Weight [kg], Hemiparesis side (R/L), Etiology</td>
</tr>
<tr>
<td>S1 15 M 165 45 R CP</td>
</tr>
<tr>
<td>S2 14 M 170 90 R CP</td>
</tr>
<tr>
<td>S3 12 M 161 46 L CP</td>
</tr>
<tr>
<td>S4 17 M 175 63 R Ischemic Stroke (8 years old)</td>
</tr>
</tbody>
</table>

B. Intervention

All participants were trained 3 times a week, receiving a total of 21 sessions lasting 30 minutes each. Each session consisted of 5 minutes of passive pedaling at 40 revolutions per minute (rpm) on a motorized cycle-ergometer (warm-up phase), 20 minutes of FES synchronized with voluntary pedaling asking the patient to maintain a cadence of 40 rpm (the cadence kept by the motor was reduced to 20 rpm) and 5 minutes of passive pedaling at 40 rpm (warm-down phase). FES was delivered to gluteus maximus, quadriceps, hamstring and tibialis anterior of both legs. The RehaStim stimulator (Hasomed GmbH, Germany) together with the MOTOmed Viva2 ergometer (Reck GmbH, Germany) were used for the intervention. The stimulation angular ranges were shifted by 180° between the two legs (anti-symmetrical stimulation strategy) and were chosen in order to provide a biomimetic stimulation strategy [16].

C. Outcome measures

To assess the effectiveness of the treatment the subjects were tested before, after training and in a 3-month follow-up visit. In what follows, we referred to the three time assessment points as PRE, POST and FU.

The condition of the subjects was evaluated by means of some clinical scales. The first one was the Winter scale intended to investigate the functional deficit level during walking of CP subjects. It ranges from I (mild deficit) to IV (severe deficit). The Observational Gait Scale (OGS) provided another evaluation of the locomotion ability: it assesses the kinematic pattern with scores that ranges from -2 (severe deficit) to 22 (no deficit). The Gross Motor Function Measurement (GMFM) was used to assess the gross motor skills and ranges from 0 (no skills correctly performed) to 100 (all skills correctly concluded). The Boyd Ashworth scale (MAS) evaluated the tibialis anterior spasticity (0 if no spasticity occurred, 4 if irreducible spasticity was obtained).

To provide a quantitative evaluation of the walking ability, a gait analysis with the optoelectronic system ELITE 2000 (BTS Company SpA, 8 cameras, 100 Hz) integrated with a force platform was performed. The extracted outcome measures were: the gait velocity (V), the step length of the paretic and healthy leg (LPL and LH), respectively, a spatial symmetry index (SI) computed as the ratio between LPL and LH, the ankle dorsiflancetion angle, and the propelling ankle power.

All patients also performed a voluntary pedaling test [6]. This test consisted of 1 minute of passive cycling, followed by 2 minutes of voluntary pedaling during which the subject was asked to maintain a constant cadence of 30 rpm. A motorized cycle-ergometer customized with resistance strain gauges mounted at the two crank arms was used to measure the torque generated by each leg during pedaling [17]. The crank angle and the torque signals were sampled at 200 Hz. The active torque profiles during voluntary pedaling were estimated by subtracting the passive torques from the measured total torques. The overall work (WTOT) produced by each leg was computed as the integral of the active torque profiles mapped as function of the crank angle.

The unbalance (U) between the overall work produced by the two legs was computed for each of the i-th revolution as follows:

\[
U(i) = \left[ \frac{W_{TOT}^{HL}(i) - W_{TOT}^{PL}(i)}{W_{TOT}^{HL}(i) + |W_{TOT}^{PL}(i)|} \right] \%
\]

obtaining values ranging from 0% (identical works) to 100% (paretic work=0).

To provide a deeper analysis of the pedaling strategy, the overall work was divided into the work produced during knee extension (W_PUSH) and the one produced during knee flexion (W_PULL).
During the pedaling test, EMG signals of the rectus femoris (RF) and biceps femoris (BF) of both legs were also acquired. A multi-channel signal amplifier system working at 1 kHz (Porti32TM, TMS International BV, The Netherlands) was used. Bipolar surface electrodes were placed over the intended muscles, as recommended by the SENIAM (Surface Electromyography for the Non-Invasive Assessment of Muscles) [18].

