Multiple-level ossification of the ligamentum flavum in the cervical spine combined with calcification of the cervical ligamentum flavum and posterior atlanto-axial membrane

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Abstract

Objective Ossification of the ligamentum flavum (OLF) is rarely identified in cervical spine and its pathogenesis has not been established. We report a case of multiple-level OLF, combined with the calcification of the cervical ligamentum flavum and posterior atlanto-axial membrane.

Clinical presentation A 42-year-old man without any systemic background presented with one month history of pain from the neck to the right shoulder and right leg numbness. Cervical computed tomography demonstrated OLF from C2 to C5, a small area of calcification of the ligamentum flavum (CLF) from C5/6 to C7/T1 and extensive calcification of the posterior atlanto-axial membrane, resulting in spinal canal stenosis. Magnetic resonance imaging showed spinal canal stenosis and severe spinal cord compression from C2 to C5. Thoracic X-ray also showed ossification of the posterior longitudinal ligament (OPLL). We performed laminectomy from C1 to C5 and resected the calcified posterior atlanto-axial membrane and OLF. Histopathological examination demonstrated calcified granules within degenerated fibrous tissue in the posterior atlanto-axial membrane and mature bony trabeculae, bone marrow and residual ligament tissue in the OLF.

Conclusions Simultaneous development of cervical OLF and CLF in this case seems unlikely to have occurred coincidentally and suggests that the pathogenesis of OLF and CLF may share a common initiation factor.

Keywords Cervical spine · Calcification · Ligamentum flavum · Ossification

Introduction

Ossification of the ligamentum flavum (OLF) frequently occurs in the lower thoracic spine [1, 2, 5–7, 11, 12, 14], but rarely in the cervical spine [3, 4, 10, 13]. Cervical OLF has been reported within a few spinal segments [10], but multi-segmental cervical OLF is extremely rare [4]. Concomitant OLF and calcification of the ligamentum flavum (CLF) has not been previously reported. We report a case with multiple-level cervical OLF combined with calcification of the posterior atlanto-axial membrane and ligamentum flavum.

Case report

A 42-year-old man with no relevant past or family history presented with pain from the neck to the right shoulder and numbness of the right leg progressing over the course of a month. Neurological examination revealed normal muscle strength and deep tendon reflexes in all four limbs. Babinski and Hoffman signs were also negative. Plain radiographs of the cervical spine showed OLF from C2 to C5 and an oval nodular lesion between C1 and C2, suggesting calcification of the posterior atlanto-axial membrane (Fig. 1). Cervical computed tomography (CT) revealed contiguous OLF from C2 to C5, a small area of CLF from C5/6 to C7/T1 and extensive calcification of the posterior atlanto-axial membrane, resulting in spinal canal stenosis (Fig. 2). Thoracic X-ray also showed ossification of the posterior longitudinal ligament (OPLL) between T6

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Fig. 1 Lateral radiography of the cervical spine in extension (*right*) and flexion (*left*). Extensive ossification of the ligamentum flavum (OLF) from C2 to C5 (*arrows*) and calcification of the posterior atlanto-axial membrane (*arrow head*). No instability was evident

Fig. 2 Preoperative CT of the cervical spine. **a** Sagittal reconstructed CT clearly showing extensive OLF from C2 to C5 (*black arrows*), small calcification of the ligamentum flavum (CLF) from C5/6 to T1/2 (*white arrows*) and calcification of the posterior atlanto-axial membrane (*arrowhead*). **b** Axial CT at the level of the axis revealing calcified posterior atlanto-axial membrane (*arrow*). **c** Axial CT at the C5 level showing the OLF causing cervical canal stenosis (*arrow*)

Fig. 3 Lateral radiography of the thoracic spine ossification of the posterior longitudinal ligament is apparent between T6 and T8 (arrows)

and T8 (Fig. 3). Cervical magnetic resonance imaging (MRI) showed spinal canal stenosis and severe spinal cord compression from C2 to C5. An intramedullary hyperintense area was observed on T2-weighted MRI (Fig. 4). The patient was not obese and had no comorbidities. Results of blood tests including blood sugar, thyroid, parathyroid, and growth hormone levels were normal.

