The Progress of the Gait Impairment and Brain Activation in a Patient with Post-stroke Hemidystonia

Satoshi YAMAMOTO, PT, PhD1, Daisuke ISHII, OT, PhD2,3, Kyoko KANAE, PT4, Yusuke ENDO, PT, MSc5, Kenichi YOSHIKAWA, PT, PhD4, Kazunori KOSEKI, PT, MSc5, Ryo NAKAZAWA, PT4, Hanako TAKANO, PT4, Masahiko MONMA, RT, PhD6, Arito YOZU, MD, PhD2,7, Akira MATSUSHITA, MD8 and Yutaka KOHNO, MD, PhD2,8

1) Department of Physical Therapy, School of Health Sciences, Ibaraki Prefectural University of Health Sciences, Japan
2) Center for Medical Sciences, Ibaraki Prefectural University of Health Sciences, Japan
3) Department of Cognitive Behavioral Physiology, Chiba University Graduate School of Medicine, Japan
4) Department of Physical Therapy, Ibaraki Prefectural University of Health Sciences Hospital, Japan
5) Department of Physical Therapy, Faculty of Health Science, Health Science University, Japan
6) Department of Radiological Sciences, School of Health Sciences, Ibaraki Prefectural University of Health Sciences Hospital, Japan
7) Department of Rehabilitation, Ibaraki Prefectural University of Health Sciences Hospital, Japan
8) Department of Neurology, Ibaraki Prefectural University of Health Sciences Hospital, Japan

ABSTRACT. Objective: We explore the effects of body weight-supported (BWS) treadmill training, including the change of cortical activation, on a patient with post-stroke hemidystonia. Patient: The patient was a 71-year-old man with left thalamus hemorrhage. His motor symptoms indicated slight impairment. There was no overactive muscle contraction in the supine, sitting, or standing positions. During his gait, the right initial contact was the forefoot, and his right knee showed an extension thrust pattern. These symptoms suggested that he had post-stroke hemidystonia. Methods: The patient performed BWS treadmill training 14 times over 3 weeks. The effects of the BWS training were assessed by a step-length analysis, electromyography and functional magnetic resonance imaging (fMRI). Results: The patient’s nonparetic step length was extended significantly in the Inter-BWS (p<0.001) and Post-BWS (p=0.025) periods compared to the Pre-BWS session. The excessive muscle activity of the right gastrocnemius medialis in the swing phase was decreased at the Inter-BWS, Post-BWS, and follow-up compared to the Pre-BWS session. The peak timing difference of the bilateral tibialis anterior muscle became significant (p<0.05) on the first day of the intervention. The fMRI revealed that the cortical areas activated by the motor task converged through the intervention (p<0.05, family-wise error corrected). Conclusion: These results suggest that there was improvement of the patient’s symptoms of post-stroke hemidystonia due to changes in the brain activity during voluntary movement after BWS intervention. Body weight-supported treadmill training may thus be an effective treatment for patients with poststroke hemidystonia.

Key words: Post-stroke hemidystonia, Body weight-supported treadmill, Functional magnetic resonance imaging, Electromyography, Step length

Hemidystonia is a movement disorder that affects the upper or lower half of the body and is caused by abnormal muscle tone. The onset of post-stroke hemidystonia occurs during the chronic stage (range: 3 months to 3 years)11. The most commonly applied treatments for acquired hemidystonia are medication and deep-brain stimulation11. Physical therapy is also sometimes used in patients with acquired hemidystonia, but its benefits are often temporary, and there has been no large-scale double-blind study indicating that physical therapy has sufficient objective benefits to jus-
Yamamoto, et al.

Fig. 1. An MRI T1-weighted image at 13 years after the onset of the patient’s cerebral hemorrhage. White arrow: The lesion area in the left thalamus.

tify its regular application for acquired hemidystonia. Indeed, systematic reviews have concluded that there is insufficient evidence to recommend any particular strategy for acquired hemidystonia\textsuperscript{2-4}. Moreover, an effective intervention for gait disorder in post-stroke hemidystonia patients has not been identified. Case reports of post-stroke hemidystonia patients are thus a valuable resource for information about the success of alternative treatments.