After a standard EMG analysis (high-pass filter at 10 Hz, rectification, low-pass filter at 5 Hz) [6], the activation profile of the two muscles for each revolution was computed by means of a spline interpolation, thus obtaining the EMG envelope as function of the crank angle. Each of the obtained profiles were normalized with respect to their maximum peak.

EMG data were used to extract information about the co-contraction of RF/BF muscles during cycling [15]. The co-contraction index (CCI) of the i-th profiles was computed as follows:

$$ CCI(i) = \frac{S_{\text{overlap}}(i)}{S_{RF}(i) + S_{BF}(i)} $$

where $S_{RF}$ and $S_{BF}$ denote the surface area under each normalized EMG profile of RF and BF, respectively. $S_{\text{overlap}}$ is the overlap area of the two EMG profiles.

Normality ranges for the outcome measures related to the pedaling test were computed on an age-matched control group (CG) of 6 healthy volunteers (age 14.3±1.0 years, height 163.0±6.6 cm, weight 56.3±7.1 kg).

**D. Statistical Analysis**

On each subject a separate statistical analysis was performed. A Kruskal-Wallis test was used to compare the outcome measures of both gait analysis and pedaling test at the different time points. If a significant difference between PRE, POST and FU assessments was found (P<0.05), a post-hoc analysis was performed comparing pairs of tests (PRE vs POST, PRE vs FU and POST vs FU).

A Mann-Whitney U test was also performed to compare the results of the pedaling test obtained by each patient with the control group at the different time points.

**III. RESULTS**

Six male patients have been recruited and two of them withdrew the study before the completion of the intervention as the training required too effort for them and their family. Details of the 4 subjects that concluded the study are reported in Tab. I. Hemiplegic CP affects S1, S2 and S3 while S4 had a stroke at the age of 8 years. All participants were novel to cycling training and electrical stimulation.

**A. Clinical test**

Table II shows the results of the clinical evaluation during the PRE test. Only a slight disability can be observed for all subjects concerning the locomotion ability and the gross motor function. In terms of Winter scale and Modified Ashworth Scale, S4 showed the most impaired situation at baseline. After the treatment no changes occurred for all clinical scales.

<table>
<thead>
<tr>
<th>Winter Scale (I-IV)</th>
<th>OGS (2-22)</th>
<th>Boyd Test (0-4)</th>
<th>GMFM (100-0%)</th>
<th>MAS (0-4)</th>
</tr>
</thead>
<tbody>
<tr>
<td>S1</td>
<td>17-21</td>
<td>3-4</td>
<td>99.15%</td>
<td>2-0</td>
</tr>
<tr>
<td>S2</td>
<td>16-21</td>
<td>3-4</td>
<td>99.21%</td>
<td>1-0</td>
</tr>
<tr>
<td>S3</td>
<td>12-19</td>
<td>3-4</td>
<td>100%</td>
<td>2-0</td>
</tr>
<tr>
<td>S4</td>
<td>17-21</td>
<td>3-4</td>
<td>99.50%</td>
<td>3-0</td>
</tr>
</tbody>
</table>

*The scale ranges are reported from severe to mild deficit. PL and HL indicates the paretic and healthy leg respectively.

**B. Gait analysis test**

Tab. III summarizes the temporal-spatial parameters of the gait analysis. Mean values and standard deviation previously obtained on an aged-matched healthy subjects control group are also reported.

The mean velocity of all the patients did not significantly change over time and was maintained for all the subjects at about 1 m/s, a value within the normality ranges.

S1 was characterized by a similar step length between the two legs already at baseline (SI=1.0) that was maintained over time. Differently, S2, S3 and S4 showed a slightly asymmetrical gait pattern at baseline: for S2 and S3 LPL was bigger than LHL while for S4 LPL was bigger than LHL. During the POST test, S3 and S4 showed a more symmetrical gait pattern, although no statistical differences were obtained between PRE and POST tests. However, all these values were comprised in the normality ranges already at baseline.

