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Case Study

Improvement and sustainability of walking ability with hybrid assistive limb training in a patient with cerebral palsy after puberty: a case report

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Abstract. [Purpose] Cerebral palsy is one of the most common causes of childhood physical disability affecting motor development. Gait training with a wearable-robot, such as the Hybrid Assistive Limb, has been reported to improve gait ability in patients with chronic motor disabilities; however, there are no reports concerning the sustained improvement of walking ability with its use in patients with cerebral palsy. We present our observations for the use of Hybrid Assistive Limb gait training in a postpubescent cerebral palsy patient. [Participant and Methods] A 17-year-old male with spastic cerebral palsy could only ambulate slightly using a crouch gait posture and with the aid of a walker. Hybrid Assistive Limb training was performed thrice weekly for 4 weeks (total of 12 sessions) along with concurrent daily physical therapy. The follow-up period was 7 months after the intervention. [Results] The intervention resulted in improvements in the patient's gait speed, proportion of the stance phase in a gait cycle, step length, and the flexion angle of the knees at initial contact and during late stance phase, which was sustained for 7 months following the intervention. [Conclusion] Our observations suggest that Hybrid Assistive Limb training may effectively improve and sustain walking ability even among postpubescent cerebral palsy patients who have a decreased walking ability.

Key words: Cerebral palsy, Hybrid Assistive Limb (HAL), Sustainability of walking ability

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INTRODUCTION

Cerebral palsy (CP) is one of the most common causes of physical disability in childhood that affect motor development. Infants with CP can neither sit nor walk as early as other children¹). Among children with CP, it is difficult to predict their final gross motor ability owing to the individual variations in growth potential. Patients are generally classified at 2-4 years of age by using the Gross Motor Function Classification System (GMFCS), which is a five-level classification system proposed by Palisano et al²). This system focuses on their voluntary movements such as walking and sitting. Hanna et al.³) reported that gross motor function peaks at about 6 years of age, following which the gait parameters deteriorate with the increase in contractures and bony deformities, especially in non-ambulatory children. A crouched gait posture is one of the most common gait pathologies in patients with CP⁴). This posture, involving reduced hip and knee extension, is adopted by patients following puberty-related changes in the body segments, in order to counter gravity^{3, 5)}.

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Robotic devices were recently used to successfully rehabilitate a patient with CP⁶⁾. Similarly, there are reports regarding the efficacy of Lokomat[®] (Hocoma, Volketswil, Switzerland), which is a passively assistive robotic walking device⁷⁾. The hybrid assistive limb (HAL; Cyberdyne, Tsukuba, Japan) is a wearable robot suit that assists the user in voluntarily controlling hip and knee motion based on signals from force/pressure sensors in the shoes or even very weak bioelectric skin surface signals⁸⁾. Gait training with HAL has been reported to improve gait ability in patients with chronic motor disabilities, such as brain stroke⁹⁾, spinal cord injury¹⁰⁾, and cervical ossification of the posterior longitudinal ligament¹¹⁾. However, there are only few reports regarding its role in patients with CP. Matsuda et al. reported that HAL training resulted in an immediately observed effect on the hip and knee joint angle, although there was no significant improvement in gait speed and step length¹²⁾.

Herein, we present our observations of the improvements and sustainability in walking ability following HAL training in a patient with CP whose walking ability decreased following puberty.

PARTICIPANT AND METHODS

A 17-year-old male (height, 152.1 cm; weight, 54.6 kg) was born at 31 weeks of gestation and sustained intraventricular hemorrhage. At the age of five, when he was seen at Ibaraki Prefectural University of Health Sciences with a diagnosis of spastic diplegia due to CP at the first time, he could walk with double elbow crutches and was enrolled in the hospital's traditional rehabilitation program, so that he was classified as GMFCS level III. Although he was a little slow to talk, his intelligence was almost normal. As he grew taller and gained weight, he used a walking device and progressively adopted a crouch gait posture. His walking ability was minimal, and he was dependent on an electric wheelchair. Prior to the current intervention, his GMFCS level was IV (non-ambulatory).