The symptoms of hemidystonia are various. Some hemidystonia patients exhibit obvious symptoms when standing or walking but not at rest\textsuperscript{5}, suggesting that hemidystonia appears when the patient is bearing his or her body weight. Body weight-supported (BWS) treadmills partially support a patient’s body weight by means of an overhead harness, a pelvic belt, and thigh straps, allowing the patient to undergo gait training. Hesse et al.\textsuperscript{6} reported that the activity of the soleus muscle was diminished in patients with BWS training compared to those without BWS training. We hypothesized that BWS treadmill training could suppress the symptoms of post-stroke hemidystonia by reducing the weight-bearing burden of patients.

Functional magnetic resonance imaging (fMRI) is a promising modality for revealing the cortical activity of patients with neurological disorders. Two neurophysiological investigations of hemidystonia have been performed\textsuperscript{7,8}, and one of these studies revealed abnormal patterns of activity in both the ipsilesional and contralesional hemispheres of hemidystonia patients\textsuperscript{8}. We hypothesized that a BWS intervention would normalize the cortical overactivity and alleviate the symptoms of post-stroke hemidystonia. We conducted the present study (1) to explore the effects of BWS treadmill training in a patient with post-stroke hemidystonia by performing a gait pattern analysis and electromyography of the patient’s lower extremities, and (2) to investigate the changes in the patient’s brain activity after BWS training.

Case Presentation

A 71-year-old Japanese man (height: 175 cm; weight: 63 kg) was admitted to our hospital for rehabilitation. Thirteen years earlier, he had been diagnosed with a cerebral hemorrhage in the left thalamus. At that time, very slight hemiplegia of the right upper and lower limbs was observed, but he could walk without using a brace and could drive a car. Approximately 6 months before his presentation at our hospital, the muscle tone in his right upper and lower limbs increased and he developed an extension thrust pattern\textsuperscript{9} that became more pronounced, making it difficult to walk. His chief complaint at that time was that his speed of walking was greatly reduced, making it impossible to travel.

An MRI T1-weighted image is provided in Figure 1. The patient’s awareness was clear, and he had good recognition (Mini-Mental State Examination: 24 points). His motor symptoms showed slight impairment (Fugl-Meyer scores: lower limbs 29/34 points, upper limbs 32/36 points; grip strength: right 15.4 kg, left 34.5 kg). The patella tendon reflex and Achilles tendon reflex of the right side were normal. The patient’s range of motion was limited at right knee extension (−10°) and right dorsiflexion (0°). Sensory impairment in the right lower limb was moderate to severe. The patient could walk 100 m without a cane. Regarding activities of daily living (ADLs), his functional level was independent or supervised (functional independence measure: 118 points).

The observation of the patient’s dystonia revealed no abnormal movements or postures in the lower extremities, upper extremities, or trunk in the supine, sitting, or standing positions. During his gait, the initial contact of the right leg was the forefoot, and his right knee showed the extension thrust pattern (Fig. 2).

This study was carried out in accord with the Declaration of Helsinki, with approval from the Ethics Committee of the Ibaraki Prefectural University of Health Sciences (approval no. 797). Written informed consent was obtained from the patient for the publication and use of the images accompanying this case report.

Methods

Intervention

The patient underwent BWS treadmill training with a body-weight support system (Unweighing System 945-480; Biodex Medical Systems, Shirley, NY) and a treadmill (Autorunner AR-200; Minato Medical Science Co., Osaka, Japan). He had had no prior experience with treadmill training, including BWS training. To help him maintain a sym-
metrical gait pattern as long as possible, we set the following parameters in the first session: (1) body support equal to 20% of his body weight, and (2) gait speed of 0.7-1.2 km/hr. In each BWS session, two skilled physical therapists assisted the patient’s leg movements and supported his balance. In the intervention session, no ankle-foot-orthosis (AFO) was used. The patient’s fatigue was assessed during the intervention. The BWS was stopped if and when the Borg scale exceeded 13 or when the patient asked to stop due to fatigue.