**TABLE III. TEMPORAL-SPATIAL PARAMETERS OF GAIT ANALYSIS**

<table>
<thead>
<tr>
<th></th>
<th>V [m/s]</th>
<th>LPL [mm]</th>
<th>LHL [mm]</th>
<th>SI</th>
</tr>
</thead>
<tbody>
<tr>
<td>PRE</td>
<td>1.1 (0.1)</td>
<td>574.3 (7.5)</td>
<td>585.1 (2.0)</td>
<td>1.0 (0.0)</td>
</tr>
<tr>
<td>S1</td>
<td>1.1 (0.1)</td>
<td>587.5 (24.0)</td>
<td>587.3 (6.3)</td>
<td>1.0 (0.0)</td>
</tr>
<tr>
<td>FU</td>
<td>1.1 (0.2)</td>
<td>598.0 (25.1)</td>
<td>611.0 (71.8)</td>
<td>1.0 (0.1)</td>
</tr>
<tr>
<td>PRE</td>
<td>1.0 (0.0)</td>
<td>560.0 (9.0)</td>
<td>589.0 (16.5)</td>
<td>0.9 (0.1)</td>
</tr>
<tr>
<td>S2</td>
<td>1.0 (0.1)</td>
<td>554.5 (13.0)</td>
<td>600.3 (52.0)</td>
<td>0.9 (0.1)</td>
</tr>
<tr>
<td>FU</td>
<td>0.9 (0.0)</td>
<td>549.0 (24.1)</td>
<td>527.0 (10.8)</td>
<td>1.1 (0.0)</td>
</tr>
<tr>
<td>PRE</td>
<td>1.2 (0.0)</td>
<td>599.3 (16.5)</td>
<td>685.0 (13.0)</td>
<td>0.9 (0.0)</td>
</tr>
<tr>
<td>S3</td>
<td>1.1 (0.1)</td>
<td>632.5 (16.1)</td>
<td>628.3 (30.3)</td>
<td>1.0 (0.0)</td>
</tr>
<tr>
<td>FU</td>
<td>1.2 (0.0)</td>
<td>604.0 (28.1)</td>
<td>725.0 (24.8)</td>
<td>0.8 (0.2)</td>
</tr>
<tr>
<td>PRE</td>
<td>1.2 (0.0)</td>
<td>681.0 (21.5)</td>
<td>591.0 (19.0)</td>
<td>1.2 (0.0)</td>
</tr>
<tr>
<td>S4</td>
<td>1.2 (0.0)</td>
<td>646.5 (27.1)</td>
<td>620.3 (20.3)</td>
<td>1.0 (0.0)</td>
</tr>
<tr>
<td>FU</td>
<td>1.3 (0.0)</td>
<td>632.0 (16.1)</td>
<td>649.0 (5.5)</td>
<td>1.0 (0.0)</td>
</tr>
</tbody>
</table>

| CG | 1.3 (0.6) | 559.8 (47.9) | 549.4 (45.1) | 1.0 (0.2) |

Values represented as mean (standard values).

*, ** indicate pairs of tests with statistical difference (p<0.05) according to the post-hoc analysis of the Kruskal-Wallis test.
The ankle dorsi-plantar flexion angle (Fig. 1) of all the patients showed a reduced range of motion and an impaired peak of the plantar flexion angle (negative peak in the figure). Two of the patients (S1 and S4) significantly improved (p-value<0.05) the ankle range of motion after training (see Fig. 1), and maintained the result at FU. The analysis of the distal kinetics in the sagittal plane revealed slight deviations for three out of four subjects already at baseline (Fig. 2). Two subjects (S1 and S4) achieved a significantly increase of the power after training (relative peak-to-peak increase of 33% and 18%, respectively), but the improvements were not maintained at FU. Differently, the patient with the most impaired ankle propelling power (S2) did not show any significant improvements in terms of ankle power; however, the EMG analysis (data not reported) during gait showed a restoration of the physiological activation timing of the tibialis anterior at toe off, that were already functional for the other 3 subjects.

C. Pedaling test

At baseline, all the subjects showed a work produced by the healthy leg significantly higher than the one of the control group (see Fig. 3). For three of them (S1, S2 and S4), this was caused by an exaggerated production of work during the pulling phase of the pedals. All the patients maintained the values over time but S3, who reduced the work in the pushing phase.

