Research Article

The voluntary driven exoskeleton Hybrid Assistive Limb (HAL) for postoperative training of thoracic ossification of the posterior longitudinal ligament: a case report

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Context: The hybrid assistive limb (HAL) is a wearable robot suit that assists in voluntary control of knee and hip joint motion by detecting bioelectric signals on the surface of the skin with high sensitivity. HAL has been reported to be effective for functional recovery in motor impairments. However, few reports have revealed the utility of HAL for patients who have undergone surgery for thoracic ossification of the posterior longitudinal ligament (thoracic OPLL). Herein, we present a postoperative thoracic OPLL patient who showed remarkable functional recovery after training with HAL.

Findings: A 63-year-old woman, who could not walk due to muscle weakness before surgery, underwent posterior decompression and fusion. Paralysis was re-aggravated after the initial postoperative rising. We diagnosed that paralysis was due to residual compression from the anterior lesion and microinstability after posterior fixation, and prescribed bed rest for a further 3 weeks. The incomplete paralysis gradually recovered, and walking training with HAL was started on postoperative day 44 in addition to standard physical therapy. The patient underwent 10 sessions of HAL training until discharge on postoperative day 73. Results of a 10-m walk test were assessed after every session, and the patient’s speed and cadence markedly improved. At discharge, the patient could walk with 2 crutches and no assistance. Furthermore, no adverse events associated with HAL training occurred.

Conclusion: HAL training for postoperative thoracic OPLL patients may enhance improvement in walking ability, even if severe impairment of ambulation and muscle weakness exist preoperatively.

Keywords: Ambulation difficulty, Ossification of the posterior longitudinal ligament of the spine, Postoperative procedures, Recovery of function, Robotics.

Introduction

The hybrid assistive limb (HAL) is a wearable robot suit that assists in voluntary control of knee and hip joint motion (Fig. 1). Signals from force-pressure sensors in the shoes and muscle action potentials detected through electrodes on the surface of the skin are processed through a computer and assisted motions are provided to the patient. Power units on the hip and knee joints on both sides consist of angular sensors and actuators, and the control system consists of a cybernic voluntary control and a cybernic autonomous control subsystem.¹

HAL has been reported to be useful in the functional recovery of various mobility disorders.²⁻⁵ Studies have shown successful outcomes for mobility disorders in the chronic phase or maintenance phase, but there are...
few reports on the use of HAL in the acute phase or early postoperative period.6,7 Therefore, the efficacy of HAL in the acute phase or early postoperative period remains unclear.

In the current case report, HAL training was performed in addition to standard physical therapy, and physical function and walking ability were evaluated before and after HAL training. We report the feasibility, safety and the recovery course of HAL training in the acute phase after surgery for thoracic ossification of the posterior longitudinal ligament (thoracic OPLL). To the best of our knowledge, only one case report has reported on the use of HAL in patients who have undergone surgery for thoracic OPLL.7 Sakakima et al. reported on the feasibility of early HAL training after surgery and its ability to enhance motor recovery of patients with residual paralysis after surgery. Patients with thoracic OPLL commonly present with myelopathy and muscle weakness of the proximal leg muscles, which leads to severe gait impairment.

Herein, we present a postoperative thoracic OPLL patient who showed remarkable functional recovery after training using HAL, although the paralysis was re-aggravated after the initial postoperative rising, requiring 3 weeks of bed rest.

Case presentation
A 63-year-old woman became aware of sensory abnormality in both legs 2 months before surgery. One month before the surgery, her gait disturbance became pronounced and she required a walking stick. Two weeks before the surgery, paralysis and strong numbness in both legs suddenly appeared and she was unable to walk. She was taken by ambulance to a local hospital, and incomplete paraplegia caused by thoracic OPLL was diagnosed. She was subsequently moved to our institute for surgery.

The neurologic examination on admission revealed muscle weakness with a manual muscle testing (MMT) score of 3/2 in the iliopsoas muscle and an MMT score of 4/4 in the quadriceps femoris, tibialis anterior, gastrocnemius, and hamstring muscles. The patient had severe sensory disturbance (touch, pain, temperature, and position) and severe numbness in both the legs in the portion below the inguinal region. There was no urinary bladder or bowel function disturbance. The results of the blood and urine tests were normal.

Computed tomography after myelography showed OPLL extending from T3 to T7, discontinuous ossification at T4/5, and vertebral fracture of T5 without posterior wall damage (Fig. 2). Magnetic resonance imaging showed strong anterior spinal cord compression at the T4/5 level, and the caudal portion of the T5 vertebral body showed low intensity on both T1- and T2-weighted images (Fig. 3). The estimated amount of intraspinal canal ossification was 70%.

Preoperative clinical evaluation showed that the Japanese Orthopaedic Association (JOA) score, excluding the upper extremities, was 5.5/11 (0-1, 5-1-3); the spinal cord independence measure score (SCIM-score)8 was 57 points (self-care: 6/20 points; respiration and sphincter management: 37/40 points; and mobility: 14/40 points); the American Spinal Injury Association (ASIA) impairment scale (AIS) was grade D; the ASIA motor score (lower limb total) was 33 points (right: 18 points; left: 15 points); the ASIA sensory score for light touch was 86 points (right: 43 points; left: 43 points); the Walking Index for Spinal Cord Injury II (WISCI II)9-11 was 8 points; and the Frankel classification was grade C.

