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CASE REPORT

Recurrence of ossification of ligamentum flavum at the same intervertebral level in the thoracic spine: a report of two cases and review of the literature

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Abstract
Purpose Ossification of the ligamentum flavum (OLF) is a possible cause of thoracic myelopathy. We report two rare cases with recurrent thoracic myelopathy caused by OLF markedly re-extended at the same intervertebral level after the primary surgery.
Methods Both patients had thoracic myelopathy caused by OLF and underwent decompressive laminectomy and resection of the OLF in the primary surgery. However, the neurological conditions gradually deteriorated following recovery after the primary surgery due to the recurrent OLF at the same intervertebral level.
Results These patients were successfully treated by revision surgery via resection of the recurrent OLF and posterior spinal fusion with instrumentation. Two years after the second surgery, the neurological disturbance was resolved satisfactorily, and re-growth of the resected ossified lesion was not observed.
Conclusions The recurrence of OLF following resection of the ossified lesions is exceedingly rare but should be noted in patients treated surgically for thoracic myelopathy due to OLF.

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Keywords Ossification of ligamentum flavum · Recurrence · Thoracic spine · Myelopathy · Posterior longitudinal ligament

Introduction

Ossification of the ligamentum flavum (OLF) is a possible cause of thoracic myelopathy [1–3]. Thoracic myelopathy caused by OLF typically progresses gradually, and surgical treatment such as decompressive laminectomy has been essentially indicated. The outcomes after surgical decompression for thoracic myelopathy due to OLF are generally good if the diagnosis is made promptly and the surgery is performed appropriately before severe neurological deficits develop [1, 2].

Previous studies have reported the recurrence of thoracic myelopathy caused by the occurrence of OLF at other intervertebral levels than that decompressed in the primary surgery [2, 4–8]. However, to our knowledge, there have been only five reported cases with recurrent OLF at the same intervertebral level after the primary surgery [4, 6, 9, 10]. The clinical features of recurrent OLF at the same intervertebral level are not well understood and the optimal treatment strategy still remains unknown.

We herein report two cases with recurrent thoracic myelopathy caused by OLF markedly re-extended at the same intervertebral level after decompressive laminectomy in the primary surgery. In these cases, the neurological conditions gradually deteriorated after sufficient recovery following the primary surgery. Both cases were successfully treated by revision surgery via resection of the recurrent OLF and posterior spinal fusion with instrumentation. In addition, we review the literatures focusing on the recurrence of OLF at the same intervertebral level

following the primary surgery. The patients were informed that their clinical data would be submitted for publication and gave their consent.

Case reports

Case 1

History

A 47-year-old female presented to a regional hospital with weakness in her bilateral lower extremities and difficulty walking. Muscle weakness was detected in the right iliopsoas and quadriceps femoris muscles. Her patella tendon reflex and Achilles tendon reflex were exaggerated bilaterally. Hyperalgesia was observed in the bilateral lower extremities and in the inguinal regions. She was neurologically diagnosed as having thoracic myelopathy. The score of the modified Japanese Orthopaedic Association (JOA) scoring system for thoracic myelopathy [2] was 2 of 11 points. Magnetic resonance imaging (MRI) showed that the spinal cord was highly compressed at T9–10 and T10–11 levels (Fig. 1). Computed tomography (CT) showed OLF at multiple intervertebral levels, from T9–10 to T12–L1. There was no ossified lesion of the spinal ligament below the T12–L1 level. Decompressive laminectomy and resection of the OLF from T9–10 to T12–L1 were performed at the regional hospital (Fig. 2). Her symptoms sufficiently recovered after the surgery. The JOA score improved to 9 of 11 points. On the imaging studies before and after the initial surgery, there was no evidence of the instability or hypermobility at the affected

