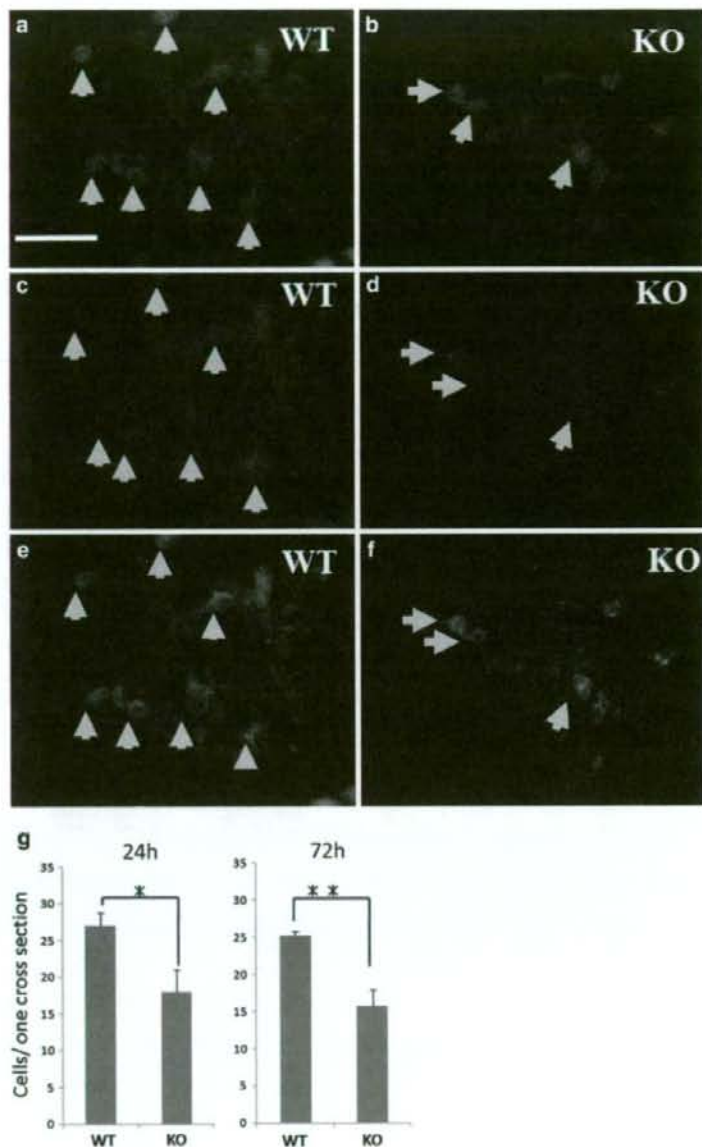


**Fig. 4** Fewer NeuN/caspase-3-active double-positive cells were counted in KO mice 24 and 72 h after spinal cord injury (SCI). We counted NeuN/caspase-3-active double-positive cells as apoptotic cells. Double-positive cells were counted and compared between genotypes. **a–f** Representative NeuN/caspase-3-active double fluorescent immunostaining 24 h after SCI. NeuN staining (**a, b**), caspase-3-active (**c, d**), and merged figures in WT and KO mice (**e, f**). Fewer neurons were caspase-3-active in KO mice (*arrow*) than WT mice (*arrow heads*). **g** Bar graphs show double-positive cell numbers in WT and KO mice 24 h (*left panel*) and 72 h (*right panel*) after spinal cord compression injury. Significantly fewer double-positive cells were counted in KO mice compared with WT mice 24 and 72 h after SCI. \* $p < 0.05$ , \*\* $p < 0.01$ . Bar 50  $\mu\text{m}$  for **a–f**



## Discussion

In this study, deletion of MIF suppressed glutamate-induced neuronal death in vitro. Recombinant human MIF exacerbated glutamate-induced neuronal death in CGN from MIF KO mice. In the in-vivo study, MIF KO mice showed a decreased number of apoptotic neurons, better neuronal survival, and better hind-limb functional recovery compared with WT mice after spinal cord compression injury.

Previously, we examined MIF expression in a mouse spinal cord compression injury model [9]. Activated microglia with amoeboid morphology that accumulated in the lesion epicenter expressed MIF mRNA and protein, and the peak of MIF expression was three days after injury. In this study, MIF deletion did not change microglia accumulation in the lesion epicenter three days after SCI. In a lipopolysaccharide-mediated endotoxemia model, MIF released from anterior pituitary glands stimulates macrophages and monocytes to secrete inflammatory cytokines

[3]. MIF is a potent inducer of TNF- $\alpha$  [3], IL-1 $\beta$  [17], and IL-6 [16]. Proinflammatory cytokines, which are secreted from macrophages and monocytes, negatively affect neuronal survival after SCI [15]. In this study, the different number of NeuN/caspase-3 positive cells may be a result of cytokine expression differences caused by the absence of MIF from accumulated inflammatory cells.

In the *in vitro* study, MIF appeared to hinder neuronal survival in WT mice, and recombinant human MIF exacerbated glutamate-induced neuronal death of CGN from KO mice. MIF regulates the proliferation and apoptosis of cells via direct effects on the tumor suppressor protein p53, implicating a role for MIF in synovial hyperplasia [16]. Delayed neuronal cell death after brain trauma is mediated by p53-dependent mechanisms [14]. The number of NeuN/caspase-3-active double-positive cells in KO mice was significantly smaller than in WT mice 24 h and 72 h after injury, suggesting MIF may also facilitate apoptosis of neurons during the first few days after injury by p53-dependent pathways *in vivo*.

Six-week hind-limb locomotor assessments revealed that KO mice showed significantly better locomotor recovery than WT mice. This result correlates with the histological finding that more neurons survived in KO mice six weeks after SCI. We could also correlate six weeks neuronal counts with neuronal cell death within a few days after injury. Less NeuN/caspase-3-positive apoptotic cells were counted in KO mice, suggesting that the initial response may determine long-term outcome.

In this study, significant locomotor recovery in KO mice was observed three to six weeks after SCI. The time course recovery of hind-limb function showed statistically significant differences between genotypes in repeated-measures ANOVA ( $p < 0.01$ ). The difference in the recovery curve between genotypes was partially explained by the difference in one to three days neuronal death or six weeks neuron counts. In addition, dorsal skin wound healing was significantly delayed in MIF KO mice compared with WT mice because of a significant reduction in fibroblast and keratinocyte migration, as observed in MIF KO mice after 1-oleoyl-2-lysophosphatidic acid treatment [19]. In the injured spinal cord, a reduction in KO mouse fibroblasts could occur because the excitotoxic properties of glutamate and the ischemic conditions cause acidosis. The difference in hind-limb locomotor score between genotypes may be derived from the wound-healing process of fibroblasts that have migrated to the lesion epicenter after SCI.

The MIF protein is present in astrocyte-like cells in the cerebral white matter and cortex [12]. The signal transductions in KO mouse astrocytes could be somewhat different than in the WT mice because MIF is upregulated in astrocytes seven days after SCI [9]. This difference may cause a glial scar that has a different molecular mechanism of MIF

signal transduction in the injured spinal cord. Elucidation of the significantly improved recovery process in KO mice after SCI may facilitate development of new therapeutic approaches in SCI.

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## Urinary Retention without Tetraparesis as a Sequel to Spontaneous Spinal Epidural Hematoma

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Tomonori Yamanishi<sup>6</sup> and Takamichi Hattori<sup>2</sup>

### Abstract

A 55-year-old man suddenly developed neck pain, tetraplegia and decreased sensation below the neck. He was diagnosed with SSEH. Surgical removal of the hematoma, and laminoplasty were performed. At 2 months after the onset of the disease, the patient regained the ability to walk. However, at 5 months after the onset of the disease, the patient remained in a state of urinary retention even though his neurological findings were normal, except for mildly brisk reflexes in the lower extremities and decreased superficial sensation below the level of T4 including the perineal area. A urodynamic study showed normal bladder sensation, despite an acontractile detrusor and an unrelaxing external sphincter upon voiding. It is postulated that the descending micturition pathways (just inside the pyramidal tracts) were selectively affected, while the ascending micturition pathways (the dorsal columns) were preserved in the present case.