We speculated that intense stress at the discontinuous ossification caused the vertebral fracture and local instability led to the compression of the spinal cord.

Posterior decompression and fusion (laminectomy for T3 to T7, and pedicle screw fixation and posterolateral fusion for T1 to T9) were performed (Fig. 4). The operation took 6 hours and 35 minutes, and the estimated blood loss was 280 mL. Intraoperative ultrasonography showed pulsation of the dural sac and favorable posterior decompression; however, the anterior compression of the OPLL remained. Intraoperative motor-evoked potential monitoring showed no change in amplitude.

The extradural drainage catheter was removed on postoperative day 2 and rehabilitation was started on postoperative day 5. On postoperative day 7, weakness...
in both the legs suddenly occurred while the patient was undergoing parallel-bar walking training. A neurologic examination revealed muscle weakness with an MMT score of 0-1/0-1 in the iliopsoas, quadriceps femoris, tibialis anterior, gastrocnemius, and hamstring muscles. The sensory disturbance in both the legs also worsened at the same time; however, numbness was slight. Plain radiographs and computed tomography scans showed appropriate laminectomy and pedicle screw fixation, and magnetic resonance imaging showed that the spinal canal was successfully decompressed and that compressive lesions such as an epidural hematoma were absent. We diagnosed the aggravation of muscle weakness to be due to residual compression from the anterior lesion and microinstability after the posterior fixation and prescribed bed rest for a further 3 weeks. The incomplete paralysis gradually recovered to an MMT score of 3/3, and after resuming rehabilitation, the patient was able to stand and walk again using the parallel bars.

Walking training with HAL started on postoperative day 44 in addition to standard physical therapy. Standard physical therapy was performed every weekday for 40 minutes and consisted of sitting position training and walking training using parallel bars with the assistance of a physical therapist. At the initiation of HAL training, the robot was fitted and sitting/standing motion was confirmed. A walking device (All-in-One Walking Trainer; Healthcare Lifting Specialist, Denmark) with a harness was used for safety, and HAL training consisted of walking on a 28-meter-long circuit several times with the assistance of 2 physical therapists and a doctor. HAL training lasted 60 minutes, including rests and time for attaching/detaching the device, and was performed 2 or 3 times a week. The patient underwent 10 sessions of HAL training until being discharged on postoperative day 73. The results of the 10-m walk test are shown in Table 1. The patient’s speed and cadence markedly improved. Clinical evaluation was performed again after the final training session: the JOA score (excluding the upper extremities) was 6.5/11 (1-1, 5-1-3); the SCIM-score improved to 84 points (self-care: 20/20 points; respiration and sphincter management: 39/40 points; and mobility: 25/40 points); the AIS was still grade D; the ASIA motor score improved to 38 points (right: 19 points; left: 19 points); the ASIA sensory score for light touch improved to 86 points (right: 43 points; left: 43 points); the WISCI II score increased to 16 points; and Frankel classification was grade D (Table 2). At discharge, the patient could walk with 2 crutches and no assistance. No adverse events associated with HAL training occurred.

**Discussion**

Recently, the feasibility of HAL training for various motility disorders associated with stroke and chronic spinal cord injury has been reported. The findings in the current study suggest that HAL training may be useful in the postoperative period of a thoracic OPLL surgery.

Figure 2. A Sagittal slice, computed tomography (CT) scan of the thoracic spine. Ossification of longitudinal ligament (OPLL) is observed in T3 to T8 level. B Axial slice, CT scan of the thoracic spine showing OPLL.
Sakakima et al. reported the only other case of HAL training for a postoperative thoracic OPLL patient also with good results. Compared with that case, HAL training was introduced 2 weeks earlier in the patient reported herein. In addition, the WISCI II score before the operation and after the final training was 8 to 16 in the current case and 0 to 8 in the previous case. In brief, the case reported in Sakakima et al. showed a more severe gait disturbance. In contrast, our case experienced postoperative aggravation of paraplegia and extra bed rest for 3 weeks. Taken together, these two cases consistently show the safety and feasibility of HAL training in the early postoperative period for thoracic OPLL.