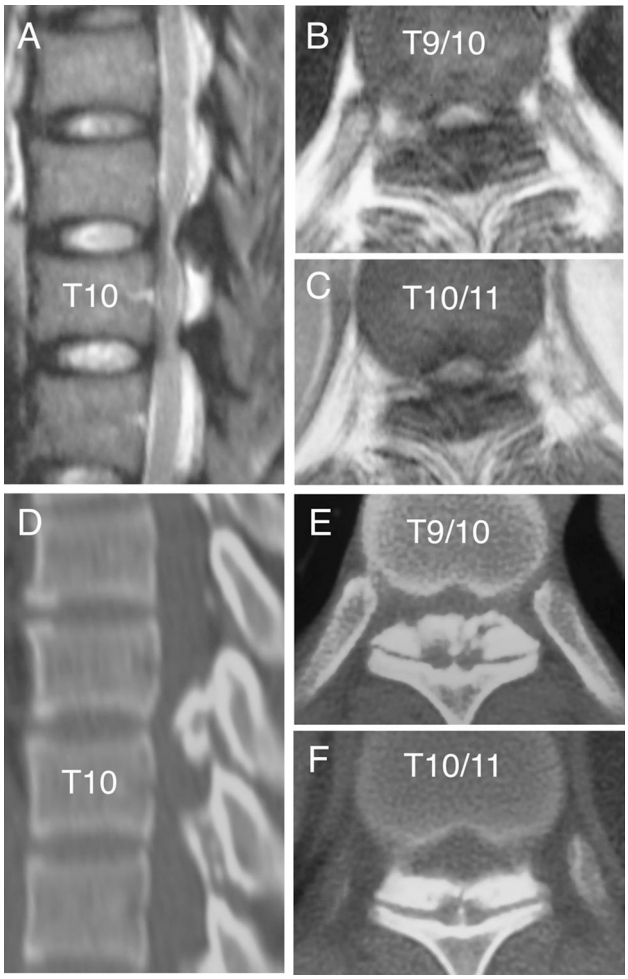

Fig. 1 Case 1. MR images and CT scans obtained before the first operation. Sagittal (**a**) and axial (**b**, **c**) T2-weighted images show the compressed spinal cord at the T9–10 and T10–11 intervertebral levels. Sagittal reconstruction (**d**) and axial (**e**, **f**) CT scans show the OLF at the T9–10 and T10–11 levels

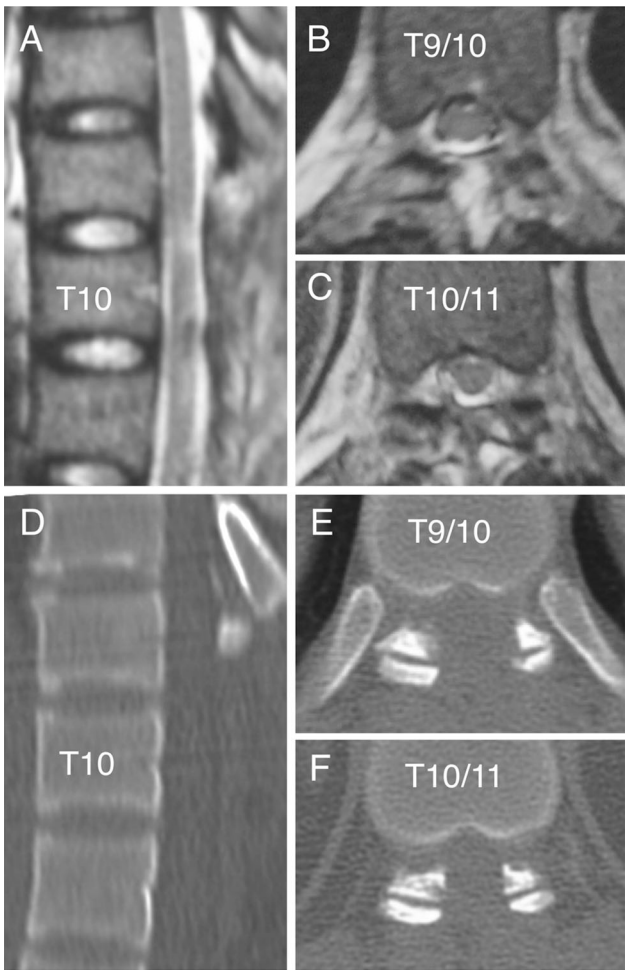

Fig. 2 Case 1. MR images and CT scans obtained after the first operation. Sagittal (**a**) and axial (**b**, **c**) T2-weighted images show the presence of cerebrospinal fluid around the spinal cord at the T9–10 and T10–11 levels, and the spinal cord was sufficiently decompressed. Sagittal reconstruction (**d**) and axial (**e**, **f**) CT scans show the OLF in the spinal canal, and the medial part of the facet joint was adequately resected at the T9–10 and T10–11 levels

