Cervical Ligamentum Flavum Ossification

—Two Case Reports—

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Abstract

A 58-year-old woman (Case 1) presented with disturbance of fine movement and gait. Magnetic resonance (MR) imaging and computed tomography (CT) demonstrated bilateral ossified ligamentum flava at the C3-4 and C4-5 levels and severe cervical canal stenosis. She underwent posterior decompression and despite strong adhesion to the dura mater, the ossified ligamentum flavum was removed without inducing liquorrhea. Her neurological symptoms improved postoperatively. A 63-year-old man (Case 2) was admitted with disturbance of fine movement and gait that had developed gradually. MR imaging and postmyelography CT demonstrated cervical canal stenosis via the ossified posterior longitudinal ligament at the C4-6 levels and ossified ligamentum flavum on the right at the C4-5 levels. He underwent right posterior decompression of C4-5. After right hemilaminectomy of C4-5, the ligamentum flavum was exposed. Neither the ligamentum flavum nor the ossification had adhered to the dura mater, so complete removal was successful and he recovered completely from his neurological symptoms. Diffuse-type ossified ligamentum flavum had adhesion to the dura mater, as in our Case 1, whereas focal-type ossified ligamentum flavum did not, as in our Case 2. This information is useful for treatment planning.

Key words: cervical ligamentum flavum, ossification

Introduction

Ossification of the ligamentum flavum usually occurs in the lower thoracic spine of middle-aged men, and is extremely rare in the cervical spine.^{4,24)} Asymptomatic ossified ligamentum flavum occurs in thoracic locations in 38.5%, lumbar in 26.5%, and cervical in only 0.9%.⁵⁾ The high rate of ossified ligamentum flavum in the thoracic spine may be due to the constant tension placed on the ligamentum flavum of the thoracic spine by kyphosis.⁷⁾ In addition, the ligamentum flavum of the thoracic spine is connected to the lumbar spine, which has wide motion, so is exposed to stress. The development of ossified ligamentum flavum of the cervical spine has been attributed to faulty development of the ligamentum flavum at the capsular portion,⁷⁾ and to the effects of the distance between the angle of the intervertebral joint and the right and left interver-

tebral joints.²⁷⁾ The ligamentum flavum does not reach the capsular portion at the C2-3 to C5-6 level, but is developed and reaches the capsular portion at and below the C6-7.⁷⁾

We treated two patients with ossification of the cervical ligamentum flavum manifesting as myelopathy.

Case Reports

Case 1: A 58-year-old woman first experienced numbness in her left hand 3 years earlier which gradually worsened and she also developed numbness in her right hand. She had no relevant past history. She was admitted to our hospital with disturbance of fine movement and gait.

On admission, neurological examination found bilateral numbness at and below the C6 level. The bilateral deep tendon reflexes were increased at and below the level of the triceps. Myoclonus and Babinski reflexes were positive on the right. She reported increased frequency of micturition.

Fig. 1 Case 1. Sagittal (A) and axial (B, C) T₂-weighted magnetic resonance images of the cervical spine at the C3-4 (B) and C4-5 (C) levels showing the low intensity mass at the site of posterior cervical cord compression.

Magnetic resonance (MR) imaging disclosed severe cervical canal stenosis and enlargement of the bilateral ligamentum flava at the C3-4 and C4-5 levels (Fig. 1), and mild stenosis at the C2-3 and C5-6 levels. T₁- and T₂-weighted MR imaging showed the ligamentum flavum as hypointense. Cervical computed tomography (CT) demonstrated bilateral ossification of the ligamentum flavum at C3-4 and C4-5 (Fig. 2). Thoracic and lumbar CT did not reveal ossification of other ligaments.

Conservative therapy failed to improve her neurological deficits, so she underwent posterior decompression. After dome laminectomy of the lower C2, laminectomy of C3-C5 levels was performed. The ligamentum flavum at the C3-4 and C4-5 levels had degenerated and manifested whitish, comparatively tight fibrous membrane in places. The degenerated ligamentum flavum was removed as far as possible to expose the ossified ligament. Drilling showed the ossification was thin and flaked off the dura mater. The ligamentum flavum of C3-4 and C4-5 was connected by a fibrous, tough, whitish membrane. Removal of the C6 lamina revealed normal dura mater, so this membrane and the ossified ligamentum flavum were detached from the dura mater by careful microscopic dissection. There was a connection between the membrane and the ligamentum flavum at the C2-3, which mimicked a fibrous band at

Fig. 2 Case 1. Axial computed tomography scans at the C3-4 (A), C4 (B), C4-5 (C), and C5 (D) levels demonstrating ossified ligamentum flavum causing severe cervical canal stenosis.

the C2 level. Decompression was obtained by opening this band.

