

Accepted Manuscript

Robot-assisted Gait Training in a Patient with Hereditary Spastic Paraplegia: A Case Report

Han Gil Seo, MD Byung-Mo Oh, MD, PhD Keewon Kim, MD



PII: S1934-1482(14)01382-3

DOI: [10.1016/j.pmrj.2014.09.008](https://doi.org/10.1016/j.pmrj.2014.09.008)

Reference: PMRJ 1345

To appear in: *PM&R*

Received Date: 17 April 2014

Revised Date: 5 September 2014

Accepted Date: 6 September 2014

Please cite this article as: Seo HG, Oh B-M, Kim K, Robot-assisted Gait Training in a Patient with Hereditary Spastic Paraplegia: A Case Report, *PM&R* (2014), doi: 10.1016/j.pmrj.2014.09.008.

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

Running head: Robot-assisted Gait Training on HSP

Robot-assisted Gait Training in a Patient with Hereditary Spastic Paraplegia: A Case
Report

Han Gil Seo, MD, Byung-Mo Oh, MD, PhD, Keewon Kim, MD

Department of Rehabilitation Medicine, Seoul National University College of Medicine,
Seoul National University Hospital, 101 Daehak-ro, Jongno-gu, Seoul 110-744,
Republic of Korea

Corresponding author:

Keewon Kim, MD, Department of Rehabilitation Medicine, Seoul National University
College of Medicine, Seoul National University Hospital, 101 Daehak-ro, Jongno-gu,
Seoul 110-744, Republic of Korea. Fax: +82-2-743-7473, Tel: +82-2-2072-0744, E-mail:

keewonkimm.d@gmail.com

1 **Robot-assisted Gait Training in a Patient with Hereditary Spastic Paraplegia: A Case**
2 **Report**

3

ABSTRACT

Robot-assisted gait training has been investigated for restoring walking through activity-dependent neuroplasticity in various neurological disorders. This case report presents the outcome of robot-assisted gait training combined with physiotherapy in a 28-year-old man with pure hereditary spastic paraplegia. The patient participated in 25 training sessions over 6 weeks. Improvements were noted in his walking speed and balance after the training. Gait kinematics and kinetics showed no remarkable changes before and after the training. Therefore, robot-assisted gait training may be useful for providing intensive gait training in patients with hereditary spastic paraplegia.

INTRODUCTION

Hereditary spastic paraplegias (HSPs) are a group of heterogeneous genetic disorders characterized by progressive distal axonopathy, mainly involving the corticospinal tract, and cause lower limb spasticity and weakness [1, 2]. Most patients present with difficulty walking or gait disturbance. Because no specific treatments exist for HSP, a symptomatic treatment approach is used [3]. Physical therapy is recommended for improving cardiovascular fitness, maintaining and improving muscle strength and gait, and reducing spasticity [4]. Anti-spasticity treatments, including oral medication, intrathecal baclofen, and botulinum toxin injection, also have been used in patients with HSP [3, 4]. However, these treatments provide short-term symptomatic improvement and gait worsen slowly over years due to advancing spasticity and general muscular deconditioning in most patients.

Robot-assisted gait training has been investigated for improving gait function in various

neurological disorders [5–8]. Theoretically, replicating a normal gait pattern using robot-assisted training restores walking through activity-dependent neuroplasticity of the spinal and supraspinal control mechanisms of walking [9]. Considering the markedly abnormal gait patterns of patients with HSP [10], intensive gait training using a robot may be beneficial for gait biomechanics and function in these patients.

To our knowledge, there has been no report on the effect of robot-assisted gait training in patients with HSP. This case report presents the outcome of robot-assisted gait training combined with physiotherapy in a patient with HSP.

Our hospital's institutional review board approved this case report, and written informed permission was obtained from the patient (IRB no: 1402-031-555).