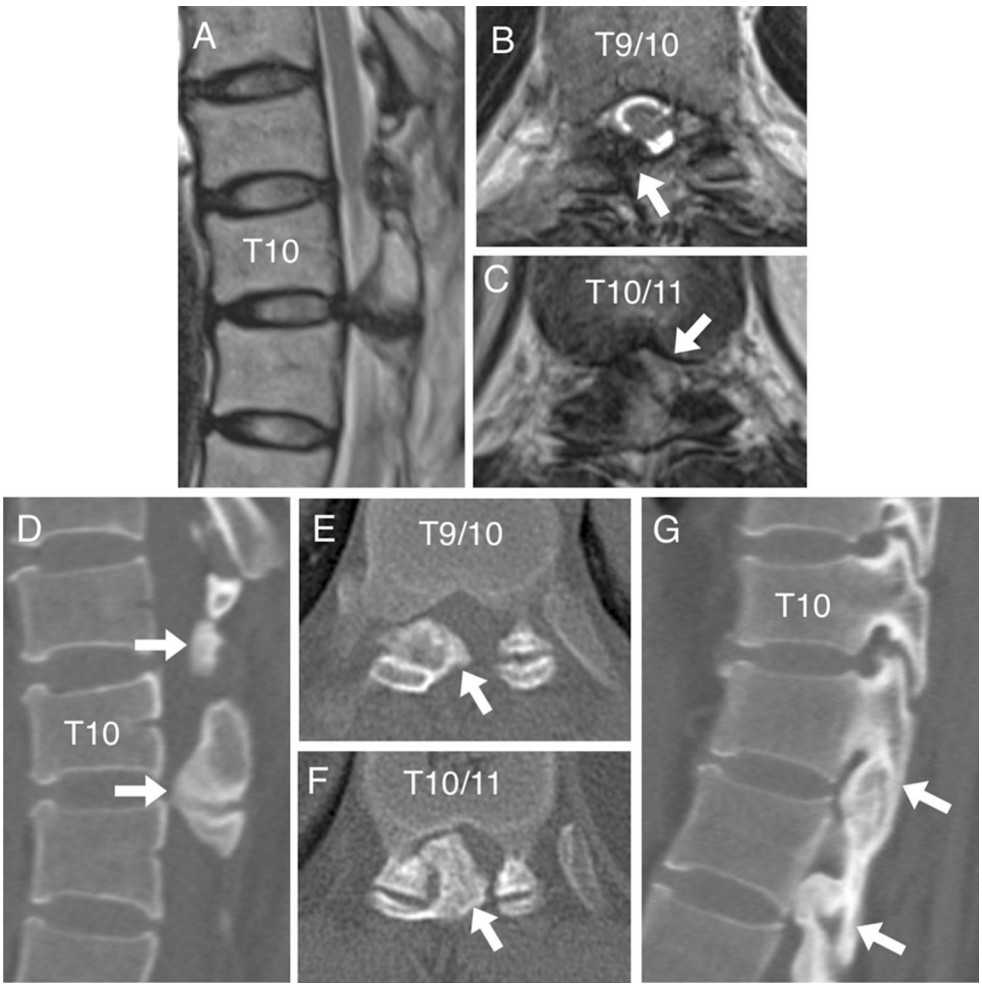

Fig. 3 Case 1. MR images and CT scans obtained before the second operation. Sagittal (**a**) and axial (**b**, **c**) T2-weighted images show the compressed spinal cord at the T9–10 and T10–11 intervertebral levels (*arrows* in **b**, **c**). Sagittal reconstruction (**d**) and axial (**e**, **f**) CT scans show the OLF markedly re-extended on the right side at the T-9–10 and T10–11 levels (*arrows* **d**–**f**). Additionally, a sagittal reconstruction (**g**) CT scan shows that the facet joints are fused at the T11–12 and T12–L1 levels (*arrows* in **g**) but not at the T10–11 level

intervertebral levels such as the increased facet joint width between the superior and inferior facets. On flexion and extension lateral radiographs of the thoracic spine, the segmental range of motions at the affected levels were a few degrees both before and after the initial surgery.

Presentation and examination

Eleven years after the initial surgery, she was referred to our hospital due to the recurrence of weakness in her bilateral lower extremities. Decreased muscle strength, hyperreflexia and hyperalgesia in the lower extremities were found again, just as before the initial surgery. The neurological diagnosis was the recurrence of thoracic myelopathy. The JOA score was 4 of 11 points.

MRI showed that the spinal cord was compressed and displaced to the left side due to the recurrence of OLF at the T10–11 level (Fig. 3). On CT, the recurrence of OLF from the T9–10 to T12–L1 intervertebral levels was observed, and the recurrent OLF at the T10–11 level was largely protruding into the spinal canal, particularly on the right side (Fig. 3). The facet joints were fused at the T11–12 and T12–L1 levels.