**Key words:** urinary retention, spinal epidural hematoma, urodynamic study

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### Introduction

Spontaneous spinal epidural hematoma (SSEH) is a rare condition that causes severe paresis and pain (1). The most appropriate treatment for SSEH is surgery, with rapid decompression of the spinal cord being the most effective (1). However, following decompression surgery, some clinical symptoms may persist. We recently examined a patient who suffered from urinary retention without tetraparesis after SSEH.

### Case Report

A healthy 55-year-old man suddenly developed severe neck pain, followed by tetraparesis. He had no history of previous illnesses such as hypertension or coagulopathy, nor did he have straining effort just prior to the onset of neck

pain. Upon admission to the Orthopedic Surgery Department in our hospital, the patient had tetraplegia and decreased sensation below the neck, and an indwelling urinary catheter was inserted. Magnetic resonance imaging (MRI) of the cervical spine showed a C2-7 SEH anteriorly but without vascular malformation on T2-weighted images (Fig. 1). Surgical removal of the hematoma together with laminoplasty gradually ameliorated the patient's tetraplegia. At 2 months after the onset of the disease, the patient regained the ability to walk, however, after the urethral catheter was removed, he was unable to urinate at all and was taught to perform clean intermittent self-catheterization (CISC) 4 times a day. At 5 months after the onset of disease, the patient was still in a state of urinary retention and was referred to our urodynamic laboratory. Upon referral, the patient had normal muscle power in the four extremities, while his deep tendon reflexes in the lower extremities were slightly brisk bilaterally, without extensor plantar reflexes. Superficial sensation

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Figure 1. MRI of the cervical cord at the time of disease onset. T2-weighted MRI images showed a C2-7 spinal epidural hematoma anteriorly (arrowheads).

determined by pin prick was moderately decreased bilaterally below the level of T4. Position and crude touch sensation in the extremities, including the perineal area, were preserved. He had constipation that needed daily laxative. He had no perspiratory dysfunction or postural syncope. A urodynamic study was performed to measure post-void residual volume and medium-fill (50 ml/min) water cystometry. Sphincter electromyography (EMG) was carried out simultaneously using a concentric needle electrode in the external anal sphincter muscles. Normal volumes were observed at the first sensation of the bladder filling, i.e., a volume of 120 ml (100 < normal < 300), and at bladder capacity, i.e., a volume of 600 ml (200 < normal < 600) during the storage phase. There was no evidence of detrusor overactivity or low-compliance detrusor. During the voiding phase, the patient showed an acontractile detrusor without urinary flow, and the sphincter EMG remained unchanged while he tried to start voiding; this was followed by considerable straining during the voiding phase. The patient had a post-void residual volume of 600 ml (normal < 30). He was started on 20 mg/day pyridostigmine, a cholinesterase inhibitor, and 60 mg/day urapidil, an alpha-adrenergic antagonist. Six months after the urodynamic assessment, the patient's difficult in-

urination had ameliorated only slightly.

Pure urinary retention rarely appears in compressive disorders; however, a remarkable feature of the present patient was that there was dissociation between his recovery from tetraplegia and urinary retention after an acute transverse lesion; that is, while his tetraplegia disappeared completely, his urinary retention remained to the extent that needed CISC over a period of 9 months. To the best of our knowledge, there have been no previous reports on urodynamic studies in SSEH. However, the results of the present study (acontractile detrusor and unrelaxing sphincter on voiding) are consistent with those found in patients in the acute spinal shock phase, which could last for more than 6 months after the initial insult on the spinal cord (2). In the case of a patient with anterior spinal artery (ASA) infarction who initially presented with urinary retention, a postmortem pathological examination suggested that the patient's urinary retention might have been due to the central cord syndrome, e.g., an initial ischemic event in the watershed area of the ASA, which involved the lateral columns that include the pyramidal tract and the adjacent long tract pathways (3). Similarly, the central cord syndrome due to cord compression with/without ischemia occurs in extramedullary disorders including SSEH (4). In addition, in a myelitis case with pure urinary retention (5), an MRI scan revealed localized inflammatory lesions that involved the lateral medullary reticulospinal tract just inside the pyramidal tract, e.g., the descending pathway to the bladder (6). Although no intramedullary lesions were visualized on MRI, it is possible that in the present patient the same area might have been selectively involved by the compressive/ischemic event by SSEH, leading to the presentation of urinary retention as an outstanding symptom. These reported cases, including ours, may indicate the vulnerability of the micturition descending fibers within the spinal cord. Nevertheless, a urodynamic study showed that bladder sensation was preserved in our patient. Since the major ascending pathways from the bladder are thought to travel in the dorsal columns (7), these pathways seemed to be preserved in the present case.

We have reported here the case of a man with urinary retention without tetraparesis as a sequel to spontaneous spinal epidural hematoma. The present case may indicate vulnerability of the micturition descending fibers within the spinal cord.

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## Paraparesis Caused by Rib Exostosis in a Child With Down Syndrome

### A Case Report

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Takashi Saisu, MD, PhD,† Shohei Minami, MD, PhD,‡ and Kazuhisa Takahashi, MD, PhD\*

#### Study Design. Case report.

**Objective.** We describe a case of paraparesis caused by a rib exostosis compressing the spinal cord.

**Summary of Background Data.** Exostoses are the most common benign tumors of bone and often arise from the metaphyses of long bones. Spinal exostoses are uncommon lesions, but when present, may compress the spinal cord and cause significant neurologic symptoms. Spinal exostoses leading to neurologic impairment typically arise from vertebrae or laminae. It is very rare for exostoses originating from rib heads to cause neurologic symptoms.

**Methods.** The patient's medical and radiographic history is reviewed as well as the relevant medical literature.

**Results.** A 10-year-old boy with multiple exostoses and Down syndrome was referred to our hospital because of difficulty walking. Careful physical examination revealed that his gait disturbance was because of paraparesis. Radiographs, computed tomography scan, and magnetic resonance imaging revealed that an exostosis arising from the left eighth rib head was compressing the spinal cord. The exostosis was resected and the patient's neurologic symptoms resolved.

**Conclusion.** Exostosis rising from the rib head can compress the spinal cord and cause neurologic deficits. Surgical resection of the rib exostosis should be strongly considered when neurologic symptoms develop.

**Key words:** multiple exostoses, rib, spinal cord compression, paraparesis, Down syndrome. *Spine* 2008;33:E911-E913

Exostoses, also known as osteochondromas, are the most common benign tumors of bone representing 36% of all benign bone tumors and 8.5% of all bone tumors.<sup>1,2</sup> Exostoses often arise from the metaphyses of long bones; spinal exostoses are uncommon.<sup>1,2</sup> Spinal exostoses that compress the spinal cord can lead to significant neurologic symptoms. Neurologically, symptomatic exostoses often arise from the vertebrae or lam-

inae and it is very rare for an exostosis originating from a rib head to cause neurologic impairment. Herein, we report a case of exostosis arising from a rib head, which compressed the spinal cord and led to paraparesis in a child with Down syndrome and multiple exostoses.

#### Case Reports

A 10-year-old boy with multiple exostoses and Down syndrome was referred to our hospital for a history of difficulty walking. He had previously been seen at several outlying hospitals for his symptoms but his gait disturbance was thought to be secondary to knee pain caused by lower extremity exostoses.

Muscle strength in the lower extremities was graded 2 to 3 and the patient could not stand. Hyperactive deep tendon reflexes were noted at the patella and Achilles tendons. Analgesia was noted below the T6 dermatomal level. Slight bowel/bladder dysfunction was observed.

Bone scintigraphy revealed multifocal uptake in his extremities. Radiographs demonstrated multiple exostoses in his extremities and pelvis. An abnormal silhouette of the left T7 pedicle and a bony mass around the head of the left eighth rib were observed in his spinal radiogram. Sagittal magnetic resonance imaging (MRI) of the thoracic spine showed a characteristic coin lesion in the spinal canal at the T8 vertebral level (Figure 1A). Axial and coronal MRI examination found that the lesion in the canal was compressing the spinal cord from the left side (Figure 1B). Myelography demonstrated a disruption of contrast filling at the T7/8 disc level and postmyelography computed tomography (CT) scan of the area showed a bony mass arising from the left eighth rib head that expanded into the spinal canal (Figure 2A-C).