Postoperatively, her neurological symptoms improved gradually and her hand numbness resolved at 3 months after surgery. Histological examination of the surgical specimens revealed elastic bundles with endochondral ossification (Fig. 3).

Case 2: A 63-year-old man noted numbness of the left hand 1 year ago, and he developed numbness of the right hand 2 months prior to visiting our hospital. He had no relevant past history. He was admitted for gradual appearance of disturbance of fine movement and gait.

On admission, neurological examination identified disturbance of fine movement and gait, and bilateral numbness at and below the C6 level. Deep tendon reflexes were increased at and below the bilateral biceps. Myoclonus and Babinski reflexes were positive bilaterally. He did not report urinary or bowel dysfunction.

MR imaging revealed cervical canal stenosis at the C4-6 levels and enlargement of the right ligamentum flavum at the C4-5 levels. Postmyelographic CT demonstrated cervical canal stenosis via the ossified posterior longitudinal ligament at the C4-6 levels and the ossified right ligamentum flavum at the C4-5 levels (Fig. 4). In addition, thoracic CT revealed mild ossification of the ligamentum flavum.

Fig. 3 Case 1. Histological examination of the surgical specimen revealing elastic bundles with endochondral ossification. Hematoxylin and eosin stain, $\times 40$.

Fig. 4 Case 2. Axial T₂-weighted magnetic resonance image (A) and computed tomography myelograph (B) at the C4-5 level showing an ossified mass on the right with bony protrusion into the canal.

Fig. 5 Case 2. Histological examination of the surgical specimen showing elastic bundles and bony tissue. Between the two tissues, a transitional state consisting of cartilage was present. Hematoxylin and eosin stain, $\times 5$.

Conservative therapy did not improve his neurological deficits, so we performed right posterior decompression at the C4-5 levels. Right hemila-

minectomy at C4-5 exposed the ligamentum flavum. There was no adhesion to the dura mater and the ligamentum flavum including the ossified ligament was removed totally.

Postoperatively, his neurological symptoms gradually improved. At 3 months after surgery, he showed full recovery of his neurological deficits. Histological examination of the surgical specimens showed intraligamentous ossification (Fig. 5).

Discussion

A total of 25 cases of cervical ossified ligamentum flavum, including ours, have been reported (Table 1).^{1-3,6,8-17,19-23,26,27} The patients were aged from 27 to 75 years (mean 57 years), 14 were male, and 22 had Japanese ancestry. In contrast, 80% or more cases of thoracic ossified ligamentum flavum occur in males.^{4,24} Bilateral involvement was noted in 15 of the 25 cases, the right side was affected in four and the left in six cases. Multilevel ossified ligamentum flavum, as in our Case 1, was reported in seven patients.

The ligamentum flavum at or below the C6-7 level extends to the capsular portion.⁷ Inadequate development of the cervical ligamentum flavum has been postulated in patients with cervical ossified ligamentum flavum.⁷ If this hypothesis is correct, the incidence should be higher at and above the C6-7 level. In the 25 reported cases, the ossified ligamentum flavum was located at C1-2 in one patient, C2-3 in seven, C3-4 in six, C4-5 in five, C5-6 in four, C6-7 in seven, and C7-T1 in two. Whether this slight variation in location reflects anatomical differences remains unclear. The C6-7 level was involved in four of the seven cases in which the ossified ligamentum flavum extended to the capsular portion. Thoracic ossified ligamentum flavum extends to the lateral capsular portion, so sufficient lateral decompression is required by drilling.²⁴ In contrast, removal of cervical ossified ligamentum flavum that does not extend to the capsular portion requires great care. Only one case of cervical ossified ligamentum flavum manifested as radiculopathy, possibly because of only slight ossification of the capsular portion. Most cases of ossified ligamentum flavum have developed by myelopathy regardless of focal or diffuse, as in our cases.