CASE PRESENTATION

A 28-year-old man with pure HSP was diagnosed with SPG4 deletion (exon 5-17 and part of the *SLC30A6* gene) by a genetic study that used the multiplex ligation-dependent probe amplification technique. The patient had undergone bilateral Achilles tendon lengthening at 6 years of age and bilateral distal hamstring lengthening, rectus femoris transfer to gracilis, and gastrocnemius lengthening at age 26. Although he could walk without assistance using a single cane and bilateral ankle-foot orthoses (AFOs), his gait gradually deteriorated.

The patient's gait disturbance mainly stemmed from bilateral lower limb spasticity and weakness; muscle tightness was not apparent during the physical examination. According to the Medical Research Council (MRC) scale, hip flexor strength was grade 3, knee extensor and ankle dorsi-/plantar-flexor strength was grade 4, and knee flexor strength was grade 2. Spasticity of hip flexors, knee flexors, and knee extensors were 1+ bilaterally according to the

Modified Ashworth Scale (MAS). Ankle planar flexor spasticity was 3 on the MAS. Ankle clonus was positive on both sides, and he had a spastic gait with excessive lumbar lordosis. The patient had been taking baclofen (20 mg bid) to reduce spasticity and was performing home-based exercises regularly.

Because intensive treatment was expected to improve gait disturbance, the patient was provided a training program that involved robot-assisted gait training in combination with physiotherapy; this protocol was based on a previous meta-analysis on stroke patients [5]. The daily training session included 30 min of robot-assisted walking using an exoskeleton-type gait robot with partial body weight support (Walkbot_S; P&S Mechanics, Seoul, Korea), 30 min of gait training on the ground, and 30 min of upper extremity exercises with functional training. He underwent 25 training sessions during 6 weeks.

The gait robot replicated a normal gait pattern on a treadmill by controlling movements of the hip, knee, and ankle joints on both sides. The speed of the robot-assisted walking was increased from 1.2 to 1.8 km/h at each session. The amount of body weight support was gradually lowered from 25% to 0% during the course of the training sessions.

Clinical and functional assessments and gait analysis were performed 1 day before and 3 days after the training program. Clinical and functional assessments included the MRC scale for muscle strength, MAS, functional ambulation category (FAC), 10-meter walk test (10MWT, average of 3 trials), 6-minute walk test (6MWT), timed up and go (TUG), and Berg balance scale (BBS). The patient was instructed to use bilateral AFOs and a single cane while performing the standing and locomotor function tests.

Gait analysis was performed using a 3D optical motion capture system with EVa Real-Time software (Motion Analysis Corp., Santa Rosa, CA). Spatiotemporal parameters and kinetics

and kinematics of the hip, knee, and ankle joints in the sagittal, coronal, and transverse planes were measured while the patient walked barefoot along an 8-m walkway using a single cane at his comfortable speed.

Muscle strength of the lower extremities did not change, except in the knee flexors, which improved from grade 2 to 3 on the MRC scale after the training. The MAS scores were unchanged in hip flexors, knee flexors, and extensors on both sides; whereas, in both ankle plantar flexors, the MAS scores reduced from 3 to 2.

Functional assessments showed some improvements after training in walking speed and balance (Table 1). The patient's 10MWT and TUG results showed a decrease in time by 1.7 s (13.5%) and 3.3 s (13.9%), respectively. The patient's BBS showed 4 points (10.3%) of improvement. Alternatively, walking endurance measured using the 6MWT showed a slight decrease by 13 m (5%) after the training. FAC showed no change.

In the gait analysis, spatiotemporal gait parameters showed a decreased cadence, speed, step width and length, and duration of swing phase after the training (Table 2). Gait kinematics and kinetics showed no remarkable change before and after the training (data not shown). Severe gait abnormalities, including a compensated Trendelenburg gait pattern, excessive flexed position of the hip joints, and decreased knee and ankle range of motion, were observed in both analyses. Left hip extension moment during stance phase and hip rotation improved toward a normal pattern; whereas, pelvic obliquity and left hip abduction during left stance phase were slightly worse after the training.