The patient underwent a T7 and T8 laminectomy and resection of the bony mass arising from the eighth rib head. The bony mass had a cartilaginous cap and entered the canal, compressing the spinal cord at the T7 vertebral level. The portion of the mass in the canal was completely resected, whereas the portion of the exostosis outside of the canal was resected as much as possible.

Pathologically, the tumor specimen was found to comprise cancellous bone with cartilaginous elements. The final pathologic diagnosis was exostosis. Postoperative CT scan revealed that the exostosis had been cleared from the canal, and MRI showed excellent decompression of the spinal cord (Figure 3A-C).

Functional motor recovery in the lower extremities was observed immediately after the surgery and the pa-

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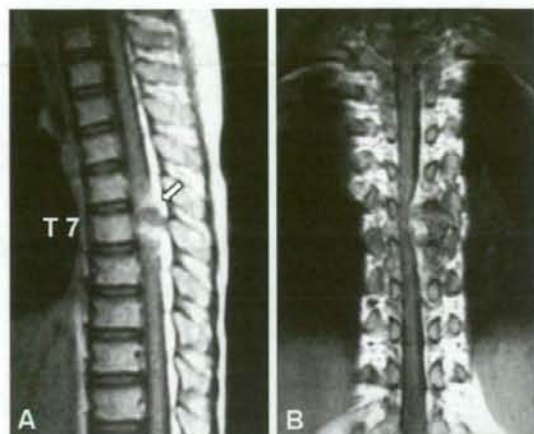


Figure 1. Sagittal T1-weighted MRI (A) demonstrated a low signal intensity mass (arrow) in the canal at the T7 vertebral level. Coronal T1-weighted MRI (B) reveals that the spinal cord is being compressed by the mass from the left side.

tient could walk unassisted 12 weeks after the operation. Twelve years after the surgery, motor function in the patient's lower extremities remains normal and he can walk without difficulty. There are no residual sensory or bowel/bladder deficits. Radiography of the thoracic spine 12 years after operative intervention was remarkable only for slight scoliosis and kyphosis.

#### Discussion

Exostoses that compress the spinal cord and cause neurologic symptoms typically arise from the vertebral pedicles or laminae.<sup>3</sup> In contrast, in the present case, CT scan confirmed that the exostosis compressing the spinal cord was instead arising from the rib head. Exostoses of the rib are often asymptomatic. When symptoms occur, they may be variable depending on the size and nature of the exostosis; previously reported symptoms include hemothorax,<sup>4</sup> pneumothorax,<sup>5</sup> pneumonia,<sup>6</sup> hiccup,<sup>7</sup> snapping scapula,<sup>8</sup> and thoracic outlet syndrome.<sup>9</sup> It is very rare that exostoses arising from the rib lead to compression of the spinal cord and neurologic symptoms. Only 6 cases have been reported previously.<sup>10-14</sup> All of the patients reported, including our patient, underwent surgical resection, which led to a positive outcome. Surgical resection should thus be strongly considered if the exostosis compresses the spinal cord and leads to neurologic deficits.

Hereditary multiple exostoses are occasional features of mental retardation syndromes, such as Langer-Giedion syndrome<sup>15</sup> and Potocki-Shaffer syndrome,<sup>16</sup> deletion syndromes caused by loss of material on chromosomes 8q24 and 11p11, respectively. To our knowledge, there have been no previous reports of multiple exostoses in constellation with Down syndrome. The association thus seems serendipitous in the present patient.

Finally, historical information regarding the nature, onset, and duration of symptoms may prove challenging

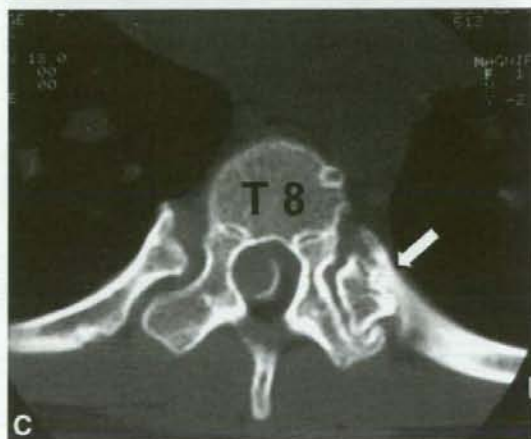
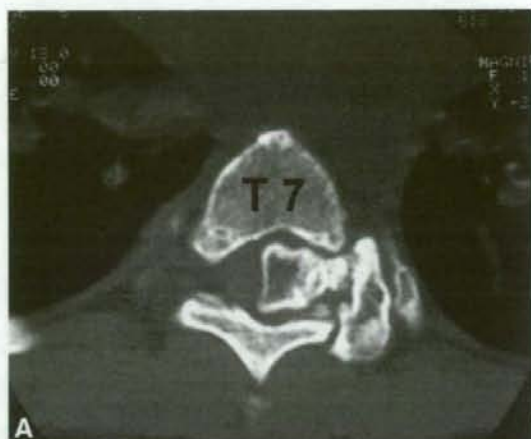


Figure 2. CT scan after myelography at the T7 level (A) T7/8 disc level (B) and T8 level (C) demonstrates an exostosis arising from the left eighth rib head (arrow) that expands into the canal.

to obtain in the patient with mental retardation. Examination of the patient may be complicated by their limited ability to comply with instructions. However, care-



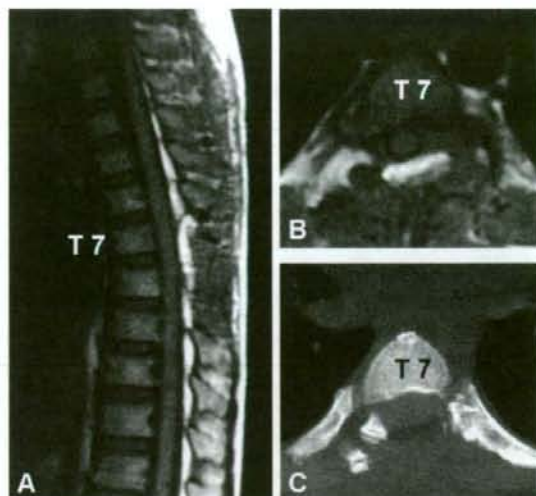


Figure 3. Postsurgical T1-weighted sagittal (A) and axial (B) MRI at the T7 level demonstrates that the spinal cord has been decompressed. CT scan at the T7 level (C) revealed that the exostosis in the canal has been removed.

ful observation in concert with methodical physical manipulation will typically allow the examiner to recognize the cause of the patient's deficit, as was possible in this case.

#### ■ Key Points

- A 10-year-old boy with multiple exostoses and Down syndrome was referred to our hospital because of difficulty walking.
- Careful physical examination revealed that his gait disturbance was because of paraparesis. Radiographs, computed tomography scan, and magnetic resonance imaging revealed that an exostosis arising from the left eighth rib head was compressing the spinal cord.

- The exostosis was operatively resected and his neurologic symptoms resolved.

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## Surgical Results and Related Factors for Ossification of Posterior Longitudinal Ligament of the Thoracic Spine

### A Multi-Institutional Retrospective Study

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#### Study Design. Retrospective multi-institutional study

**Objective.** To describe the surgical outcomes in patients with ossification of the posterior longitudinal ligament in the thoracic spine (T-OPLL) and to clarify factors related to the surgical outcomes.

**Summary of Background Data.** Detailed analyses of surgical outcomes of T-OPLL have been difficult because of the rarity of this disease.

**Methods.** The subjects were 154 patients with T-OPLL who were surgically treated at 34 institutions between 1998 and 2002. The surgical procedures were laminectomy in 36, laminoplasty in 51, anterior decompression via anterior approach in 25 and via posterior approach in 29, combined anterior and posterior fusion in 8, and sternum splitting approach in 5 patients. Instrumentation was conducted in 52 patients. Assessments were made on (1) The Japanese Orthopedic Association (JOA) scores (full

score, 11 points), its recovery rates, (2) factors related to surgical results, and (3) complications and their consequences.