Operation

We resected the recurrent OLF at the T9–10 and T10–11 levels using a navigation system to accurately identify the positions between the spinal canal and the ossified lesion during surgery. The OLF at the T10–11 was strongly adhered to the dura mater, and therefore, the ossified lesion was removed with the dura mater (Fig. 4). The dura mater was repaired using an artificial patch. The facet joint on the right side at the T10–11 level was widely resected to remove the re-ossified lesion safely, and the posterior fusion with instrumentation from T8 to L2 was added. Both

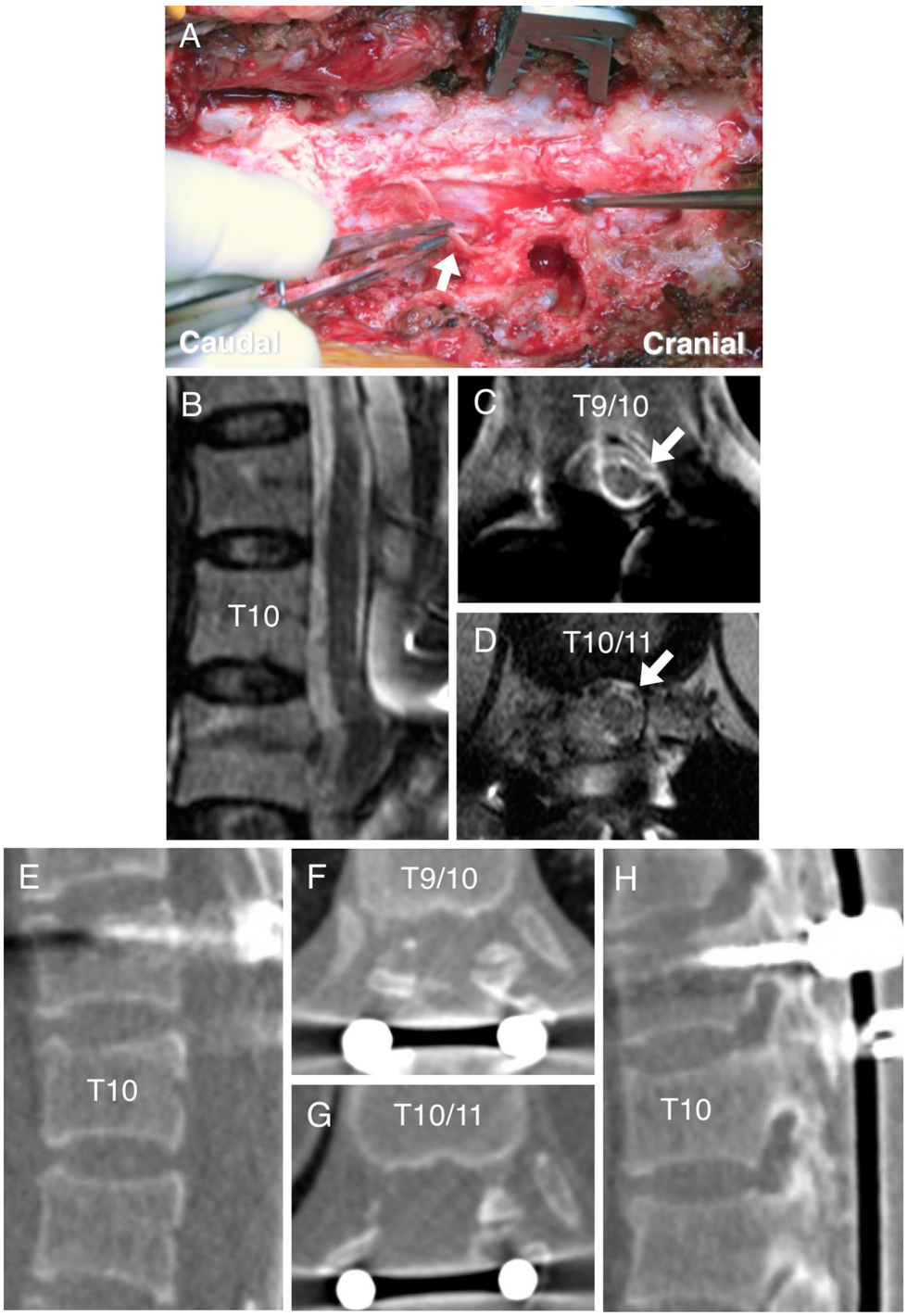

Fig. 4 Case 1. Intraoperative photograph and MR images and CT scans obtained after the second operation. The recurrent OLF was strongly adhered to the dura mater, and therefore, the ossified lesion was removed with the dura mater at the T10–11 level (*arrow* in **a**). Sagittal (**b**) and axial (**c**, **d**) T2-weighted images show that the spinal cord is sufficiently decompressed at the T9–10 and T10–11 levels after the second operation (*arrows* in **c**, **d**). Sagittal (**e**) and axial CT scans (**f**, **g**) show that the OLF in the spinal canal and the facet joint on the right side are widely resected. Additionally, sagittal CT scan on the *left side* (**h**) shows the facet joints are fused at the T9–10 and T10–11