As previously reported by Matuda et al.¹³, HAL walking training was performed thrice weekly for 4 weeks (total 12 sessions) along with concurrent daily physical therapy. We utilized HAL lower limb type S size (target 145–165 cm) with Cybernic Voluntary Control (CVC) mode (Fig. 1). The CVC mode of HAL can support the wearer's voluntary motion according to muscle activity⁸). Further, flexion/extension balance and assist torque at the hip and knee joints were optimized. To avoid falls, a walking device (All-in-One Walking Trainer, Ropox A/S, Naestved, Denmark) with a harness was used. Each 60-minute training session consisted of pre-training (10 min) including single-leg extension-flexion motion, standing and sitting exercise, time for wearing and removing the device (20 min), and time for HAL training (30 min).

Walking ability was assessed with motor analysis by using a commercial 3D motion capture system (Vicon Nexus, Oxford, UK) at 5 instances: before and immediately after the intervention (the next day), and 1, 4 and 7 months following the intervention. The patient was asked to walk at a self-selected walking speed using a walker and ankle-foot orthoses. Two to three trials were captured at each analysis, from which a representative trial was identified for subsequent analysis. Several gait parameters, including gait speed, proportion of gait cycle, step length and range of motion of hip and knee joint were assessed.

To examine the influence of the intervention on the patient's general health, we analyzed his heart rate and degree of fatigue by using a Borg scale. All clinically significant adverse events were recorded for every session.

The Human Ethics Review Committee of Ibaraki Prefectural University of Health Sciences approved this study (No. 682). Written informed consent was obtained from the patient's parents before this intervention was performed.



Fig. 1. Gait training by using Hybrid Assistive Limb.

RESULTS

The total gait training time during the intervention was 238 minutes (mean, 19.9 min per session). The mean degree of fatigue during the training, as measured on the Borg scale, was 11.9 (range, 11–13). There were no severe adverse events that interrupted the intervention.

The gait parameters are summarized in Table 1. Gait speed was higher than the pre-intervention values at all post-intervention assessments. The percentage of right stance phase and double support phase reduced following the intervention and never returned the pre-intervention levels. The changes in the left stance phase were variable, although its percentage was lesser at 1 and 7 months following the intervention. Bilateral step lengths were greater from the first post-intervention month, and remained so until the last follow-up visit at 7 months.

The joint kinematics are summarized in Table 2. The hip flexion angle at initial contact was found to be higher than the pre-intervention value for the right hip at assessment immediately following the intervention, and at the 4- and 7-month assessment; and for the left hip immediately following the intervention, and at 1- and 7-month assessment. The hip flexion angle in the late stance phase increased immediately following the intervention in both the hips, but subsequently decreased to pre-intervention levels at the next follow-up assessment. The arc of hip motion, defined as the difference of hip flexion angle between initial stance and late stance phase, was found to be lesser than the pre-intervention value immediately following the intervention in the right hip, and between 1 and 7 months in the left hip. The knee flexion angle at initial contact was lesser than the pre-intervention value for the right knee at

	Pre-training	Immediate post-training	1 month	4 months	7 months
Gait speed (m/min)	17.9	14.2	21.9	21.5	21.2
Percentage of gait cycle (%)					
Right stance phase	76	68	70	74	58
Left stance phase	77	80	76	78	70
Double support phase	53	36	36	40	48
Step length (m)					
Right	0.29	0.26	0.32	0.39	0.39
Left	0.30	0.21	0.41	0.33	0.31

Table 1. Gait parameters

Table 2. Joint kinematics: flexion is positive in hip and knee joint

	Pre-training	Immediate post-training	1 month	4 months	7 months
Hip flexion angle (°)					
Initial contact					
Right	27.9	50.7	22.3	34.2	30.9
Left	28.1	47.2	29.8	27.2	32.8
Late stance phase					
Right	-4.2	24.8	-5.8	-1.0	-0.9
Left	-4.6	20.3	-9.7	-11.2	-1.7
Arc of hip motion					
Right	32.1	25.9	28.1	35.2	31.8
Left	32.7	26.9	39.5	38.4	34.5
Knee flexion angle (°)					
Initial contact					
Right	29.6	43.5	22.2	18.6	22.9
Left	33.2	28.1	29.1	19.8	25.5
Late stance phase					
Right	26.3	25.7	17.7	23.7	14.0
Left	32.3	30.8	22.3	18.4	17.6

1-, 4- and 7-months following the intervention; and for the left knee immediately following the intervention, until 7 months later. The knee flexion angle during the late stance phase was found to be reduced immediately following the intervention and the change had persisted until the 7-month assessment for both the knees. The improvement in the knee joint kinematics following the intervention was sustained up to 7 months following the intervention.