DISCUSSION

To the authors' knowledge, this is the first case report that has examined the effect of robot-

assisted gait training on walking in a patient with HSP. Contrary to our expectations, no remarkable improvement in gait biomechanics was observed, despite some functional improvement. Nevertheless, the findings provide information about the effect and therapeutic target of physical therapy in patients with HSP.

In late-onset HSP (after age 6 years), gait disturbance usually worsens insidiously over many years [3]. Physical therapy has been used in patients with HSP for symptomatic improvement of lower extremity strength, gait, and cardiovascular condition. However, evidence of effectiveness is still lacking. Because it is difficult to alleviate gait disturbances in patients with HSP by using conventional physical modalities, novel therapeutic approaches deserve consideration.

Patients with HSP have markedly abnormal gait patterns caused by spasticity and weakness of the lower extremities [10]. Although the gait patterns vary among patients, abnormal patterns include lower gait velocity, shorter step length, larger step width, decreased knee range of motion, and excessive hip flexion. We expected that robot-assisted gait training would improve abnormal gait pattern by replicating a normal gait pattern. However, the findings suggest that it is challenging to ensure activity-dependent neuroplasticity, which is a postulated mechanism of robot-assisted training in stroke and spinal cord injuries, in patients with HSP. The difference may result from the pathophysiology of HSP, a length-dependent distal degeneration of axons in the corticospinal tract [2], rather than a focal injury of the central nervous system. Some of the functional improvements observed in our patient may have resulted from compensation to intensive training.

One point that should be considered is the number of training sessions. Twenty-five training sessions over 6 weeks, in our case, were relatively less than 24–60 sessions over 8–16 weeks, as used in previous studies on the effects of locomotor training after incomplete spinal cord

injury [6]. Therefore, the amount of training in our study may not have been enough to induce neuroplasticity. However, a recent study reported that 12 sessions of robot-assisted gait training over 4 weeks improved speed and functional mobility in a certain group of patients with spinal cord injuries [11]. The essential components required for locomotor training, such as timing, intensity, frequency, and duration, are still a matter of debate in patients with spinal cord injury.

Of note, in this study, the robot-assisted gait training was performed without AFOs, while balance and locomotion tests were performed with the AFOs to simulate gait function in real life. Using AFOs, the tests may not reflect changes in the kinetics or kinematics of the ankles. Nevertheless, functional tests showed clinically meaningful changes after the training. The improvement in the gait speed, TUG, and BBS were clinically significant when the threshold of minimal detectable change (MDC) for elderly and neurological conditions were referenced (gait speed, 0.06 m/s to 0.29 m/s; TUG, 2 s to 5 s; and BBS, 2 points to 5 points) [12]. Meanwhile, the MDC in the 6MWT was 20 m to 106 m; thus, a decrease of 13 m in our patient was not clinically significant [12]. To supplement the results of functional tests, gait analysis was performed without AFOs before and after the training. However, the trajectories and moments of the ankle did not show remarkable changes.

Although robot-assisted gait training has been used mainly in patients with stroke or spinal cord injury, it has been recently used in patients with Parkinson disease, a degenerative neurologic disorder [7, 8]. Although the results were controversial, some studies reported improved walking ability and balance after robot-assisted gait training compared to that with conventional physical therapy [7, 8]. It was suggested that not only reinforcing neural circuits but also reconditioning through the augmented physical activity was a plausible explanation for the effect of robot-assisted training [8]. The current study's findings indicate that

functional improvement without a definite change in gait parameters may be because of the intensive physical activity during robot-assisted gait training. Therefore, improving the functional aspects of gait via intensive training may be an appropriate goal for gait training in patients with HSP.