**Results.** (1) The mean JOA score before surgery was  $4.6 \pm 2.0$  and  $7.1 \pm 2.5$  after surgery. The mean recovery rate was  $36.8\% \pm 47.4\%$ . (2) The recovery rate was 50% or higher in 72 patients (46.8%). Factors significantly related to this were location of the maximum ossification (T1–T4) (odds ratio, 2.43–4.17) and the use of instrumentation (odds ratio, 3.37). (3) The frequent complications were deterioration of myelopathy immediately after surgery in 18 (11.7%) and dural injury in 34 (22.1%) patients.

**Conclusion.** The factors significantly associated with favorable surgical results were maximum ossification located at the upper thoracic spine and use of instrumentation. T-OPLL at the nonkyphotic upper thoracic spine can be treated by laminoplasty that is relatively a safe surgical procedure for neural elements. The use of instrumentation allows correction of kyphosis or prevention of progression of kyphosis, thereby, enhancing and maintaining decompression effect, and its use should be considered with posterior decompression.

**Key words:** thoracic spine, ossification of posterior longitudinal ligament, surgical outcome, spinal instrumentation. **Spine 2008;33:1034–1041**

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Thoracic myelopathy caused by ossification of the posterior longitudinal ligament of the thoracic spine (T-OPLL) is usually progressive and responds poorly to conservative therapy, making surgery the only effective treatment option. Despite advancement in surgical techniques and tools employed for surgery of T-OPLL, favorable surgical results are not always achieved. Fujimura *et al*<sup>1</sup> investigated the surgical outcomes at a mean follow-up of 35 months after anterior decompression and fusion in 48 T-OPLL patients. They reported favorable overall results, but that the results were poorer in patients with a longer morbidity period, massive ossification, and ossification of other ligaments in association with T-OPLL. Matsuyama *et al*<sup>2</sup> investigated the surgical results in 21 patients with T-OPLL, and reported post-

operative deterioration of thoracic myelopathy in 5 of these 21 patients.

According to a radiologic study conducted by Ohtsuka *et al*,<sup>3</sup> the prevalence of T-OPLL was 0.8% in 1058 subjects from the general population in a rural town in Japan, which was significantly lower than that of 3.2% for OPLL of the cervical vertebrae. Thus, since the T-OPLL is a rather uncommon condition<sup>3-5</sup> and the number of patients visiting a single institute is limited, detailed analysis of operated cases has been difficult. A Research Group for Ossification of the Spinal Ligament sponsored by the Japanese Ministry of Health, Labor and Welfare and constituted by members from major Japanese institutions engaged in the treatment of spinal diseases, conducted a multi-institutional retrospective survey of patients who underwent surgery for T-OPLL. This report describes the results of the analyses conducted by this group with regards to the surgical outcomes, factors related to the surgical outcomes, and perioperative complications in patients with T-OPLL.

#### Materials and Methods

The survey pertained to T-OPLL patients who underwent surgery during the 5-year period from 1998 to 2002 at any one of the 34 institutions where the members of the research group belonged. In July 2004, questionnaires were sent to each institution by the secretary office of the present survey. Each institution was requested to fill in the questionnaire. The data were recovered by the end of December 2004, and a total of 198 operated cases were collected. The analysis was conducted on the data obtained from 154 of 198 patients who had postoperative follow-up period of at least 1 year and whose important data including their sex, age, preoperative neurologic status, surgical methods and results, and major complications were not missing.

The study group consisted of 62 males and 92 females, with a mean age of 56.8 years (range, 27-79 years). The mean follow-up period was 3.0 years (range, 1-6 years).

The items investigated were the patients' demographic data, the underlying disease, presence/absence of comorbidity, details of the history of spinal surgery, radiologic findings [x-ray, magnetic resonance imaging (MRI), and computed tomography], surgical methods and surgical results, complications, and the surgical outcomes.

The morphology of the T-OPLL, level of the ossified lesions, and the kyphosis angle of the thoracic vertebrae (T3, 4 to T12) were determined radiologically. The morphology of T-OPLL was classified as the linear type, beaked type, continuous waveform type, continuous cylindrical type, or the mixed type (composed of at least 2 of these types), according to the classification established by the research group in 1993 (Figures 1-3).<sup>6</sup> The thoracic vertebral levels of the maximum ossification and maximum cord compression were determined by computed tomography and MRI, and the presence/absence of an intramedullary high-intensity lesion was assessed on T2-weighted MR images.

The surgical outcomes were assessed by the Japanese Orthopedic Association (JOA) score for thoracic myelopathy (total of 11 points), which was derived from the JOA scoring system for cervical myelopathy by eliminating the motor and sensory scores for the upper extremity (Table 1). The recovery rate was calculated using the preoperative JOA score (points) and the

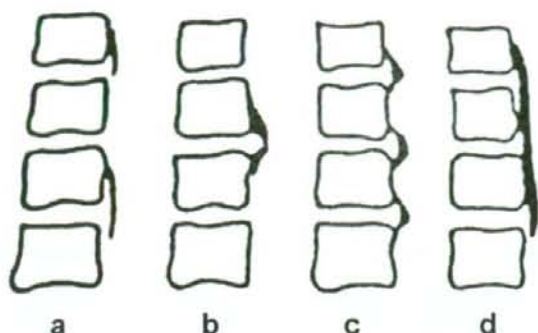


Figure 1. Classification of OPLL.<sup>6</sup> (a), linear type; (b) beaked type; (c), continuous waveform type; (d), continuous cylindrical type. Mixed type is defined as a combination of 2 or more different types.

JOA score at the follow-up, according to the following formula; Recovery rate = (JOA score at follow-up - preoperative JOA score)/(11 - preoperative JOA score) × 100 (%). The Frankel classification modified by Bradford *et al*<sup>7</sup> was also used for evaluation of the surgical outcomes.

#### Statistical Analysis

Stata 9 software (Stata Corp., College Station, TX) was used for the statistical analysis. The surgical outcomes and the factors related to the outcomes were assessed by logistic regression analysis. Age- and sex-adjusted odds ratios and their 95% confidence intervals were demonstrated.



Figure 2. Beaked type of OPLL compressing the spinal cord. Sagittal reconstruction of CAT scan of a 64-year-old man demonstrating a beaked type of OPLL.



Figure 3. Continuous cylindrical type of OPLL. Tomogram of a 48-year-old woman showing a continuous cylindrical type of OPLL.

## Results

### Clinical Data

The initial symptom was numbness of the lower extremities in 87 (56.5%), gait disturbance in 72 (46.8%),

Table 1. JOA Scoring System for Thoracic Myelopathy

Category	Score (Point)
Motor function	
Lower extremity	
Unable to stand and walk by any means	0
Unable to walk without a cane or other support on a level	1
Walks independently on a level but needs support on stairs	2
Capable of fast but clumsy walking	3
Normal	4
Sensory function	
Lower extremity	
Apparent sensory disturbance	0
Minimal sensory disturbance	1
Normal	2
Trunk	
Apparent sensory disturbance	0
Minimal sensory disturbance	1
Normal	2
Bladder function	
Urinary retention and/or incontinence	0
Sense of retention and/or dribbling and/or thin stream and/or incomplete continence	1
Urinary retardation and/or pollakiuria	2
Normal	3

Table 2. Demographics and Clinical Data of Patients

Sex	
Male	62
Female	92
Mean age	56.8 yr (range, 27–79)
Mean follow-up	3.0 yr (range, 1–6)
Mean morbidity period	24.3 mo (range, 1–183)
Initial symptoms	
Numbness in the lower extremities	87 (56.7)
Gait disturbance	72 (46.8)
Weakness of lower extremities	47 (30.5)
Sensation of trunk strangulation	23 (14.9)
Urinary disturbance	5 (3.2)

weakness of the lower extremities in 47 (30.5%), strangulating sensation of the trunk in 23 (14.9%), and urinary disturbance in 5 (3.2%) patients (some patients presented with multiple symptoms). The mean morbidity period from the onset of the initial symptom to surgery was 24.3 months (range, 1–183 months). The prevalence of underlying diabetes mellitus was 22.1% (34 of 154). Twelve (7.8%) of the 154 patients had a history of depression of the thoracic spine, and 29 (18.8%) of the 154 patients had a history of surgery in the spine other than the thoracic vertebrae; of these 29 patients, 23 had surgery for OPLL of the cervical spine, and the remaining 6 had surgery for lumbar spinal diseases (Table 2).