Timeline

The patient underwent BWS treadmill training sessions 14 times over a 3-week period, for a duration of ≤20 min/session and no more than one session per day. We evaluated both the immediate and long-term effects of the BWS training. To determine the immediate effect of BWS training in this patient, we performed three evaluations on the first day of the intervention—namely, a step-length analysis and electromyography (EMG) assessment before (Pre-BWS), during (Inter-BWS), and after (Post-BWS) the BWS training.

Before starting the BWS treadmill training, the patient performed a treadmill gait trial with 20% body-weight support for approx. 1 min. The outcome data during the BWS training were from five gait cycles at from 25 sec to 35 sec after the start of the treadmill gait. The long-term effects of the BWS training were assessed by EMG and fMRI at 3 weeks (3 weeks) and 4 weeks (Follow-up) after the first day of the training. In the evaluation sessions, no AFO was used.

The patient also underwent conventional physical therapy (including overground gait training and ADL training) plus occupational therapy (i.e., training of the right upper arm using a hand-ergometer and ADL training) throughout the training period. He used a metal double-upright AFO on his right lower extremity. The AFO has two Klenzak joints set at 0° with plantarflexion rod stops and at 20° with dorsiflexion rod stops. There were no medication changes affecting the relief of the patient’s muscle tonus during the BWS training or during the patient’s evaluation for this report.

Outcome

Step lengths of the patient’s paretic foot and nonparetic foot

For the spatial quantification of the patient’s gait, we performed a step-length analysis using a video camera (frame rate: 60 Hz; HDR-CX 470, Sony Marketing, Tokyo) and analysis software (Kinovea, ver. 0.8.26). Gait event detection was performed using videographic techniques with visual detection. Visual detection was performed by direct observation of the video, without using any tools from the software. For event detection, the rater reported the video frame in which the foot first made contact with the floor after the swing phase (Initial contact), and the first instant of foot-off from the floor after the stance phase (Foot-off).

For step length analysis, the paretic step length (SL_paretic) (in cm) was defined as the distance between anterior-posterior positions (Fig. 3A; the walking direction is presented as a positive value) from the nonparetic initial contacts to the paretic initial contacts. The nonparetic step length (SL_nonparetic) was defined as the distance between anterior-posterior positions from the paretic initial contacts to the nonparetic initial contacts (Fig. 3B). When the patient’s nonparetic leg could not overtake the paretic leg, the nonparetic step length was negative (Fig. 3C). The SL_paretic and SL_nonparetic values were measured for five consecutive gait cycles.

Step-length asymmetry in the gait (SL_asymmetry) was defined as 100% × (SL_paretic − SL_nonparetic)/(SL_paretic + SL_nonparetic) for each walking cycle. An index of 0% indicates perfect symmetry; the magnitude represents the degree of asymmetry, and the sign indicates the direction of the asymmetry. That is, a positive index indicates a larger step length for the paretic leg during the paretic step. The mean and standard deviation of the SL_paretic, SL_nonparetic, and SL_asymmetry values were...
data was described previously. To calculate the %MVC data, the method using the 2-sec MVC was divided by the 100% MVC value (recorded each day) to calculate the %MVC (100% MVC). The iEMG data during movement were divided by a 2-sec EMG segment was selected and averaged for 2 sec time in order to calculate the integrated EMG (iEMG) in the 100 msec.

EMG patterns at the maximum voluntary contraction (MVC) of the TA and GM muscles were recorded for ≥5 sec at the first day and 3 weeks of the intervention, and at follow-up. Using visual inspection, the smallest variation of 100 msec, TE = 40 msec, flip angle = 80°, number of slices = 32, slice thickness = 3.2 mm, matrix = 64 × 64, voxel size = 3.4 mm³, total time = 5 min. The run began with three dummy volumes to allow for T1 equilibration effects; these volumes were subsequently discarded. The patient was scanned during the performance of two tasks that were performed based on a block design (task 30 sec, rest 30 sec); each task involved a different movement (right foot dorsiflexion, left foot dorsiflexion). Each task was performed in five blocks interspersed with rest blocks.