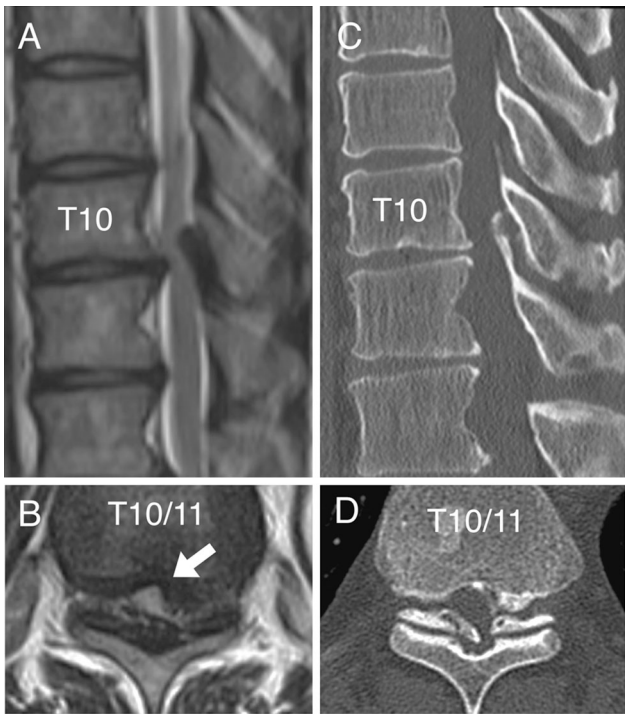

Fig. 5 Case 2. MR images and CT scans obtained before the first operation. Sagittal (**a**) and axial (**b**) T2-weighted images show the compressed spinal cord at the T10–11 intervertebral level (*arrow* in **b**). Sagittal reconstruction (**c**) and axial (**d**) CT scan shows OLF and OPLL at T10–11 level

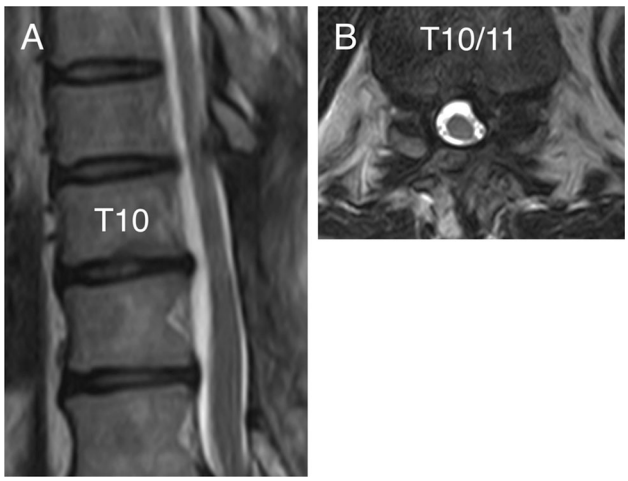

Fig. 6 Case 2. MR images obtained after the first operation. Sagittal (**a**) and axial (**b**) T2-weighted images show the presence of cerebrospinal fluid around the spinal cord at the T10–11 level, and the spinal cord was sufficiently decompressed

autografts from the iliac bone and local bone were used to achieve the bony fusion of the lateral side of the facet joints.