### Radiologic Findings

The morphology of T-OPLL was classified as the linear type in 8 patients (5.2%), the beaked type in 45 patients (29.2%), the continuous waveform type in 46 patients (29.9%), the continuous cylindrical type in 26 patients (16.9%), and the mixed type in 29 patients (18.8%). The level of maximum ossification was located between the first to fourth thoracic vertebrae (T1–T4) in 69 patients, T5–T8 in 62 patients, and T9–T12 in 23 patients. The mean anteroposterior diameter of the lesion at the level of the maximum ossification was  $6.7 \pm 2.0$  mm (range, 1.5–12.0 mm). On T2-weighted MRI, an intramedullary high-intensity lesion was recognized in 83 (63.8%) of the 130 patients in whom the lesion could be evaluated. With regard to association of T-OPLL with ossification of other spinal ligaments, cervical OPLL was recognized in 97 patients (63%), and ossification of the yellow ligament in the thoracic spine was recognized in 96 patients (62.7%).

**Surgical Methods.** Laminectomy was conducted in 36 patients, laminoplasty in 51 patients, anterior decompression and fusion *via* an anterior extrapleural or transpleural approach in 25 patients, anterior decompression *via* a posterior approach (the method reported by Ohtsuka *et al*<sup>8</sup>) in 29 patients, circumferential decompression and fusion *via* a combined anterior and posterior approach reported by Tomita *et al*<sup>9</sup> in 8 patients, and anterior decompression and fusion *via* a sternal splitting approach in 5 patients (Table 3).<sup>10</sup> When the surgical procedures selected for different morphologic types of T-OPLL were assessed, the linear type was most frequently treated by laminectomy, the beaked type by

**Table 3. Type of OPLL and Surgical Methods**

	No	Type of OPLL				
		Linear 8 (5.2)	Beaked 45 (29.2)	Continuous Waveform 46 (29.9)	Continuous Cylindrical 26 (16.9)	Mixed 29 (18.8)
Laminectomy	36	5 (62.5)	8 (17.8)	9 (19.6)	7 (26.9)	7 (24.1)
Laminoplasty	51	0	16 (35.6)	19 (41.3)	11 (42.3)	5 (17.2)
Anterior decompression <i>via</i> anterior approach	25	0	12 (26.7)	4 (8.7)	2 (7.7)	7 (24.1)
Anterior decompression <i>via</i> posterior approach	29	3 (37.5)	5 (11.1)	12 (26.1)	6 (23.1)	3 (10.3)
Circumferential decompression	8	0	2 (4.4)	2 (4.3)	0	4 (13.8)
Sternum splitting approach	5	0	2 (4.4)	0	0	3 (10.3)

Values inside parentheses indicate percentages.

laminoplasty, and anterior decompression and fusion *via* anterior approach, the continuous waveform type and the continuous cylindrical type by laminoplasty, and the mixed type by laminectomy, and anterior decompression and fusion *via* anterior approach.

When the procedures selected for different levels of maximum ossification were assessed, laminoplasty was conducted in 50% of all patients with maximum ossification at the level of T1–T4, laminectomy, laminoplasty, and anterior decompression and fusion *via* anterior approach were conducted at almost the same frequency in patients with maximum ossification at the level of T5–T8, whereas laminectomy was conducted in 52% of all the patients with maximum ossification at the level of T9–T12 (Table 4).

Augmentation by spinal instrumentation was conducted in 52 patients (33%), posterior instrumentation in 50 patients, and anterior instrumentation in the remaining 2 patients. Instrumentation was combined with laminectomy in 52.8%, with laminoplasty in 21.6%, with anterior decompression and fusion *via* anterior approach in 20.0%, with anterior decompression *via* posterior approach in 34.5%, with circumferential decompression and fusion in 87.5%, and with sternal splitting approach in none of the patients. The ossified lesion was excised in 48 patients (31.2%), thinned and floated in 25 patients (16.2%), and left untouched in 81 patients (52.6%). Intraoperative electrophysiologic monitoring

**Table 4. Level of OPLL and Surgical Methods**

	No	Level of OPLL		
		T1–T4 69(44.8)	T5–T8 62 (40.3)	T9–T12 23(14.9)
Laminectomy	36	6 (8.7)	18 (29.0)	12 (52.2)
Laminoplasty	51	35 (50.7)	15 (24.2)	1 (4.3)
Anterior decompression <i>via</i> anterior approach	25	4 (5.8)	16 (25.8)	5 (21.7)
Anterior decompression <i>via</i> posterior approach	29	16 (23.2)	8 (12.9)	5 (21.7)
Circumferential decompression	8	3 (4.3)	5 (8.1)	0
Sternum splitting approach	5	5 (7.2)	0	0

Values inside parentheses indicate percentages.

was conducted in 77 patients (50.0%), and intraoperative ultrasonography was conducted in 51 patients (33.1%). Seven patients (4.5%) underwent additional decompression during the follow-up period.

**Surgical Outcomes.** The mean JOA score was  $4.6 \pm 2.0$  before surgery,  $6.9 \pm 2.4$  at 1 year after surgery,  $7.0 \pm 2.4$  points at 3 years after surgery, and  $7.1 \pm 2.5$  points at the final follow up, with a mean recovery rate of  $36.8\% \pm 47.4\%$  at the final follow-up. A mean recovery rate at the follow-up was  $36.9\% \pm 23.3\%$  in patients treated by laminectomy,  $39.9\% \pm 39.6\%$  by laminoplasty,  $26.6\% \pm 46.8\%$  by anterior decompression *via* anterior approach,  $29.7\% \pm 53.3\%$  by anterior decompression *via* posterior approach,  $64.1\% \pm 28.2\%$  by circumferential decompression,  $48.1\% \pm 27.2\%$  by sternum splitting approach.

The pre- and postoperative modified Frankel classification was tabulated in Table 5. The paralysis improved by at least one grade in 107 patients (69.5%), remained unchanged in 38 patients (24.7%), and deteriorated by at least one grade in 9 patients (5.8%).

**Factors Related to the Surgical Outcomes.** The recovery rate was 50% or higher in 72 patients (46.8%), and factors related to the recovery rate of 50% or higher were assessed, including age, sex, preoperative morbidity period, preoperative JOA score, morphologic type of the ossified lesion, anteroposterior diameter of the ossified lesion, kyphosis angle of the thoracic vertebra, intramedullary high-intensity lesion on T2 weighted MR images, level of maximum ossification, surgical method, combined use of instrumentation, and reoperation, and presence/absence of diabetes mellitus. The cut-off value of 50% was used, because the recovery rate of 50% or

**Table 5. Surgical Outcomes**

	JOA Scores	Modified Frankel Classification						
		A	B	C	D1	D2	D3	E
Preop.	$4.6 \pm 2.0$	2	8	33	40	44	24	3
Follow-up	$7.1 \pm 2.4$	1	4	7	13	38	67	24

The recovery rate of JOA scores was  $36.8\% \pm 47.4\%$ . Improvement in Frankel grade was obtained in 107 patients (69.5%).