The image processing and statistical analysis were performed using the SPM12 program (ver. r7487; Wellcome Department of Cognitive Neurology, London, UK). First, to correct for dislocations caused by head motion, all images were realigned. The realigned images were then normalized to the Montreal Neurological Institute template brain supplied with SPM12. Finally, the images were smoothed using an 8-mm Gaussian kernel.

Statistical analysis
The effects of BWS treadmill training on the patient’s step length and the asymmetry on the days of the training period were assessed with a one-way repeated measures analysis of variance (ANOVA) with the time (Pre-BWS, Inter-BWS, Post-BWS) as a within-subject factor. Bonferroni’s correction was used for post hoc comparisons when the ANOVA revealed significant differences. The level of statistical significance was set at p<0.05.

For the iEMG and %gait cycle of the EMG peak amplitude data, we conducted a one-way ANOVA with the time (Pre-BWS, Inter-BWS, Post-BWS or the first day of the intervention, 3 weeks, or follow-up) as a within-subject factor. Holm’s correction was used for post hoc comparisons when the ANOVA revealed significant differences. When two peaks were observed in a single gait cycle, they were analyzed separately at the stance phase (during initial contact to foot-off) and swing phase (during foot-off to the next initial contact). The data were analyzed using R (ver. 2.8.1), a language and environment for statistical computing and graphics (http://cran.r-project.org/).

For the fMRI, a general linear model (GLM) analysis was performed to obtain average brain responses associated with the task. Regions of interest (ROIs) in the left hemisphere and their right hemispheric homologues were created using the Neuromorphometrics atlas of the sensory-
The Progress of Post-stroke Hemidystonia

Fig. 4. Examples of the nonparetic step length on the first day of the training period. A: Pre-BWS. B: Inter-BWS. C: Post-BWS.

Table 1. Step length and asymmetry on the first day of the training period

<table>
<thead>
<tr>
<th></th>
<th>Pre-BWS</th>
<th>Inter-BWS</th>
<th>Post-BWS</th>
</tr>
</thead>
<tbody>
<tr>
<td>SL_{nonparetic}, cm</td>
<td>-16.2 (0.7)</td>
<td>6.8 (2.8)*</td>
<td>-12.0 (1.4)*†</td>
</tr>
<tr>
<td>SL_{paretic}, cm</td>
<td>48.7 (1.3)</td>
<td>21.7 (1.1)*</td>
<td>43.9 (2.9)*†</td>
</tr>
<tr>
<td>SL_{asymmetry}, %</td>
<td>200.1 (9.6)</td>
<td>53.3 (16.6)*</td>
<td>175.3 (9.7)*†</td>
</tr>
</tbody>
</table>

Values are mean (SD). *p<0.05 vs. Pre-BWS. †p<0.05 vs. Inter-BWS.

SL_{nonparetic}: Nonparetic step length. SL_{paretic}: Paretic step length. SL_{asymmetry}: Step length asymmetry, defined as 100% \times (SL_{paretic} – SL_{nonparetic})/(SL_{paretic} + SL_{nonparetic}).

EMG

In the iEMG of the patient’s Rt-GM muscle in the stance phase, the EMG activity at Inter-BWS was lower than that at Pre-BWS (Fig. 5). The EMG activity at Post-BWS was higher than that at Pre-BWS. The iEMG of the Rt-GM muscle at Pre-BWS exhibited a peak at the right swing phase (Fig. 5). This signal was gradually decreased throughout the sessions (Pre-BWS > Inter-BWS > Post-BWS session). This signal was also decreased at 3 weeks after the first day of the training and during the follow-up session.

At the peak amplitude of the iEMG on the first day of the intervention, the results of the one-way repeated measures ANOVA for SL_{asymmetry} showed a main effect of the difference in the time (F(2, 12) = 200.27, p<0.001) (Table 1), and in the Bonferroni correction, the SL_{asymmetry} at Inter-BWS was significantly smaller than those at the Pre-BWS (p<0.001) and Post-BWS (p<0.001) periods. The SL_{asymmetry} at Post-BWS was significantly larger than that at Pre-BWS (p=0.033) (Table 1).

Results

On the first day of the intervention, the patient reported feeling fatigue (Borg scale: 12), and the intervention was discontinued after 12 min. The second and third intervention periods were each 18 min. The duration of the 4th to 14th interventions was 20 min.

