Seiichi Odate et al., J Spine Neurosurg 2012, 1:2
http://dx.doi.org/10.4172/2325-9701.1000104

Journal of Spine & Neurosurgery

Case Report

A SCITECHNOL JOURNAL

Intolerable Low Back Pain Due to Ossification of the Ligamentum Flavum at the L5–S1 Level

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Abstract

A 37-year-old man presented with low back pain and leg pain. A CT scan identified ossification of ligamentum flavum (OLF) at the L5–S1, causing foraminal stenosis. AnL5 nerve root block resolved his low back pain and leg pain completely. However, the symptoms recurred after the effect of the local anesthetic wore off. Because of persistent and intolerable low back pain, he underwent surgery, partial facetectomy and removal of the ossified ligaments followed by posterior instrumented fusion at L5–S1. Histopathology of the specimen revealed no apparent degenerative changes in the ligament. The patient's symptoms had disappeared completely at the final follow-up.

This case is different from previously reported cases: the chief complaint was intolerable low back pain; the patient was a young adult; it did not involve degeneration of the spinal elements; and did not involve coexisting ossification of other spinal ligaments. Although the pathogenesis of OLF remains still unclear, it is possible that OLF of the lower lumbar spine causes severe low back pain and is treatable by surgery.

Keywords: Ossification of ligamentum flavum; Lower lumbar spine; Intolerable low back pain; Radiculopathy; Foraminal stenosis; Hyperthyroidism

Abbreviations: OLF: Ossification of Ligamentum Flavum; MRI: Magnetic Resonance Imaging; CT: Computed Tomography; ODI: Oswestly Disability Index; OPLL: Ossification of the Posterior Longitudinal Ligament

Introduction

Ossification of ligaments in the thoracic and cervical spine has been widely recognized to cause myelopathy [1,2]. However, there are only a few reports of patients with ossification of the ligamentum flavum (OLF) in the lumbar spine presenting with radiculopathy [3-5]. We describe a rare case of a patient presenting with intolerable low back pain due to OLF located at the L5–S1 level.

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Received: November 03, 2012 **Accepted:** December 05, 2012 **Published:** December 03, 2012

Case Report

History and examination

A 37-year-old man visited our hospital complaining of intolerable low back pain and bilateral leg pain. He reported a history of recurrent low back pain since the age of 30 years. The low back pain had gradually deteriorated and he had suffered from severe pain every day for the previous year. The leg pain had occurred for 6 months. Although, the patients complained both low back pain and leg pain, leg pain was relatively slight but low back pain was serious problem for him. He had been treated for hyperthyroidism for the past 7 years, but his family's medical history was unremarkable. He was admitted to our hospital for further examination and treatment. A neurologic examination revealed mild muscle weakness with 4/5 strength in the tibialis anterior and extensor hallucis longus muscles. He had sensory disturbance at the L5 area in both legs. His patellar tendon and Achilles tendon reflexes were normal. Laboratory blood and urine tests showed no abnormal findings. Plain radiographs of the lumbo sacral spine and flexion and extension radiographs showed no degenerative changes (Figures 1A and 1B). Sagittal T2-weighted magnetic resonance imaging (MRI) demonstrated no degenerative changes in the lumbar spine (Figure 2A). Axial T2-weighted MRI revealed no obvious compression of the neural elements (Figure 2B). Computed tomography (CT) revealed high-density lesions, suggesting the presence of OLF in the capsular portion at the L5–S1 level without degenerative changes in the facet joints (Figure 3A). Foraminal stenosis due to the OLF was suspected at the L5–S1 level (Figure 3B). No ossification of any other spinal ligaments was observed on CT. Immunologically, he was not human leukocyte antigen type B-27 and did not show any radiographic changes in the iliosacral joint such as erosion or sclerosis, so the possibility of ankylosing spondylitis was excluded at that time.

Initial treatment

AnL5 nerve root block (0.5 mL of 1% Lidocaine) resolved his low back pain and leg pain completely. However, the symptoms recurred after the effect of the local anesthetic wore off. Oswestly Disability

(A) (B)

Figure 1: Lateral radiographs of the spine in flexion (A) and extension (B) showed no abnormality.

Index (ODI) was 35 of 50 points. Because of persistent and intolerable low back pain, he underwent surgery.