The paretic leg of S3 and S4 showed a reduced W_TOT mainly caused by an impaired production of work in the pulling phase. W_PULL significantly improved after the treatment for both the patients and S4 maintained the result at FU. Also S1 and S2 showed a reduced W_PULL that, for S2, was significantly improved at POST test and maintained at FU.

The unbalance between works (see Fig. 3) was significantly higher than the control group for three out of four subjects (S2, S3 and S4). After the treatment a significant improvement was obtained for S3 that maintained the results at follow-up. An improving trend was highlighted also for S4 that showed a statistically significant difference only at FU.

Finally the analysis of the co-contraction index (Fig. 4) showed that for all the subjects the healthy leg had a co-contraction between the rectus femoris and the biceps femoris similar to the one of the control group. Differently, a higher level of co-contraction with respect to the control group was observed for the paretic leg muscles of two subjects (S2 and S3).

After training, S2 showed a significant improvement that was maintained at follow up (P-value<0.01), while S3 did not improve over time. Concerning S1 and S4, the CCI
values of the paretic leg were within the control group ranges already at baseline.

![CCI- Paretic leg](image)

*Figure 4: Comparison of the co-contraction index for the paretic leg of the four patients over time (PRE, POST and FU). Median and interquartile ranges (error bars) are shown. Green area indicates the control group values.

† indicates significant difference with Kruskal Wallis test (p<0.05)
* indicates significant difference with Mann-Whitney U test (p<0.05)

IV. DISCUSSION

A case series study assessing the effectiveness of a 7-week FES-augmented voluntary cycling training has been conducted on six hemiparetic adolescents.

The feasibility of the treatment was confirmed: all subjects correctly responded to the stimulation and well tolerated the treatment. Two patients withdrew the study, as the effort required was not compatible with their every-day life activities.

The preliminary results of the present study did not provide clear evidence about the effectiveness of the training in improving motor recovery and walking ability. Indeed, from a clinical point of view, no changes occurred over time. Concerning the performance during gait, some distal improvements were observed for S1 and S4 both in terms of ankle dorsi-plantar flexion angle and ankle propelling power. The pedaling test showed a significant increase of work produced by the paretic leg during knee flexion for S2, S3 and S4. Some improvements in terms of symmetry of the pedaling were also observed for S3 and S4. Finally, the training was effective in restoring a physiological co-contraction level between rectus femoris and biceps femoris for S2.

The slight variation in the biomechanical parameters observed during gait and pedaling were not translated into clinically significant improvements. However, it is important to underline that all of the participants were characterized by a slight disability already at baseline: all of the clinical scores were closed to the ceiling, thus limiting the possible effects of the intervention. Moreover, the gait velocity obtained by all the subjects was about 1 m/s, a value classified by Tilson and colleagues [19] as representative of people with no substantial walking limitations, thus confirming the slight impairment in terms of walking ability highlighted from the clinical scales. In the future, it might be interesting to investigate the effectiveness of the intervention on a group of patients with a higher level of impairment. Indeed, the subject with the most severe impairment at baseline (S4, Winter Scale II) was the only one who obtained a slight improvement both in terms of range of motion and propelling power at the ankle during walking and in terms of symmetry during pedaling.

Beyond the small group of patients recruited, another limitation of the study is the absence of a control group of patients performing a standard cycling training. However, in a previous study [20] performed on a group of 62 CP infants, no significant improvements were achieved with 30 sessions over 12 weeks of traditional cycling training.

The present work represents a preliminary step for the proper design of a randomized controlled study aimed at assessing the effectiveness of FES-augmented voluntary cycling training on hemiparetic adolescents. Since the training was safe and the instrumentation was relatively cheap, the treatment could be also delivered in a domestic setting, thus improving patients’ compliance and limiting the number of drop-outs.

The possibility to investigate the cortical changes induced by the training (e.g., through transcranial magnetic stimulation or functional-magnetic resonance imaging) could also provide new insights into the mechanisms of the neurological recovery.

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REFERENCES


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