The step lengths of the patient’s paretic foot and nonparetic foot

Based on the results of the one-way repeated measures ANOVA for the SL_{asymmetry} data, a main effect was observed for the difference in the time (F(2, 12) = 221.27, p<0.001) (Fig. 4, Table 1). In the Bonferroni correction, the SL_{asymmetry} value at the Inter-BWS period was significantly larger than those of the Pre-BWS and Post-BWS periods (both p<0.001). The SL_{asymmetry} value at the Post-BWS period was significantly larger than that at the Pre-BWS period (p=0.025; Fig. 4, Table 1). The one-way repeated measures ANOVA for the SL_{asymmetry} data revealed a main effect for the difference in the time (F(2, 12) = 275.36, p<0.001) (Table 1). In the Bonferroni correction, the SL_{asymmetry} value at Inter-BWS was significantly smaller than those at the Pre-BWS (p<0.001) and Post-BWS (p<0.001) periods (Table 1).

Motor area (precentral cortex, postcentral cortex, supplementary motor area, superior parietal lobe, supramarginal cortex, and angular gyrus). The numbers of activated voxels (showing activation above a threshold of p<0.05, family-wise error [FWE] corrected) were counted in the ROIs in each hemisphere.
Fig. 5. Integrated electromyographic (iEMG) results of the gastrocnemius medialis (GM) muscle and tibialis anterior (TA) muscle on the first day of the intervention, at 3 weeks, and at follow-up. White arrow: The right (Rt)-GM muscle during the mid-stance phase on the first day of the intervention. Black arrow: A peak of the right swing phase in the Rt-GM at Pre-BWS on the first day of the intervention. Solid line: Pre-BWS session. Dotted line: Inter-BWS session. Dashed line: Post-BWS session. %MVC: %muscle voluntary contraction. The timings of foot-off are indicated with white triangles for the Pre-BWS, filled triangles for the Inter-BWS, and diamond symbols for the Post-BWS session.
there was a main effect of time (F(2, 12) = 14.6, p<0.001).

The use of the Holm correction showed that the peak amplitude of the iEMG in the Lt-TA swing phase at Inter-BWS became significantly smaller than that at Pre-BWS (p=0.001). Similarly, the peak amplitude of the iEMG in the Lt-TA swing phase of the Post-BWS period became significantly larger than that at Inter-BWS (p=0.001).

At the peak timing of the iEMG on the first day of the intervention, regarding the results of the Rt-TA muscle, there was a main effect of time (F(2, 12) = 10.16, p=0.003) (Fig. 6, Table 2). In the Holm correction, the peak timing of the iEMG in the Rt-TA muscle of the Post-BWS period became significantly earlier than that at Inter-BWS (p=0.002). For the Lt-TA stance phase results, there was a main effect of time (F(2, 12) = 14.2, p<0.001). With the Holm correction, the peak timing of the iEMG in the Lt-TA stance phase of Inter-BWS became significantly earlier than that at Pre-BWS (p=0.023). Similarly, the peak timing of the iEMG in the Lt-TA stance phase Post-BWS became significantly later than that at Inter-BWS (p<0.001). Concerning the results of the Lt-TA swing phase, there was a main effect of time (F(2, 12) = 15.6, p<0.001). With the Holm correction, the peak timing of the iEMG in the Lt-TA swing phase at Inter-BWS became significantly earlier than that at Pre-BWS (p<0.001). Similarly, the peak timing of the iEMG in the Lt-TA swing phase of Post-BWS became significantly later than that at Inter-BWS (p=0.019).

