Case report

Thoracic myelopathy caused by calcification of the ligamentum flavum

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Abstract

Calcification of the ligamentum flavum (CLF), which is a rare disorder that can potentially cause myelopathy, occurs uncommonly in the thoracic spine. Here, we report a rare case of thoracic myelopathy caused by CLF in a 78-year-old man. Magnetic resonance imaging (MRI) showed posterior spinal cord compression by a hypo-signal intense mass, and computed tomography (CT) revealed CLF and vacuum disc phenomenon at T10/11. After undergoing posterior decompression and instrumented fusion (T9–T12), the patient's gait difficulties improved. The pathogenesis of CLF is largely unknown; however, it involves accumulation of calcium pyrophosphate dehydrate crystals (CPPD), and CLF from CPPD deposition tends to occur within a thickened and hypertrophic ligament. CLF occurs predominantly in the cervical spine and less frequently in the lumbar spine, with few cases involving the thoraco-lumbar spine. The thoracic spine is characterized by hypomobility; however, the thoraco-lumbar spine has a mobile segment which may potentiate CLF formation. Decompression with fusion surgery can be useful for treating patients with thoraco-lumbar CLF.

Key words: calcification of the ligamentum flavum, thoracic myelopathy, decompression with fusion surgery

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Introduction

Calcification of the ligamentum flavum (CLF) is a rare disorder, primarily affecting middle-aged Asian women, and it can cause myelopathy due to posterior cord compression^{1–3}. Unlike ossification of the ligamentum flavum (OLF), CLF shows no mature bone formation within the ligament. The two most common sites where CLF occurs are the cervical and lumbar spine segments, while the thoracic spine is seldom affected^{4, 5}. Here we describe a rare case of thoracic myelopathy caused by CLF. This case report was conducted in accordance with the Declaration of Helsinki, and written

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informed consent was obtained from the patient for publication of this case report and any accompanying images.

Case report

A 78-year-old man presented with a significant history of hypertension and a history of CABG 5 years prior. His body mass index (BMI) was 27.5 kg/m² (body weight, 75 kg; height, 165 cm). He had had bilateral femoral numbness for one month that had gradually spread to the entire leg, as well as two weeks of progressive gait difficulties. He exhibited incomplete spastic paraplegia and was unable to stand or walk alone. Hyperactive deep tendon reflexes were found for both lower extremities along with bilateral muscle weakness (manual muscle testing [MMT], 3-4/5 level), diminished bilateral pain sensation upon pinprick test below the groin, and a disturbance of urination. Magnetic resonance imaging (MRI) showed posterior spinal cord compression caused by a hypointense mass (T10/11) as well as an intramedullary high-intensity signal change on the T2-weighted image. Computed tomography (CT) revealed CLF, along with a vacuum disc phenomenon at T10/11 and bony fusion in front of the vertebra at T11-12; slight CLF was also seen

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Figure 1 a, b Preoperative MRI of the thoraco-lumbar spine: a. T2-weighted sagittal image. b. T2-weighted axial image (T10/11 level) showing the hypointense mass causing dorsal cord compression and intramedullary high-intensity signal change. c, d, e. Pre-operative CT of the thoraco-lumbar spine: c, d. sagittal CT reconstruction showing calcification of the ligamentum flavum (arrowhead) and the vacuum disc phenomenon at the T10/11 disc. d. axial CT scan (T10/11 level) showing calcification of the ligamentum flavum (arrowhead).



Figure 2 Postoperative radiograph after posterior decompression followed by instrumented fusion from T9–T12. a. anterior-posterior view, b. lateral view.



Figure 3 Intraoperative photograph demonstrating the calcified ligamentum flavum at the T10/11 level (arrows) post-laminectomy.

at L3/4 (Figure 1). The patient underwent posterior decompression followed by instrumented fusion from T9–T12, and pedicle screws were inserted two vertebrae above and below to ensure stability at T10/11 (Figure 2). Adhesion of the calcified ligamentum flavum to the posterior dura mater was observed (Figure 3). The patient's postoperative course was uneventful, and the numbness and muscle weakness of the bilateral lower extremities were ameliorated (MMT, 5/5 level). He gradually recovered from the gait disturbance and could ambulate with the aid of a walker six months after treatment.