Although symptoms were not severe, surgical intervention was selected in consultation with the patient because of severe spinal stenosis with the intramedullary hyperintensity on MRI. We performed posterior decompressive surgery from C1 to C5 and resected the calcified posterior atlanto-axial membrane and cervical OLF from C2 to C5. We attempted resection of the OLF through hemilaminectomy. However, because the ossified lesion was very large and the dura had ossified, we performed laminectomy and resected the calcified posterior atlanto-axial membrane and OLF after thinning the ossified dura

using a high-speed drill. We were able to preserve the arachnoid membrane, without any leakage of cerebrospinal fluid.

Histological findings demonstrated calcified granules within degenerated fibrous tissue in the calcified tissue (Fig. 5, right) and mature bony trabeculae, bone marrow and ligament tissue in OLF (Fig. 5, left). Three years postoperatively, the patient is asymptomatic. CT has demonstrated no regrowth of OLF or calcification, and no cervical deformity.

Discussion

Although thoracic OLF is relatively common in Japan, the epidemiology and etiology of cervical OLF remains obscure. Investigation of 1,736 volunteers of southern Chinese origin revealed radiological OLF in 3.8 % of individuals, with small cervical OLF found in only 4 individuals [5]. Symptomatic OLF is frequently located in the lower thoracic spine [1, 2, 5–7, 11, 12, 14] and symptomatic OLF of the cervical spine is extremely rare [3, 4, 10, 13]. On the other hand, CLF characteristically occurs in the cervical spine of older women and rarely involves the thoracic spine [9]. This difference in common locations of OLF and CLF suggests that local factors are related to the formation of OLF and CLF.

Miyazawa et al. [10] reviewed the literature (including Japanese articles), and found 22 detailed reports of cervical OLF. Of these, 18 cases were located at a single intervertebral level and 4 cases involved two intervertebral levels. Only Christiano et al. [4] have reported on a case of multiple-level OLF of the cervical spine (C2–T3), which was seen in a patient with idiopathic hypercalcemia. Concomitant OLF and CLF has not been reported previously.

The relatively strong association between OPLL and cervical OLF suggests a systemic hyperostotic background to cervical OLF [10]. Upregulation of systemic factors including hormones and growth factors associated with ectopic ossification have been thought to be involved in the initiation and development of spinal ligament ossification [8]. However, the true etiology of OLF remains almost completely unknown. The present patient did not show any systemic background.

The etiology of OLF is thought to be different from that of CLF [9]. In CLF, calcified granules from calcium hydroxyapatite are characteristically deposited in the degenerated ligament, with no mature bone forming within the ligament [9]. The present patient showed cervical OLF, thoracic OPLL and cervical CLF. This combination raises the possibility that the same factor may have initiated ossification and calcification in these spinal ligaments.

Fig. 4 Preoperative MRI of the cervical spine. **a** T1-weighted sagittal MRI demonstrating compression of the posterior cervical cord by the OLF. **b** On T2-weighted sagittal MRI, the OLF appears hypointense

(arrows) and the calcified posterior atlanto-axial membrane appears isointense (arrowhead). **c** The cervical spinal cord shows an intramedullary hyperintense area (arrows)

Fig. 5 Photomicrograph of a section from the resected OLF and calcified posterior atlanto-axial membrane (haematoxylin-eosin stain, original magnification $\times 100$). *Left* OLF shows fragmented and focally irregular osseous trabeculae (black arrow) and intraligamentous

ossification (white arrow). *Right* the posterior atlanto-axial membrane shows calcification (white arrow) within degenerated fibrous tissue (black arrow)

In this patient, because the extent of cervical OLF was very large and the dura had ossified, laminoplasty and hemilaminectomy were not possible and we employed laminectomy for resection of the OLF. This patient was rather young. Meticulous follow-up is needed to observe progression of ossification or calcification in other spinal regions.

Conclusions

The patient in this case lacked any systemic background for ectopic ossification, yet displayed multiple-level cervical OLF, thoracic OPLL and cervical CLF. Both cervical OLF and cervical CLF are uncommon diseases. Simultaneous development of cervical OLF and CLF thus seems unlikely

to have occurred coincidentally. We suggest that the pathogenesis of OLF and CLF may share a common initiation factor.

Conflict of interest No benefits in any form have been or will be received from any commercial party related directly or indirectly to the subject of this article.

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