**Table 6. Factors Related to Surgical Outcomes (50% or Higher Recovery Rate of JOA Scores)**

Factors	% of Patients*	Odds Ratio (95% Confidence Interval)	P
Sex			
Female	48.8	1.00	
Male	47.5	0.95 (0.49–1.83)	0.877
Age			
<49	66.7	1.00	
50–59	46.7	0.44 (0.18–1.06)	0.067
60–69	41.0	0.35 (0.13–0.91)	0.032
≥70	29.4	0.21 (0.06–0.74)	0.015
Morbidity period			
<1 yr	58.5	1.00	
1–3	50.0	0.71 (0.31–1.59)	0.400
>3	34.0	0.46 (0.19–1.07)	0.073
Preop. JOA scores			
<5	51.4	1.00	
≥5	44.9	0.75 (0.38–1.46)	0.396
Type of OPLL			
Beaked/continuous wave form	51.2	1.00	
Mixed	65.4	1.87 (0.72–4.84)	0.196
Linear	16.7	0.20 (0.02–1.86)	0.157
Continuous cylindrical	34.6	0.61 (0.23–1.59)	0.311
Anteroposterior diameter of OPLL			
<5 mm	57.9	1.00	
5–10	46.9	0.73 (0.26–2.04)	0.551
≥10	70.0	1.81 (0.33–9.88)	0.492
Kyphosis angle on MRI			
<30 degrees	48.4	1.00	
≥30	51.5	1.10 (0.54–2.24)	0.794
Level of OPLL			
T1–4	59.7	1.00	
T5–8	40.0	0.41 (0.19–0.87)	0.020
T9–12	31.8	0.24 (0.08–0.71)	0.010
Surgical methods			
Anterior decompression via anterior approach/Sternum splitting approach	46.7	1.00	
Anterior decompression via posterior approach	53.6	1.20 (0.40–3.43)	0.770
Circumferential decompression	87.5	8.20 (0.84–79.98)	0.071
Laminectomy/laminoplasty	43.5	0.90 (0.35–2.07)	0.727
Use of instrumentation			
No	38.7	1.00	
Yes	63.0	3.40 (1.57–7.2)	0.002
No. of surgeries			
Single	61.0	1.00	
Two or more	46.2	0.48 (0.14–1.69)	0.251
Diabetes mellitus			
No	47.4	1.00	
Yes	50.0	1.17 (0.53–2.60)	0.702
Intradiscal high-intensity lesion			
No	44.2	1.00	
Yes	51.8	1.40 (0.65–3.02)	0.391

Sex- and age-adjusted odds ratio and the 95% confidence interval is shown except that for sex and age.

\*The numbers showing percentage of patients who obtained recovery rate of 50% or higher at the follow-up in each group.

†Statistically significant difference.

higher has been considered to be good to excellent surgical outcomes in the previous literature.<sup>11</sup> As a result, maximum ossification at T1–T4 [odds ratio; 1 for T1–T4 *vs.* 0.41 (95% confidence interval; 0.19–0.87,  $P = 0.02$ ) for T5–T8 and 0.24 (0.08–0.71,  $P = 0.01$ ) for T9–T12] and combined use of instrumentation with surgery [odds ratio; 1 for without instrumentation *vs.* 3.40 (1.57–7.2,  $P = 0.002$ ) for with instrumentation] were associated significantly with the better outcomes (Table 6). None of the other factors was significantly related to the surgical outcome; however, the outcome tended to be favorable in patients treated by circumferential decompression and fusion and in patients with a morbidity period of less than 1 year, and in patients younger than 50 years of age.

**Complications.** The following perioperative complications were recognized: deterioration of thoracic myelopathy immediately after the surgery in 18 patients (11.7%); epidural hematoma in 3 patients (1.9%); dural injury resulting in cerebrospinal fluid leakage in 34 patients (22.1%); respiratory complications in 8 patients (5.2%); hoarseness in 2 patients; ileus, esophageal fistula, meningitis, myocardial infarction, and enteritis in 1 patient each.

In patients with neurologic deterioration, the grades of paralysis were Frankel A in 3 patients, B in 7, C in 4, D in 4, and surgical procedures employed were laminectomy in 3 patients (8.3%), laminoplasty in 2 (3.9%), anterior decompression *via* anterior approach in 5

(20.0%), anterior decompression and fusion via posterior approach in 6 (20.7%), and circumferential decompression in 2 (25.0%). In this group of patients, the ossified lesion was excised in 7 patients (*i.e.*, deterioration occurred in 14.2% of 48 patients who underwent excision), thinned and floated in 7 patients (*i.e.*, 28% of 25 patients who underwent thinning and floating), and left untouched in 4 patients (*i.e.*, 4.9% of 81 patients whose ossified lesion was left untouched). Thus, neurologic deterioration was observed more frequently in surgical procedures in which the ossified lesion was excised or thinned and floated than those in which the lesion was left untouched.

Measures against immediate neurologic deterioration were administration of steroid in 14 patients, surgical evacuation of hematoma in 2. In 12 patients (66.7%), the paralysis started to recovery in 5.6 days, on average (1–30 days), after the index surgery. The paralysis improved in 1 of 3 patients with Frankel A, 6 of 7 patients with Frankel B, 2 of 4 patients with Frankel C, and all 4 patients with Frankel D paralysis. Cerebrospinal fluid leakage was managed conservatively in 26 patients, and surgically in 8 patients.

#### ■ Discussion

In the present study, improvement by at least one grade in the modified Frankel classification was obtained in 69.5% of the T-OPLL patients after surgical treatments, and mean recovery rate of the JOA score was 37%. Thus, moderate improvement of myelopathy was obtained after surgery in T-OPLL patients. However, compared with recovery rates of cervical OPLL reported in the literature, which ranged from 43% to 63%,<sup>11–13</sup> the recovery rates of T-OPLL were much lower. This is possibly attributable to the following reasons: (1) posterior decompression alone for T-OPLL is minimally effective because of kyphosis of the thoracic spine except in the upper thoracic spine where some lordosis exists; (2) the blood flow to the thoracic spinal cord is less than that to the cervical spinal cord; (3) anterior approach to the thoracic spine is more difficult than that to the cervical spine, making decompression surgery more technically demanding.

Fujimura *et al*<sup>14</sup> have identified several factors related to poor surgical results, including a long morbidity period, extensive OPLL, and ossification of other spinal ligaments. Matsuyama *et al*<sup>2</sup> have reported that many patients with the beaked-type of OPLL showed exacerbation of the neurologic symptoms after surgery. Tokuhashi *et al*<sup>15</sup> have reported that the efficacy of posterior decompression for OPLL may be poorer in patients with a large kyphosis angle on preoperative MRI. In the present survey, the factors that were found to be significantly related to the surgical outcomes were the level of maximum ossification at the upper thoracic spine and the combined use of instrumentation with decompression surgery.

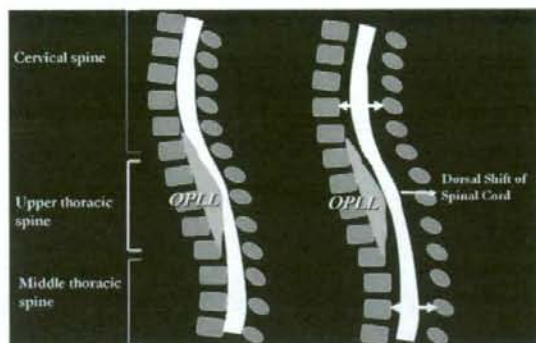


Figure 4. Schematic drawing describing the mechanism of decompression of the spinal cord by laminoplasty in patients with OPLL at the upper thoracic spine. Since the spinal curvature is usually lordotic or only slightly kyphotic at the cervicothoracic junction, dorsal shift and decompression of the spinal cord can be expected by posterior decompression alone.

Surgical method most frequently used for T-OPLL at the upper thoracic spine was laminoplasty. Since the spinal curvature is usually lordotic or only slightly kyphotic at the cervicothoracic junction, dorsal shift and decompression of the spinal cord can be expected by posterior decompression alone (Figure 4), and, therefore, T-OPLL at this level can be successfully treated by laminoplasty, which is relatively safe and is rarely associated with neurologic complications. The use of instrumentation allows correction of kyphosis or prevention of progression of kyphosis, and stabilization of the spine, thereby, enhancing and maintaining decompression effect.<sup>16</sup> Yamazaki *et al*<sup>17</sup> have reported a patient whose neurologic symptoms gradually deteriorated after laminectomy resulting in severe paraplegia. However, the patient obtained neurologic recovery after fusion with posterior instrumentation conducted 4 weeks after the initial decompression surgery. Nakanishi *et al*<sup>18</sup> have reported that a patient who had significant reduction of spinal evoked potential after laminectomy regained the potential level immediately after the addition of posterior instrumentation. Thus, the use of instrumentation should be considered when posterior decompression is conducted.