At the peak amplitude of the iEMG on the first day of the intervention, at 3 weeks, and at the Follow-up, the results of the one-way repeated measures ANOVA for the Rt-TA muscle showed no main effect (F(2, 12) = 2.7, p=0.11) (Fig. 7, Table 3). In the Lt-TA stance phase results, no main effect of time was observed (F(2, 12) <0.1, p=0.94), whereas in the results of the Lt-TA swing phase, there was

Table 2. Peak amplitude and peak timing on integrated electromyography (iEMG) of the tibialis anterior (TA) muscles on the first day of intervention

<table>
<thead>
<tr>
<th></th>
<th>Pre-BWS</th>
<th>Inter-BWS</th>
<th>Post-BWS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rt-TA</td>
<td>iEMG (%MVC)</td>
<td>51.3 (14.2)</td>
<td>53.9 (14.1)</td>
</tr>
<tr>
<td></td>
<td>%gait cycle</td>
<td>56.3 (5.8)</td>
<td>65.2 (4.8)</td>
</tr>
<tr>
<td>Lt-TA stance</td>
<td>iEMG (%MVC)</td>
<td>71.0 (17.6)</td>
<td>30.6 (7.7)*</td>
</tr>
<tr>
<td></td>
<td>%gait cycle</td>
<td>15.8 (9.7)</td>
<td>2.2 (1.6)*</td>
</tr>
<tr>
<td>Lt-TA swing</td>
<td>iEMG (%MVC)</td>
<td>71.0 (9.6)</td>
<td>43.1 (5.3)*</td>
</tr>
<tr>
<td></td>
<td>%gait cycle</td>
<td>83.1 (3.6)</td>
<td>74.9 (1.4)*</td>
</tr>
</tbody>
</table>

Values are mean (SD). *p<0.05 vs. Pre-BWS. †p<0.05 vs. Inter-BWS.

Fig. 6. The iEMG of five gait cycles of the right and left TA muscles on the first day of the intervention. The peak amplitude of the TA is indicated with crosses or circles. A, D: Pre-BWS session. B, E: Inter-BWS session. C, F: Post-BWS session. The timings of foot-off are indicated with white triangles for the Pre-BWS, filled triangles for the Inter-BWS, and diamond symbols for the Post-BWS session.
The iEMG of gait cycles of the right and left TA muscles on the first day of the intervention, at 3 weeks, and at follow-up. Peak amplitudes of the TA are shown as crosses or circles. A, D: The first day of the intervention. B, E: 3 weeks after the first day of the intervention. C, F: follow-up. The timings of foot-off are indicated with white triangles.

**Table 3.** Peak amplitude and peak timing on integrated electromyography (iEMG) of the TA muscles on the first day of intervention, at 3 weeks, and at follow-up

<table>
<thead>
<tr>
<th></th>
<th>First day of intervention</th>
<th>3 weeks</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rt-TA</td>
<td>iEMG (%MVC)</td>
<td>51.3 (14.2)</td>
<td>44.0 (5.2)</td>
</tr>
<tr>
<td></td>
<td>%gait cycle</td>
<td>56.3 (5.8)</td>
<td>59.4 (5.4)</td>
</tr>
<tr>
<td>Lt-TA stance</td>
<td>iEMG (%MVC)</td>
<td>71.0 (17.6)</td>
<td>68.1 (17.0)</td>
</tr>
<tr>
<td></td>
<td>%gait cycle</td>
<td>15.8 (9.7)</td>
<td>20.6 (7.0)</td>
</tr>
<tr>
<td>Lt-TA swing</td>
<td>iEMG (%MVC)</td>
<td>71.0 (9.6)</td>
<td>67.9 (8.5)</td>
</tr>
<tr>
<td></td>
<td>%gait cycle</td>
<td>83.1 (3.6)</td>
<td>81.3 (1.5)</td>
</tr>
</tbody>
</table>

Values are mean (SD). *p<0.05 vs. the first day of the intervention. †p<0.05 vs. 3 weeks.

a main effect of time (F(2, 12) = 6.7, p=0.01). With Holm’s correction, the peak amplitude of the iEMG in the Lt-TA swing phase of the Follow-up became significantly larger than those at Pre-BWS (p=0.03) and 3 weeks (p=0.02).

At the peak timing of the iEMG on the first day of the intervention, at 3 weeks, and at the Follow-up, the results of the one-way repeated measures ANOVA for the Rt-TA muscle showed no main effect (F(2, 12) = 0.5, p=0.64) (Fig. 7, Table 3). In the Lt-TA stance phase results, there was no main effect of time (F(2, 12) <2.3, p=0.14), and in the results for the Lt-TA swing phase, there was also no main effect of time (F(2, 12) <2.6, p=0.11).