Discussion

In this case, posterior decompression with fusion surgery for thoracic CLF improved the patient's lower limb function. The pathogenesis of CLF is largely unknown, however it can be composed of calcium pyrophosphate dehydrate crystals (CPPD)^{4, 6–9)}. CPPD deposition predominantly occurs in the cervical spine of middle-aged to older women, and CLF from CPPD deposition tends to occur within a thickened and hypertrophic ligament, which is commonly caused by the degenerative changes of aging^{1, 4, 5)}. The mechanical load placed on the ligament causes the ligamentum flavum cell to deposit calcium¹⁰⁾.

CLF occurs predominantly in the cervical spine and less commonly in the lumbar spine^{4, 5)}, with few cases involving the thoraco-lumbar spine, which is characterized by hypomobility^{3, 4, 11}; however, the thoraco-lumbar area does have a mobile segment due to the free end of the 11th and 12th ribs. The current patient's CLF was at T10/11, and the intervertebral vacuum phenomenon at T10/11 suggests segmental hypermobility and/or instability, both of which may potentiate CLF formation. Moreover, bony fusion in front of the vertebra at the distal adjacent segment T11/12 might affect local instability at T10/11. Early surgical decompression of the spinal cord is a demonstrated treatment performed for CLF^{2, 5, 9}. In this case, we focused on the local instability at the CLF, and chose posterior decompression with instrumented fusion instead of decompression alone. Postoperatively, the patient was observed to have marked improvement in his lower limb function.

Conclusion

Thoracic CLF, a rare disease which causes spinal cord compression with myelopathy, can rarely occur at only the thoraco-lumbar level, due to the mobile segment present in this region of the spine. Early surgical intervention can provide a satisfactory outcome, and decompression with fusion surgery can be a better option for patients with thoracolumbar CLF.

Conflict of interest: The authors declare that there they have no conflict of interest related to the present study.

References

- Miyasaka K, Kaneda K, Sato S, et al. Myelopathy due to ossification or calcification of the ligamentum flavum: radiologic and histologic evaluations. AJNR Am J Neuroradiol 1983; 4: 629–632. [Medline]
- 2. Ahn DK, Lee S, Moon SH, et al. Ossification of the ligamentum flavum. Asian Spine J 2014; 8: 89–96. [Medline] [CrossRef]
- Muthukumar N, Karuppaswamy U. Tumoral calcium pyrophosphate dihydrate deposition disease of the ligamentum flavum. Neurosurgery 2003; 53: 103–108, discussion 108–109. [Medline] [CrossRef]
- Giulioni M, Zucchelli M, Damiani S. Thoracic myelopathy caused by calcified ligamentum flavum. Joint Bone Spine 2007; 74: 504–505. [Medline] [CrossRef]
- Inoue H, Seichi A, Kimura A, et al. Multiple-level ossification of the ligamentum flavum in the cervical spine combined with calcification of the cervical ligamentum flavum and posterior atlanto-axial membrane. Eur Spine J 2013; 22(Suppl 3): S416–S420. [Medline] [CrossRef]
- Berghausen EJ, Balogh K, Landis WJ, et al. Cervical myelopathy attributable to pseudogout. Case report with radiologic, histologic, and crystallographic observations. Clin Orthop Relat Res 1987; 217–221. [Medline]
- Brown TR, Quinn SF, D'Agostino AN. Deposition of calcium pyrophosphate dihydrate crystals in the ligamentum flavum: evaluation with MR imaging and CT. Radiology 1991; 178: 871–873. [Medline] [CrossRef]
- Seki S, Kawaguchi Y, Ishihara H, et al. Lumbar spinal stenosis due to a large calcified mass in the ligamentum flavum. Asian Spine J 2013; 7: 236–241. [Medline] [CrossRef]
- 9. Roet M, Spoor JKH, de Waal M, et al. Extensive calcification of the ligamentum flavum causing cervical myelopathy in a Caucasian woman. Springerplus 2016; 5: 1927. [Medline] [CrossRef]
- Chao YH, Huang SY, Yang RC, et al. Tissue transglutaminase is involved in mechanical load-induced osteogenic differentiation of human ligamentum flavum cells. Connect Tissue Res 2016; 57: 307–318. [Medline] [CrossRef]
- 11. Shiguematsu FY, de Souza EC, Zimmermann AF, *et al.* Thoracic myelopathy due to calcification of the ligamentum flavum with hyperproteinorachia and responsive to steroid therapy: case report. Rev Bras Reumatol 2012; 52: 438–446. [Medline]