In the present study, there was no statistically significant difference in the surgical outcomes among patients treated by different surgical methods. Fujimura *et al*<sup>1</sup> and Ohtani *et al*<sup>19</sup> have reported that the anterior decompression is more radical and reasonable for T-OPLL in the kyphotic thoracic spine. On the other hand, Ohtsuka *et al*<sup>8</sup> and Tsuzuki *et al*<sup>20</sup> have reported that they have respectively obtained relatively favorable surgical outcomes using their posterior decompression procedures. Tomita *et al* reported the circumferential decompression method with good surgical outcomes. In the present study, although statistically not significant, the surgical outcomes of Tomita's method tended to be more favorable than those of other surgical methods. Further studies on a greater number of patients are necessary to de-

termine the differences in the clinical outcomes among various surgical procedures.

Surgery for T-OPLL was associated with a high rate of complications. Exacerbation of neurologic symptoms immediately after the surgery and cerebrospinal fluid leakage caused by dural injury were major issues of concern. Diverse explanations have been proposed to explain the neurologic deteriorations immediately after the surgery, including direct spinal cord injury during excision of the ossified lesion, progression of kyphosis after posterior decompression, epidural hematoma, etc. There are also cases in which no specific cause can be identified. As described earlier in this article, laminectomy alone may lead to exacerbation of paralysis in some patients, leading to the recommended use of instrumentation in conjunction with posterior decompression. When complete excision is attempted, unexpected spinal cord injury or cerebrospinal fluid leakage caused by dural injury may develop. Thinning and floating of the ossified lesion, which have been considered to be safer than complete excision, did not reduce the incidence of neurologic complications. Some attempts to make surgery for T-OPLL safer and to improve the surgical outcomes have been made, including the uses of electrophysiologic monitoring, a navigation system during excision of the ossified lesion,<sup>21</sup> and intraoperative ultrasonography for confirmation of decompression during posterior decompression, etc. Although the frequency of neurologic deterioration immediately after the surgery was as high as 11.7%, paralysis can be expected to recover spontaneously to some extent, except for the patients who developed Frankel A paralysis.

The limitations of the present study included retrospective natures of the study, the small number of patients despite the large number of participating institutions, great variations of surgical methods among institutions, all of which may make reliable statistical analysis difficult. Nonetheless, this is the largest study of surgically treated patients with T-OPLL that is a rare disease, and the results are expected to provide some guidelines for selection of the surgical treatment method in the patients with different types and levels of T-OPLL. The results may also serve as basic data for prospective studies that are planned in the near future.

#### ■ Key Points

- Multi-institutional retrospective study of surgically treated patients with ossification of posterior longitudinal ligament in the thoracic spine was conducted.
- The mean recovery rate of Japanese Orthopedic Association Scores was 36.8% ± 47.4%.
- Factors significantly related with favorable surgical outcomes were location of the maximum ossification at the upper thoracic spine and use of spinal instrumentation.

- Multi-institutional retrospective study of 154 surgically treated patients with ossification of posterior longitudinal ligament in the thoracic spine was conducted.
- Factors significantly related with favorable surgical outcomes were location of the maximum ossification at the upper thoracic spine and the use of spinal instrumentation.
- T-OPLL at the upper thoracic spine can be treated safely by laminoplasty.
- The use of instrumentation should be considered with posterior decompression for T-OPLL at the middle and lower thoracic spine.

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## Cervical myelopathy in patients with ossification of the posterior longitudinal ligament

### Clinical article

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**Object.** The authors assessed the clinical course in patients with a narrowed cervical spinal canal caused by ossification of the posterior longitudinal ligament (OPLL), but who have no or only mild myelopathy. Additionally, the authors analyzed the factors contributing to the development and aggravation of myelopathy in patients with OPLL-induced spinal canal stenosis.

**Methods.** Between 1997 and 2004, the authors selected treatments for patients with cervical OPLL in whom the residual space available for the spinal cord was  $\leq 12$  mm. Treatment decisions were based on the severity of myelopathy at presentation. Twenty-one patients with no or mild myelopathy (defined as a Japanese Orthopaedic Association [JOA] scale score  $\geq 14$  points) received conservative treatment, with a mean follow-up period of 4.5 years. In 20 patients with moderate or severe myelopathy (JOA scale score  $< 14$  points), the authors performed surgery via an anterior approach. The clinical course in these patients was assessed with the JOA scale and the OPLL types were classified. The authors evaluated the range of motion between C-1 and C-7, the developmental segmental sagittal diameter, the percentage of spinal canal diameter occupied by the OPLL (% ratio), and the residual space available for the spinal cord on cervical radiographs; T2-weighted MR images were examined for high signal changes (HSCs).

**Results.** In the conservative treatment group, 8 patients showed improvement, 12 remained unchanged, and 1 patient's condition became slightly worse during the observation period. Fifteen patients in this group had mixed-type, 3 had continuous-type, 2 had localized-type, and 1 had a segmental-type OPLL. In the surgically treated group, there were 12 patients with segmental-type, 10 patients with mixed-type, and 1 with localized-type OPLL. The mean range of motion at C1-7 was  $36.4^\circ$  in the conservatively treated group and  $46.5^\circ$  in the surgical group ( $p < 0.05$ ). No significant difference was seen between the groups in terms of developmental segmental sagittal diameter, % ratio, or residual space available for the cord. No HSCs were noted in the conservative group, while 17 patients in the surgical group had HSCs ( $p < 0.05$ ).

**Conclusions.** In the present study, the authors demonstrate that the mobility of the cervical spine and the type of OPLL are important factors contributing to the development and aggravation of myelopathy in patients with OPLL-induced spinal canal stenosis. The authors advocate conservative treatment in most patients with OPLLs who have no or only mild myelopathy, even in the presence of spinal canal narrowing. (DOI: 10.3171/2008.10.SPI08480)

**KEY WORDS** • myelopathy • ossification • posterior longitudinal ligament • stenosis

**I**N patients with OPLL of the cervical spine, the heterotopic ossified mass frequently occupies a considerable area of the spinal canal, resulting in a decrease in space available for the spinal cord (Fig. 1 upper). When compression myelopathy secondary to OPLL is

severe, most surgeons suggest surgical decompression of the spinal cord from an anterior and/or posterior approach.<sup>6</sup> The optimal treatment for patients with cervical OPLL and decreased space for the cord who have no or only mild myelopathy has been controversial, however. Some authors have advocated early surgery in patients with mild myelopathy,<sup>12</sup> and some have even suggested prophylactic surgery in patients without myelopathy.<sup>2</sup> In contrast, other authors have recommended conservative treatment.<sup>7</sup> In deciding on the best treatment for a particular patient with cervical OPLL who has no or only mild

Abbreviations used in this paper: DSSD = developmental segmental sagittal diameter; HSC = high signal change; JOA = Japanese Orthopaedic Association; OPLL = ossification of the posterior longitudinal ligament; ROM = range of motion.

## Cervical myelopathy in patients with OPLL

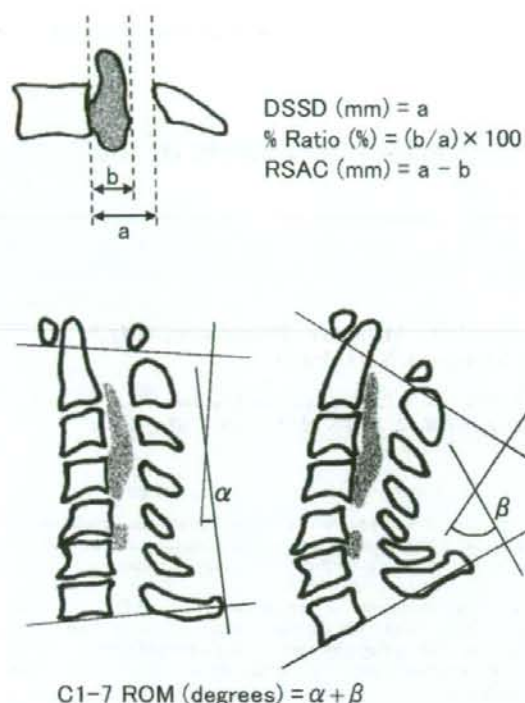


Fig. 1. Schematic drawings of radiographic parameters. In lateral cervical radiographs (upper), we measured the DSSD, % ratio, and the residual space available for the spinal cord (RSAC). In flexion and extension radiographs (lower), we evaluated the ROM between C-1 and C-7.

myelopathy, it is important to predict the neurological course as precisely as possible. To date, however, there have been only a few reports on neurological changes in patients who receive conservative management of their disease.<sup>10</sup> In addition, the factors that contribute to myelopathy in patients with cervical OPLL and spinal canal narrowing have yet to be fully established.