The outcome for all previously reported patients treated by excision of the ossified ligamentum flavum was excellent although all but one patient manifested myelopathy. The surgical outcome in our patients, both of whom experienced comparatively rapid myelopathy, was also good. The preoperative symptom duration associated with thorac-

Table 1 Reported cases of ossification of the cervical ligamentum flavum

Author (Year)	Sex/ Age (yrs)	Ancestry	Level	Extension to capsular portion	Other ossifica- tion	Site	Type	Adhe- sion
Kirita et al. (1973) ¹⁰⁾	F/27	Japanese	C6-7, C7-T1	—	?	bil	?	?
Nagashima (1975) ¹⁷⁾	M/64	Japanese	C6-7	+	+	lt	?	?
Kaneda et al. (1977) ⁹⁾	M/46	Japanese	C2-3, C3-4	?	+	bil	?	?
Kamakura et al. (1978) ⁸⁾	F/61	Japanese	C5-6, C6-7	—	+	bil	?	?
Kubota et al. (1981) ¹³⁾	M/39	Japanese	C2-3	—	+	bil	?	+
	F/47	Japanese	C2-3	—	+	bil	?	+
Ota et al. (1982) ²⁰⁾	F/75	Japanese	C5-6	—	—	rt	?	+
Minami et al. (1985) ¹⁴⁾	M/40	Japanese	C7-T1	—	—	bil	d	+
Seichi et al. (1988) ²²⁾	M/70	Japanese	C6-7	+	—	lt	f	—
Tanaka et al. (1988) ²⁷⁾	M/70	Japanese	C6-7	+	+	lt	?	?
Hoshida et al. (1989) ⁶⁾	M/55	Japanese	C2-3	—	+	lt	?	?
Sato et al. (1989) ²¹⁾	F/70	Japanese	C4-5	—	—	bil	f	—
	F/72	Japanese	C4-5, C5-6	—	—	bil	f	—
Shimada et al. (1990) ²³⁾	M/51	Japanese	C2-3	—	+	bil	d	—
Kobayashi et al. (1991) ¹¹⁾	M/61	Japanese	C3-4	—	+	lt	f	—
Nishiura et al. (1992) ¹⁹⁾	F/52	Japanese	C5-6	+	—	rt	f	?
Takayama et al. (1993) ²⁶⁾	M/55	Japanese	C3-4, C4-5	+	—	rt	d	—
Doi et al. (2000) ²⁾	F/72	Japanese	C3-4, C6-7	—	—	bil	d	+
Kruse et al. (2000) ¹²⁾	?/72	American	C3-4	—	—	bil	d	?
Mizuno and Nakagawa (2002) ¹⁵⁾	M/64	Japanese	C6-7	+	+	lt	f	?
Chou et al. (2004) ¹⁾	F/40	Chinese	C2-3	—	—		f	?
Nadkarni et al. (2005) ¹⁶⁾	M/30	Indian	C1-2	—	—	bil	f	—
Fukao et al. (2006) ³⁾	M/72	Japanese	C2-3	+	—	bil	d	+
Present Case 1	F/58	Japanese	C3-4, C4-5	—	—	bil	d	+
Present Case 2	M/63	Japanese	C4-5	—	+	rt	f	—

d: diffuse type, f: focal type, ?: no description.

ic ossified ligamentum flavum was significantly shorter in patients with excellent than in those with fair outcomes.²⁴⁾ Understanding of the clinical symptoms is necessary for early diagnosis and treatment because delays may result in a worse post-treatment functional outcome.²⁴⁾

In our Case 1, the ligamentum flavum was swollen with membrane-like ossification on its underside. In Case 2, the ossified ligamentum flavum continued from the lamina, formed a bone projection to the spinal canal, and caused compression of the dura mater. These observations suggest differences in the etiology and progression of the ossified ligamentum flavum. In Case 2, the ossified ligamentum flavum did not adhere to the dura mater and was easily removed as one mass. In Case 1, removal was complicated by the presence of a fibrous membrane between the swollen ligamentum flavum and the dura mater, and by strong adhesion to the dura mater.

A previous case had a fibrous thin membrane on the underside of the ossified ligamentum flavum,⁷⁾ and a patient with long-term compression had swollen epidural membrane and venous plexus reminiscent of a fibrous band.^{25,26)} A patient presented with myelopathy due to ligamentum flavum thick-

ening and the presence of abnormal fibrous tissue, consisting of slender fibroblasts embedded in a dense collagenous matrix, at the cervicothoracic junction.¹⁸⁾ Excessive development may have been the consequence of continual movement and stimulation. In our Case 1, prolonged strong compression by the swollen ligamentum flavum may have induced the growth of epidural fibrous tissue, and additional chronic dynamic and static stress may have led to the ossification. This patient required surgical intervention despite the possibility of adhesion of the tissue to the dura mater.

Three previously reported cases presented with adhesion to the dura mater and, like our Case 1, the ossified ligamentum flavum was the diffuse type. None of the reported cases with focal ossified ligamentum flavum, like our Case 2, had dura mater adhesion. We stress that patients with diffuse-type ossified ligamentum flavum may present with dura mater adhesion. This knowledge is important for surgical planning.

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