**Functional MRI**

For both the left and right dorsiflexion tasks, not only the lower-limb representation area of the motor cortex but also other areas (the left and right precentral cortex, left and right postcentral cortex, left and right supplementary motor area, left and right superior parietal lobe, left and right supramarginal cortex, and right angular gyrus) were activated at Pre-BWS (Fig. 8, Table 4). The total voxel count decreased throughout the study period: Pre-BWS > 3 weeks > Follow-up (Fig. 8, Table 4).
The Progress of Post-stroke Hemidystonia

The first day of the intervention

3 weeks

Follow-up

Rt-DF task

Lt-DF task

Fig. 8. Cortical activation by the left or right dorsiflexion task on the first day of the intervention, at 3 weeks, and at follow-up. The images at Z=64 mm above the anterior-posterior commissural plane are shown. The left side of the figure corresponds to the left “lesioned” hemisphere. The statistical threshold was set to the FWE-corrected p-value <0.05. Rt-DF: right dorsiflexion; Lt-DF: left dorsiflexion.

Table 4. Voxel counts of the cortical activation by the left and right dorsiflexion tasks in the ROIs on the first day of the intervention, at 3 weeks, and at follow-up

<table>
<thead>
<tr>
<th></th>
<th>Hemisphere</th>
<th>Baseline</th>
<th>3 weeks</th>
<th>Follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rt-DF task</td>
<td>Left</td>
<td>5,806</td>
<td>2,109</td>
<td>1,023</td>
</tr>
<tr>
<td></td>
<td>Right</td>
<td>958</td>
<td>1,073</td>
<td>373</td>
</tr>
<tr>
<td>Lt-DF task</td>
<td>Left</td>
<td>2,414</td>
<td>811</td>
<td>885</td>
</tr>
<tr>
<td></td>
<td>Right</td>
<td>1,570</td>
<td>1,082</td>
<td>811</td>
</tr>
</tbody>
</table>

Voxels counts were calculated above the statistical threshold that was set to the FWE-error-corrected p-value <0.05. Rt-DF: right dorsiflexion. Lt-DF: left dorsiflexion.

Discussion

Our findings for this patient with post-stroke hemidystonia demonstrated that (1) the patient’s gait pattern was modified immediately after the BWS treadmill training, and the modification pattern was maintained to the follow-up period; and (2) there was a change in the cortical activation in the sensory-motor area evoked by the voluntary movement after the BWS intervention period.

The iEMG of the patient’s Rt-GM muscle in the right mid-stance phase at Inter-BWS was decreased compared to that at Pre-BWS on the first day of the intervention. This result indicates that the patient’s excessive muscle activities for gait were suppressed by the body weight support that we provided. The step-length analysis revealed a significant extension of the nonparetic step length at Inter-BWS compared to the Pre-BWS session, suggesting an extension of the paretic stance phase. In addition, the patient’s gait symmetry was improved at Inter-BWS compared to the Pre-BWS session. These results suggest that the body weight support reduced the patient’s symptoms of post-stroke hemidystonia in his lower extremities.

Moreover, the significant extension of the patient’s nonparetic step length and the improvement of his gait
symmetry lasted until the Post-BWS session. The iEMG of the Rt-GM in the right mid-stance phase at Post-BWS was increased compared to that at Pre-BWS. In healthy subjects, the GM muscle must push off the ground in the stance phase. The reason why the iEMG of the patient’s Rt-GM in the right mid-stance phase at Post-BWS was increased compared to that at Pre-BWS on the first day of the intervention is unclear, because we did not measure the data of the trajectory of the center of gravity and the left-right ratio of the stance time. Presumably, the increase in the iEMG of the patient’s Rt-GM muscle during the mid-stance phase of the post-BWS compared to the pre-BWS was due to the increased weight bearing of the right lower limb and the increased activity of the Rt-GM (which is the antigravity muscle) as a result of the increased shift of the center of gravity toward the right side in the right stance phase.