Since 1997, we have selected treatments for patients with cervical OPLL based principally on the severity of myelopathy at the initial consultation, regardless of the degree of residual space available for the cord. In patients with OPLL who have no or only mild myelopathy (cervical myelopathy JOA scale score  $\geq 14$  points), we have taken a conservative approach. In patients with moderate or severe myelopathy (JOA scale score  $< 14$  points), we have performed surgery via an anterior approach.

The purpose of the present study was to assess the clinical course in patients with spinal canal narrowing due to OPLL, but who have no or mild myelopathy. We also investigated the factors contributing to the development and aggravation of myelopathy in patients with OPLL-induced spinal canal stenosis. Based on our results, we discuss the appropriate timing of surgery in patients with cervical OPLLs.

TABLE 1: Clinical data\*

Characteristic	Conservative Tx Group (21 patients)	Surgical Tx Group (20 patients)
sex		
male	12	16
female	9	4
age at 1st visit (yrs)	61.1 $\pm$ 8.7 (48-75)	—
age at op (yrs)	—	59.4 $\pm$ 10.4 (42-74)
FU period (yrs)		
1st visit to final FU	4.2 $\pm$ 2.7 (1-10)	—
op to final FU	—	3.6 $\pm$ 0.8 (3-5)

\* Except as otherwise noted, values are expressed as the means  $\pm$  SDs with the range in parentheses. Abbreviation: FU = follow-up.

## Methods

### Patient Population

Between January 1997 and December 2004, 99 patients with cervical OPLLs in whom the residual space available for the cord at the cervical spine was  $\leq 12$  mm visited our institute for an initial consultation. We selected a treatment procedure for these patients based on extent of myelopathy: patients with no or mild myelopathy (JOA scale score  $\geq 14$  points) were followed up conservatively, and patients with moderate or severe myelopathy (JOA scale score  $< 14$  points) underwent surgical treatment.

Of our 99 patients, 21 had a JOA scale score  $\geq 14$  points at the initial consultation and received conservative treatment (Table 1). The conservative group included 12 men and 9 women, with a mean age at initial consultation of 61.1 years (range 48-75 years). The mean follow-up period from the first visit to the final examination was 4.2 years (range 1-10 years). Eleven patients underwent follow-up for  $< 5$  years and 10 patients for  $\geq 5$  years. Of the 99 patients, 78 had JOA scale scores  $< 14$  points; these patients underwent surgical treatment via an anterior approach. To match the 2 groups in terms of numbers, we specifically chose patients who underwent surgery between 2002 and 2004. Of these 24 patients, 20 patients attended follow-up for  $\geq 1$  year and were included in our final analysis. The surgery group included 16 men and 4 women with a mean age at surgery of 59.4 years (range 42-74 years). In all 20 patients, surgeries were performed with anterior excision of OPLL foci and placement of an arthrodesis with a strut autograft using either fibula or iliac crest. After surgery, these patients attended follow-up for a mean of 3.6 years (range 3-5 years).

### Clinical Assessment

The JOA scale was used to evaluate the severity of cervical myelopathy.<sup>6</sup> Patients with mild myelopathy who increased their JOA scale score by  $\geq 1$  point were said to have improved, and those with a decrease of  $\geq 1$  point

TABLE 2: Clinical results

Parameter	Conservative Tx Group (21 patients)	Surgical Tx Group (20 patients)
JOA scale score		
1st visit	15.6 ± 0.9 (14–17)	—
before op	—	10.2 ± 2.3 (6–13.5)
at FU	16.4 ± 1.0 (14.5–17)	15.1 ± 1.8 (10–17)
postop recovery ratio (%)	—	74.6 ± 20.6 (30.8–100)
outcome after conservative tx*		
improved (%)	8 (38.1)	—
unchanged (%)	12 (57.1)	—
worsened (%)	1 (4.8)	—

\* Improvement was defined as a ≥ 1-point gain in JOA scale score, unchanged status was gain or loss of < 1 point, and worsening was defined as the loss of > 1 point. Except as when otherwise noted, values are presented as means ± SDs with the range in parentheses.

were said to have worsened. When JOA scale scores increased or decreased by < 1 point, this was recorded as unchanged. In surgically treated patients, scores obtained before surgery and at the final follow-up examination were evaluated and a recovery rate was calculated.<sup>6</sup>

#### Radiographic Assessment

Using lateral cervical radiographs, we measured the DSSD, the percentage of the spinal canal diameter occupied by the OPLL (% ratio), and the residual space available for the cord (Fig. 1 upper). The types of OPLL were classified as continuous type, mixed type, segmental type, and localized type, according to the criteria of the Japanese Investigation Committee on the Ossification of the Spinal Ligaments.<sup>14</sup> We evaluated the ROM between C-1 and C-7 on flexion and extension radiographs (Fig. 1 lower), and spinal cord HSCs were assessed on T2-weighted MR images.

#### Statistical Analysis

The Mann-Whitney U-test and Fisher exact probability test were applied for statistical analyses. Probability values < 0.05 were considered significant; results are presented as the means ± SDs.

### Results

Of the 21 patients in the conservative treatment group, 6 showed no sign of myelopathy and 15 had mild myelopathy at the initial consultation. In the 6 patients without myelopathy, radiographs were originally obtained because the patients complained of neck stiffness or pain. All 6 remained neurologically unchanged during

TABLE 3: Radiographic data at the initial consultation\*

Parameter	Conservative Tx Group	Surgical Tx Group
OPLL type (no. of patients)		
continuous	3	0
mixed	15	7
segmental	1	12†
localized	2	1
DSSD (mm)		
mean ± SD	14.9 ± 1.3	15.0 ± 1.1
range	13–17	13–17
% ratio		
mean ± SD	46.6 ± 11	40.0 ± 12
range	25–63	25–61
RSAC (mm)		
mean ± SD	7.9 ± 1.5	9.3 ± 1.9
range	6–11	6–12
C1–7 ROM (°)		
mean ± SD	36.4 ± 14.0	46.5 ± 9.3†
range	15–62	26–64
ROM (no. of patients)		
≥35°	11	19†
<35°	10	1
HSC status (no. of patients)		
negative	21	3
positive	0	17†

\* RSAC = residual space available for cord.

† Indicates statistically significant difference compared to the conservative treatment group ( $p < 0.05$ ).

the follow-up period. Of the 15 patients with myelopathy, 8 showed improvement, 6 remained unchanged, and in 1 patient the myelopathy worsened. The mean JOA scale score at the initial consultation in the conservatively treated group was 15.6 points (range 14–17 points), and 16.4 points at follow-up (range 14.5–17 points) (Table 2). In the surgical group, the mean preoperative JOA scale score was 10.2 points (range 6–13.5 points) and 15.1 points at the most recent follow-up (range 10–17 points). The mean recovery rate was 74.6% (range 30.8–100%).

Cervical radiographs and MR images obtained at the initial consultation were analyzed to clarify the morphological characteristics of the OPLLs. In the conservative treatment group, 15 patients had mixed-type, 3 had continuous-type, 2 had localized-type, and 1 had a segmental-type OPLL. In the surgical treatment group 12 patients had segmental-type, 7 had mixed-type, and 1 had a localized-type OPLL (Table 3). The incidence of segmental-type OPLL was significantly higher in the surgical than the conservative treatment group ( $p < 0.05$ ).

The mean DSSD was 14.9 mm (range 13–17 mm) in the conservative group and 15 mm (range 13–17 mm) in the surgically treated group. The % ratio was 46.6%