CONCLUSION

This is the first case report that presents the effect of robot-assisted gait training in a patient with HSP. Gait biomechanics did not improve despite some functional gain in gait speed and balance. These findings suggest that in patients with HSP, improvement in functional, rather than abnormal gait parameters, should be considered as the appropriate therapeutic target. Robot-assisted gait training may be useful for providing intensive gait training in patients with HSP.

References

1. Salinas S, Proukakis C, Crosby A, Warner TT. Hereditary spastic paraplegia: clinical features and pathogenetic mechanisms. *Lancet Neurol* 2008;7:1127-1138.
2. Blackstone C, O'Kane CJ, Reid E. Hereditary spastic paraplegias: membrane traffic and the motor pathway. *Nat Rev Neurosci* 2011;12:31-42.
3. Fink JK. Hereditary spastic paraplegia. *Curr Neurol Neurosci Rep* 2006;6:65-76.
4. Fink JK. Hereditary Spastic Paraplegia Overview. In: Pagon RA, Adam MP, Bird TD, Dolan CR, Fong CT, Stephens K, editors. *GeneReviews(R)*. Seattle (WA); 2000 [updated 2014].
5. Mehrholz J, Elsner B, Werner C, Kugler J, Pohl M. Electromechanical-assisted

training for walking after stroke. *Cochrane Database Syst Rev* 2013;7:CD006185.

6. Morawietz C, Moffat F. Effects of locomotor training after incomplete spinal cord injury: a systematic review. *Arch Phys Med Rehabil* 2013;94:2297-2308.

7. Picelli A, Melotti C, Origano F, Waldner A, Gimigliano R, Smania N. Does robotic gait training improve balance in Parkinson's disease? A randomized controlled trial. *Parkinsonism Relat Disord* 2012;18:990-993.

8. Picelli A, Melotti C, Origano F et al. Robot-assisted gait training in patients with Parkinson disease: a randomized controlled trial. *Neurorehabil Neural Repair* 2012;26:353-361.

9. Behrman AL, Bowden MG, Nair PM. Neuroplasticity after spinal cord injury and training: an emerging paradigm shift in rehabilitation and walking recovery. *Phys Ther* 2006;86:1406-1425.

10. Klebe S, Stolze H, Kopper F et al. Gait analysis of sporadic and hereditary spastic paraplegia. *J Neurol* 2004;251:571-578.

11. Hubli M, Dietz V. The physiological basis of neurorehabilitation--locomotor training after spinal cord injury. *J Neuroeng Rehabil* 2013;10:5.

12. Steffen T, Seney M. Test-retest reliability and minimal detectable change on balance and ambulation tests, the 36-item short-form health survey, and the unified Parkinson disease rating scale in people with parkinsonism. *Phys Ther* 2008;88:733-746.

Fig. 1. Gait kinematics and kinetics in the (A) sagittal, (B) coronal, and (C) transverse planes before and after the training program. The red and blue lines represent gait parameters before and after the training, respectively. Green shadow indicates the normal range of each parameter.

192 Table 1. Outcomes of the functional assessments before and after the training program

Functional assessments	Before	After
FAC	3	3
10MWT* (sec)	12.6	10.9
6MWT (m)	249	236
TUG (sec)	23.8	20.5
BBS	39	43

193 *Mean value of 3 trials. The patient used bilateral ankle-foot orthoses and a single cane
 194 during the tests. FAC, functional ambulation category; 10MWT, 10-meter walk test; 6MWT,
 195 6-minute walk test; TUG, timed up and go; BBS, Berg balance scale.

196

197 Table 2. Spatiotemporal gait parameters before and after the training program

Gait parameters	Before	After
Cadence (steps/min)	45.8	39.0
Speed (cm/s)	33.7	22.9
Step width (cm)	15.4	13.8
Step length (cm)		
Right	43.2	32.5
Left	46.4	36.2
Step time (s)		
Right	1.34	1.52
Left	1.28	1.55
Swing (% cycle)		
Right	30.3	27.4
Left	27.0	20.9

198

199