Three possibilities may underlie the improvement in walking ability for postoperative thoracic OPLL patients with HAL training. First, patients practice walking using voluntary intentions assisted by HAL, which might induce a feedback effect between the central and peripheral nervous systems. Barbeau et al. has mentioned the importance of sensory inputs in locomotor training with animal and human studies demonstrating that sensory inputs such as maximum weight facilitates proper trunk posture and are essential to maximizing functional recovery. Belda-Lois et al. have reviewed the “top-down approach” in gait rehabilitation after stroke in which rehabilitation is driven by neural plasticity. In addition, motor learning is an important concept in robotic neurorehabilitation. With HAL training, assisted motion from the patient’s voluntary drive can form the proprioceptive feedback loop; thus, repetitive voluntary training may induce motor learning. Plautz et al. revealed in their animal experiments that motor learning is a prerequisite

Table 1  The results of a 10-m walk test at initial training and final training

<table>
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<tr>
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<th>Speed (m/min)</th>
<th>Cadence (steps/min)</th>
<th>Stride (m/step)</th>
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<tr>
<td>At initial training</td>
<td>15.94</td>
<td>43.82</td>
<td>0.22</td>
</tr>
<tr>
<td>At final training</td>
<td>31.78</td>
<td>77.86</td>
<td>0.24</td>
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factor in driving representational plasticity and that repetitive motor activity alone does not produce functional recovery. This result also supports our hypothesis that assisted voluntary motion with HAL may induce feedback and motor learning, and subsequent functional recovery. Motor learning through neurofeedback has traditionally used electroencephalography. Recently, real-time functional magnetic resonance imaging (rt-fMRI) has been demonstrated to allow for high spatial resolution and imaging of activity across the entire brain within a short time, all done non-invasively. Yang-ten Fang et al. have reported that neuroplastic changes and functional recovery induced by robot-assisted therapy in post-acute stroke patients as well as fMRI findings of the brain were related to functional recovery. Therefore, we believe that rt-fMRI may give useful information regarding the mechanism of functional recovery associated with HAL training in the near future. Secondly, similar to other robotic-assisted locomotor training, HAL and the All-in-One Walking Trainer supports standing steadiness and reduces the amount of labor required of the assistant.

**Figure 4.** A AP view, plain radiograph of the thoracic spine, immediately after surgery. Posterior decompression and fusion (laminectomy for T3 to T7 and pedicle screw fixation and posterolateral fusion for T1 to T9) were performed. B Lateral view, plain radiograph of the thoracic spine, immediately after surgery.

**Table 2** The results of the cervical JOA score excluding upper extremity, ASIA classification, ASIA motor score (lower extremity), ASIA sensory score (lower extremity), SCIM-score, WISCI II score, and Frankel classification at pre-op and at discharge

<table>
<thead>
<tr>
<th>Cervical JOA score excluding upper extremity</th>
<th>ASIA classification</th>
<th>ASIA motor score (lower extremity)</th>
<th>ASIA sensory score (lower extremity)</th>
<th>SCIM-score</th>
<th>WISCI II</th>
<th>Frankel classification</th>
</tr>
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<tr>
<td>Pre-op</td>
<td>5.5/11</td>
<td>D</td>
<td>18/15</td>
<td>43/43</td>
<td>57</td>
<td>8</td>
</tr>
<tr>
<td>At discharge</td>
<td>6.5/11</td>
<td>D</td>
<td>19/19</td>
<td>43/43</td>
<td>84</td>
<td>16</td>
</tr>
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especially in patients with muscle weakness in the proximal muscles of the legs.\textsuperscript{25} Furthermore, voluntary movement may be difficult for a patient with a paraparesis due to spinal diseases such as OPLL in the acute phase. However, HAL enables one to practice walking in the acute phase of motor function disorder using the wearer’s intension. Thus, HAL enables more repeatable, precise, sustainable, and progressive training. Thirdly, training using an advanced robotic device may affect the patient’s motivation for rehabilitation. We routinely collect questionnaires including questions on expectation, tiredness, actual feeling of effect, and free comments. Further experience and analysis of such data may reveal the psychological effect of HAL training.

A limitation of the current study is that the remarkable recovery of physical function may have been achieved not only through HAL training but also through the patient’s natural postoperative course after decompression as well as through standard physical therapy. Thus, we cannot definitively show the beneficial effects of HAL in the current study. Case control and randomized control studies may clarify the effect of HAL training in the future. However, the previous case report by Sakakima et al. and the current report have shown the safety and feasibility of HAL training for patients in the early post-operative period of thoracic OPLL.

The mechanism by which HAL affects the central and nervous system is unclear; however, there have been some reports showing possible favorable change and plasticity of the central nervous system via sensory feedback.\textsuperscript{12–21,24} We believe that the best advantage of HAL over normal physical rehabilitation is the extremely synchronized enhancement of voluntary command from the brain to the muscles. HAL enables patients to voluntarily move their legs, even patients with severe muscle weakness; thus, we expect this movement may induce favorable sensory feedback to the central nervous system. Nevertheless, combined evaluation by radiological and neurophysiological assessment over time is necessary to reveal the mechanism of change, and clinical research should be performed including these assessments. Further experience with early HAL training for postoperative patients is needed.

Conclusion
HAL training for postoperative thoracic OPLL patients may enhance improvement in walking ability, even if severe impairment of ambulation and muscle weakness exist preoperatively. Early commencement of HAL training appears to be effective.

Acknowledgments
We gratefully acknowledge Mrs. Mayuko Sakamaki for her assistance during training and data collection.

Disclaimer statements
Conflicts of Interest None

Funding This work was supported by Industrial Disease Clinical Research Grants of the Ministry of Health Labour and Welfare, Japan (14060101-01).

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References