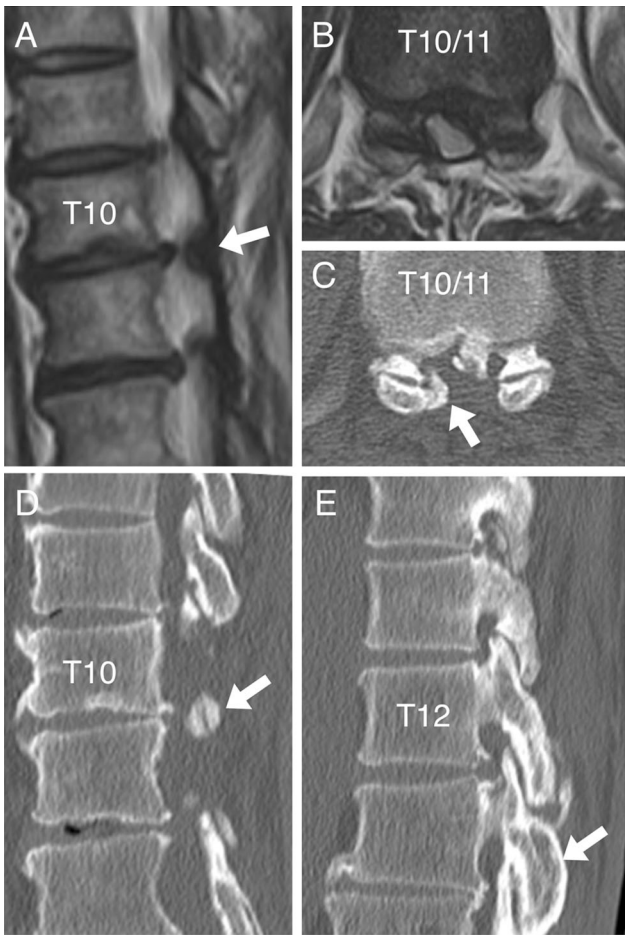

Fig. 7 Case 2. MR images and CT scans obtained before the second operation. Sagittal (**a**) and axial (**b**) T2-weighted images show the compressed spinal cord at the T10–11 intervertebral level (*arrow* in **a**). Axial (**c**) and sagittal reconstruction (**d**) CT scans show the OLF markedly re-extended on the right side at the T10-11 levels (*arrows* in **c** and **d**). Additionally, a sagittal reconstruction (**e**) CT scan shows the facet joints are fused below the T12-L1 level (*arrow* in **e**)

Postoperative course

MRI after the reoperation revealed the sufficiently decompressed spinal cord at the T10–11 level (Fig. 4). Her symptoms were relieved, and her muscular strength was improved. The JOA score improved to 5 of 11 points at 2 years after the reoperation.

Case 2

History

A 54-year-old male presented to a regional hospital with weakness of his bilateral lower extremities. Weakness of iliopsoas muscle and exaggerated patella tendon reflex and Achilles tendon reflex were detected on bilateral sides. Hypoalgesia was found in the inguinal regions and the