DISCUSSION

This was the case report of a post-pubertal patient with CP in whom a sustained improvement in walking ability was noted following HAL training, without any adverse events.

The proportion of double support phase in the gait cycle and the hip flexion angle at initial contact improved immediately following the intervention. This suggests that the patient was able to move his legs forward with greater ease than before the intervention. Also, the arc of hip motion and the flexion angle of knee joint during the late stance phase appeared to have improved, along with improvement in the gait speed and step length. Endo et al. previously reported that improvement of muscle activity around the knee joint during the stance phase had good influence on the standing posture, so that the flexion angle of the hip was increased during the swing phase¹². Matsuda et al. observed improvements in the hip and knee joint angles immediately following a single session of HAL training¹⁴. However, they did not study if this improvement was sustained for a prolonged period of time.

A few previous studies reported that intensive physiotherapy resulted in an improvement in walking speed and stride length. Bower et al. concluded that in their trial for children aged 3 to 12 years with bilateral CP at level III to V on the GMFCS, the effectiveness of intensive physiotherapy on gross motor ability was very little. This advantage appeared to reduce over the subsequent 6 months if the physiotherapy reverted to conventional intensity¹⁵. In the current case, although the increase of hip flexion angle returned about initial value 7 months after, the decrease of knee flexion angle during stance phase affected improvement of step length. HAL training can provide proper walking image as well as changes in range of joint motion and result in sustainable walking ability for over 7 months in the post-pubertal patient with CP.

It is well known that real-time feedback improves gait and posture in patients with CP^{7, 9)}. Recently, Saita et al. reported on the role of biofeedback during HAL in stroke rehabilitation¹⁶⁾. According to this literature, HAL training in a patient with subacute stroke results in the cortical activation of the primary motor cortex of the ipsilesional hemisphere, and therefore leads to immediate improvement¹⁶⁾. In this case, interactive biofeedback may have influenced the patient's gait pattern and memory. Because the improvement in his walking ability persisted for 7 months following the intervention, it may be assumed that the voluntary motion induced by robotic-assisted rehabilitation remained in his memory.

Robotic assisted gait training has been described in a patient with CP, wherein a Lokomat device, a Gait Trainer GT1, and a CP Walker were used⁶). Lokomat uses a computer-driven exoskeleton to regulate and monitor gait parameters in individuals with compromised gait pattern⁷). However, the intensity of brain activity, which is the premise of motion, increases only during conditions that include voluntary motion planning, conducted exercise, and feedback, and does not change with passive exercise or simple repetitive automatic exercise¹⁷). In this case, our patient walked voluntarily by using the CVC mode, which may have helped him in gaining a proper walking pattern. The walking to move forward more positively than before obtained from the gait training was physically difficult to reproduce without HAL, and deterioration of walking parameters was observed at immediately following HAL training. Later, it is assumed that improvement and sustainability of walking ability and posture were obtained because the body responded to the acquired image of movement. Similar effect was reported in rehabilitation with HAL after spinal cord injury¹⁸).

The scope of our observations in this single case report is limited. It is difficult to find out that improvement has been achieved over time after the intervention and that the effect has been sustained by this case only. There is a need for large prospective studies with long-term follow-up of patients with CP in order to establish HAL training as a newer effective therapy. Such a study may also be able to determine the optimum session time, frequency, and total number of HAL interventions.

In conclusion, HAL training has the potential to result in sustained improvement in the walking ability of patients with CP even in cases that present after puberty.

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The authors declare that there is no conflict of interest regarding the publication of this article.

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