In healthy subjects, there is no activity of the GM muscle in the swing phase. Jung et al. reported excessive GM muscle activity in a dystonia patient. In our patient, excessive Rt-GM muscle activity was observed at the Pre-BWS session. These results suggest that excessive GM activity in the right swing phase is one of the characteristics of post-stroke hemidystonia. Our patient’s excessive Rt-GM activity in the right swing phase was decreased at the Inter-BWS, Post-BWS, the 3-week time point, and the follow-up period compared to that at the Pre-BWS session, indicating that the BWS intervention improved the EMG activity pattern in the right swing phase.

The iEMG findings of the TA muscles in our patient demonstrated that the BWS training resulted in a change in the peak amplitude and timing on the first day of intervention. These changes did not occur steadily across time from the start to the end of the training period. These results imply that the carry-over effect of the TA muscle is not obtained before or after training.

Dystonic movements are associated not only with the cortico-basal-ganglia and cerebello-cortical motor network but also with overactivation in the sensorimotor cortex. Our patient’s fMRI results showed that the cortical areas activated by a motor task of left or right dorsiflexion were converged, which suggests that the converged fMRI activation patterns were related to the symptom improvement in this patient. Previous studies have shown that neural-activity convergence is caused by the progression of motor learning, which suggests that our patient’s excessive Rt-GM muscle activity was decreased through motor learning.

Several limitations of this study should be noted. First, the patient presented with motor and sensory impairments. Due to these impairments, the MVC measurement could not be performed stably. At the time of MVC measurement, an increase in the amplitude of myoelectric activity was observed, and the patient was able to exert some muscle strength. However, the activity of the GM muscles during gait exceeded 100% MVC. In previous reports on MVC in stroke patients, torque-measurement sensors were used to evaluate the accuracy of MVC measurements. In our present study, however, we did not measure torque, and thus the accuracy of the MVC value could not be verified. Therefore, the low reliability of the %MVC data is one of the limitations to the interpretation of EMG in this study. Second, we analyzed the EMG data during the BWS training from 25 sec to 35 sec after the start of the treadmill gait. Over this interval, no muscle fatigue was expected in this patient. Meyer et al. reported that healthy adults’ familiarization with treadmill walking requires 6 minutes; this finding indicates that our analysis time window was not long enough for the patient to become familiar with treadmill walking. These factors should be taken into consideration when interpreting the results of EMG data during BWS training. Third, we identified walking events using a video camera. Peterson et al. reported that there was an average difference of 60 ms between the identification of walking events using a video camera and the identification of walking events using the floor reaction force, and thus that the video camera was highly reliable for identifying walking events. Moreover, Peterson et al. used a video camera with a sampling frequency of 30 Hz, whereas our video camera had a sampling frequency of 60 Hz. Due to this higher time resolution, our analysis would have been more accurate than that of Peterson et al. Nonetheless, because we did not measure the floor reaction force or foot pressure using sensors, the accuracy of the identification of walking events in our study could not be verified. Fourth, the task used in the fMRI examination was ankle dorsiflexion movement, which is affected mainly by the portion of the primary motor area where the lower limbs are represented. In our patient, not only the muscles of the right lower extremity but also those of the right upper extremity were overactive due to dystonia. In the ankle dorsiflexion movement, activation was observed not only in the areas represented by the lower limbs in the primary motor area, but also across a wider region. In addition, the patient was undergoing arm training using a hand-ergometer as occupational therapy. These results suggest that the changes in brain activity that occurred in his case may have included effects other than those due to the BWS intervention.

Conclusion

Our patient with a chronic-stage thalamic hemorrhage performed 14 sessions of BWS treadmill training over a 3-week period. The patient’s gait pattern was modified immediately after the BWS treadmill training, and the modification pattern was maintained through the follow-up period. There was a change in the cortical activation in the sensory-motor area evoked by the voluntary movement after the BWS intervention period. These results suggest that there
was improvement of the symptoms of post-stroke hemidystonia due to changes in the brain activity during voluntary movement after the BWS intervention. Body weight-supported treadmill training may thus be an effective treatment for patients with post-stroke hemidystonia.

Conflict of Interest: The authors state that they have no conflicts of interest to disclose.

References