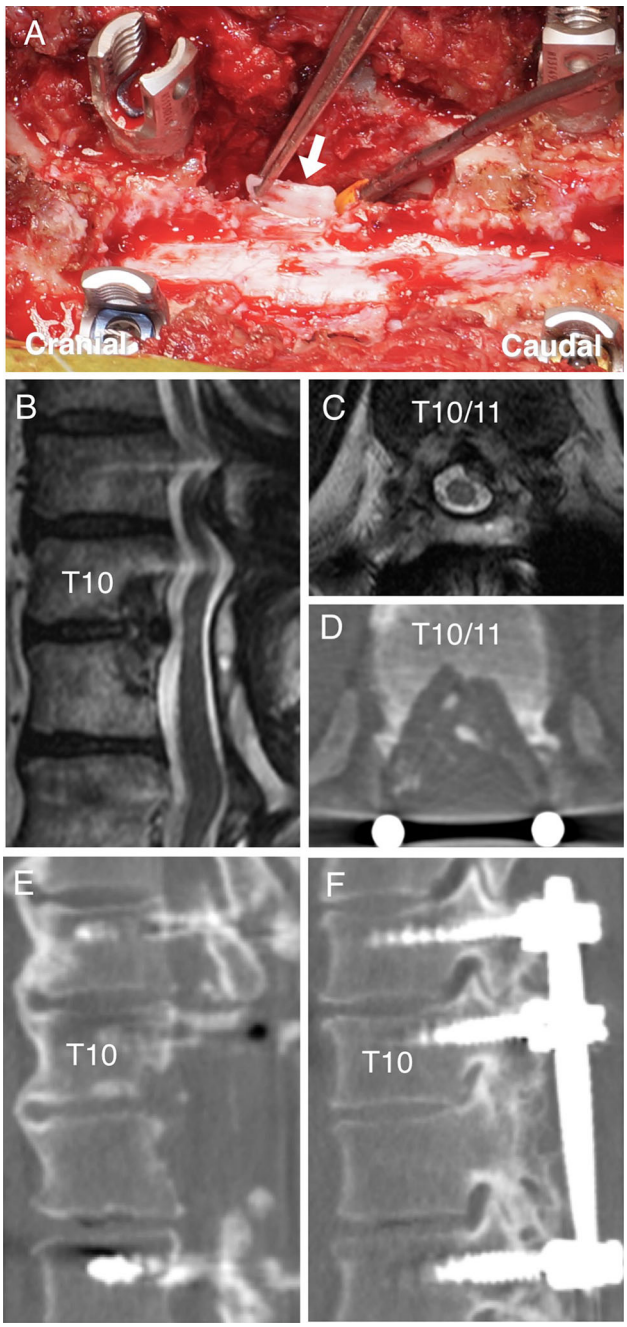

Fig. 8 Case 2. Intraoperative photograph and MR images and CT scans obtained after the second operation. The recurrent OLF was removed at the T10–11 level (*arrow* in **a**). Sagittal (**b**) and axial (**c**) T2-weighted images show that the spinal cord is sufficiently decompressed at the T10–11 level after the second operation. An axial (**d**) and sagittal (**e**) CT scan shows that the OLF and OPLL as well as the medial facet joints on the bilateral sides are resected. Sagittal CT scan on the *left side* (**f**) shows the facet joints are fused at the T10–11 and T11–12

bilateral lower extremities. His neurological diagnosis was thoracic myelopathy. The JOA score was 9 of 11 points. MRI revealed the compressed spinal cord at the T10–11 level (Fig. 5). CT showed the OLF and ossification of the posterior longitudinal ligament (OPLL) at the T10–11 level. In addition, segmental OPLL from the L1–2 to L4–5 and OLF at the L4–5 and L5–S1 caused narrowing of the lumbar spinal canal at the multiple intervertebral levels. Decompressive laminectomy and resection of the OLF at the T10–11 level were performed at the regional hospital (Fig. 6). Posterior decompression and fusion were also performed at the L1–5 levels. The JOA score improved to 11 of 11 points. On the imaging studies before and after the initial surgery, there was no evidence of the instability or hypermobility at the affected intervertebral levels. On flexion and extension lateral radiographs of the thoracic spine, the segmental range of motions at the affected levels were a few degrees both before and after the initial surgery.

Presentation and examination

Four years after the initial surgery, he was referred to our hospital due to the recurrence of weakness in his bilateral lower extremities and difficulty walking. Neurological findings similar to prior to the initial surgery were observed, and the recurrence of thoracic myelopathy was considered. His JOA score was 7 of 11 points.

MRI showed that the spinal cord was compressed at the T10–11 level (Fig. 7). On CT, the recurrence of the OLF and an enlargement of OPLL were found at the T10–11 level. In addition, posterior fusion due to the initial surgery was detected at the L1–5 level.

Operation

In the second surgery, we resected the recurrent OLF at the T10–11 level (Fig. 8) and removed the OPLL by the Otsuka method [11]. The facet joints were widely resected at the T10–11 to remove the re-ossified lesion safely, and the posterior fusion with instrumentation from T9 to L1 was added. Both autografts from the iliac bone and local bone were used to achieve the bony fusion of the lateral side of the facet joints.

Postoperative course

His neurological symptoms were sufficiently recovered. The JOA score improved to 10 of 11 points at 2 years following the second surgery.

Discussion

The prevalence of OLF in the thoracic spine observed on CT scan is 26–63.9% among the normal population [3, 12, 13]. A portion of the population with OLF in the thoracic spine has neurological symptoms of thoracic

Table 1 Summary of reported cases of recurrent OLF at the same intervertebral level in the thoracic spine

References	Age (years)	Sex	Level	Initial surgery	Regression (months)	Second surgery	Condition of the adjacent segments
Yonenobu et al. [6]	ND	ND	ND	Decomp	66	Decomp	ND
Okada et al. [10]	ND	ND	ND	Decomp	ND	Decomp	ND
	ND	ND	ND	Decomp	ND	Decomp	ND
Ando et al. [9]	36	M	T10/11	Decomp	11	Decomp and fusion	After posterior fusion of upper and lower segments in initial surgery
Nakata et al. [4]	ND	M	T11/12	Decomp	96	Decomp	Spontaneous fusion of upper and lower segments with OALL
Present report	47	F	T10/11	Decomp	132	Decomp and fusion	Spontaneous facet fusion of lower segments
	54	M	T10/11	Decomp	48	Decomp and fusion	After posterior fusion of lower segments in initial surgery

Age age (years) at initial surgery, *F* female, *M* male, *Level* level of recurrent OLF, *Time to regression* time (months) to regression after the initial surgery, *Decomp* posterior decompression, *Decomp and fusion* posterior decompression and fusion with instrumentation, *ND* no description

myelopathy. The number of patients with thoracic myelopathy due to OLF is 1.6 per 100,000 people annually in Japan [14]. According to our registration database of spinal surgery at Tohoku University Spine Society [1], the annual number of patients who underwent surgery for thoracic myelopathy due to OLF was 0.6 per 100,000 people. For surgical treatment of thoracic myelopathy caused by OLF, decompressive laminectomy without fusion has been essentially indicated and the long-term clinical outcomes are generally good [1]. Accordingly, thoracic myelopathy due to the recurrence of OLF at the same intervertebral level is exceedingly rare. To our knowledge, only 5 reported cases with recurrent thoracic myelopathy caused by OLF re-extended at the same intervertebral level after the primary decompressive surgery have been reported (Table 1) [4, 6, 9, 10].

The risk factors for OLF are considered to be ageing [3, 12], genetic predisposition [15], metabolic abnormalities [16], mechanical stress [17, 18], and fusion of adjacent intervertebral levels [18, 19]. The ossified lesion has a potential to extend beyond the normal size of the ligamentum flavum and can have various types of morphology [1]. During the initial decompression surgery for OLF, the ligamentum flavum is widely resected to decompress the spinal cord [20]. However, the capsular portion of the ligamentum flavum can partially remain when the lateral part of the facet joints is preserved to prevent segmental instability [1, 21]. The residual ligamentum flavum may contain tissue with the potential for osteogenesis to re-extend the OLF. The OLF should be resected as much as possible to reduce the risk of recurrence of the ossified lesion.

Nakata et al. [4] reported one case who underwent reoperation for recurrent OLF at the same intervertebral level 8 years after posterior decompression of the OLF at the T11–12 level. In this case, ossification of the anterior longitudinal ligament (OALL) was widely observed at adjacent intervertebral levels but not at the T11–12 level. The concentration of mechanical stress at the T11–12 level that was not fused by OALL might be one of the causes of the recurrent OLF [9, 18, 19]. Additionally, it has been reported that OLF markedly developed at adjacent intervertebral levels following primary surgery to perform posterior decompression and fusion for OPLL in the thoracic spine [22, 23]. In our cases, the facet joints were fused at adjacent intervertebral levels but not at the recurrent level in both cases. These reports suggested that not only the genetic predisposition for ossification of the spinal ligaments but also increased local mechanical stress due to segmental fusion at the adjacent levels might be a possible cause of enlargement of the OLF.

In both of our two cases, the OLF recurred at the thoracolumbar junction as the same as previous reported cases [4, 9]. The intervertebral range of motion at the thoracolumbar junction is generally larger than that of other region of the spine [24]. The concentrations of mechanical stress on the focal site caused by the fusion of adjacent levels and the larger intervertebral range of motion at the thoracolumbar junction might have caused the re-extension of the OLF.

The neurologic manifestations of thoracic myelopathy can be exacerbated by not only static spinal cord compression caused by the ossified lesions but also the intervertebral mobility placing mechanical stress on the spinal cord [19, 25]. Therefore, the decompression of the spinal

cord as well as the spinal fusion with instrumentation to obtain stability of the lesion can help to improve the neurologic manifestation [9, 25]. In the revision surgery for our two cases, the facet joints at the affected level had to be widely resected to remove the re-ossified lesion safely, and therefore the spinal fusion with instrumentation was needed to prevent postoperative segmental instability. Consequently, the neurological symptoms were sufficiently improved after the reoperation. The spinal fusion using instruments stabilized the intervertebral level at the lesion and may have consequently contributed to the neurological recovery following the revision surgery. The spinal fusion with instrumentation to reduce the focal mechanical stress of the affected intervertebral level may additionally help to prevent the re-extension of the OLF after decompressive surgery [9].

Overall, a combination of spinal cord decompression and spinal fusion with instrumentation might be considered to obtain optimal neurological recovery and to prevent re-extension of the OLF in the case that the OLF is located at the thoracolumbar junction and the adjacent levels are widely fused preoperatively, and postoperative segmental instability due to wide bony resection is highly expected.

Conclusion

We herein reported two extremely rare cases successfully treated by revision surgery for a recurrent OLF at the same intervertebral level in the thoracic spine after the primary surgery. The possibility of the recurrence of OLF following resection of the ossified lesion should be noted in patients with OLF in the thoracic spine.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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