Operation

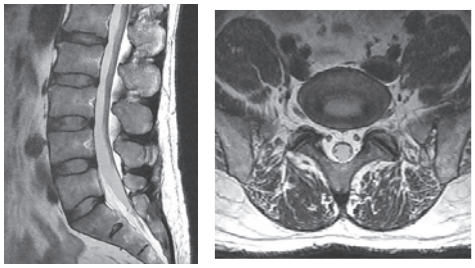
Decompressive partial facetectomy of L5–S1 and removal of the OLF were performed. There was iatrogenic destabilization after the decompression, then posterior instrumented fusion was performed (Figure 4). Both L5 nerve roots were severely compressed by the OLF in the intervertebral foramen. The patient's severe low back pain and leg pain disappeared completely after surgery and ODI was 3 points at the follow-up examination 2 year later.

Histopathology

Tissue sections were prepared from the removed OLF and stained with hematoxylin and eosin. Histopathology of the surgical specimen revealed ossification resulting in trabecular bone formation in the ligamentum flavum (Figure 5A). Notably, there were no findings of an inflammatory reaction or degenerative changes in the elastic fibers (Figure 5B).

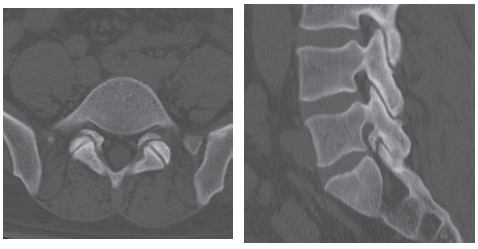
Discussion

The majority of cases of OLF occur at the lower third of the thoracic or the thoracolumbar spine [1,2]. Only a few authors have published cases that involved patients with OLF surgically treated in the lumbar spine [3-5]. Kurihara et al. [6] reviewed lumbosacral roentgenograms of 2403 outpatients and showed that OLF of the lumbar spine was found in 206 (8.6%) patients and that the most commonly involved levels were the upper and middle lumbar spine. They also showed that patients with OLF in the lumbar spine demonstrated a high incidence of ossification of the other spinal ligaments, such as ossification of the posterior longitudinal ligament (OPLL) throughout the spine. However, in the present case, the OLF was localized only in the lower lumbar spine, at level L5–S1 and no ossification of other spinal ligaments was observed. Yano et al. [5] reported a case of a 27-year-old woman with OLF in the lower lumbar spine presenting with radiculopathy. The OLF existed at L4–L5 and L5–S1 levels in the spinal canal portion involving degeneration of the ligamentum flavum. Interestingly, in the present case, the ossification existed more laterally, in the capsular portion of the ligamentum flavum and did not involve degeneration of the ligamentum flavum. This capsular outgrowth resulted in foraminal stenosis and L5 radiculopathy.



(A) (B)

Figure 2: T2-weighted MRI demonstrated no degenerative change or stenosis; Sagittal (A) and axial (B) views at the L5–S1 level.



(A) (B)

Figure 3: (**A**) Axial CT image demonstrating ossification of the ligamentum flavum at the L5 –S1 level. (**B**) A Parasagittal CT image revealed lesions causing foraminal stenosis at the L5–S1 level.

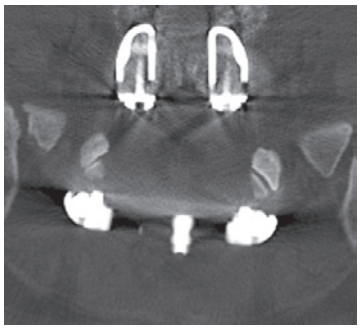
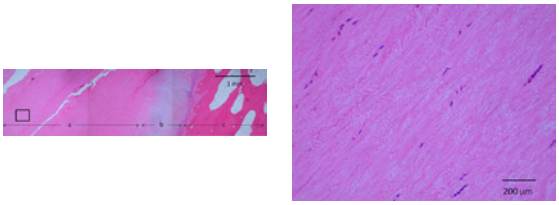


Figure 4: Postoperative axial CT image demonstrating complete resection of the ossified lesion with additional instrumented fusion.



(A) (B)

Figure 5: (**A**) Photomicrograph of the specimen of OLF obtained at the L5–S1 level during surgery, showing different steps of the ossification process (hematoxylin and eosin staining). The ligament (**a**) is transforming into cartilaginous tissue (**b**) and subsequently into trabecular bone (**c**). (**B**) Magnified view of the square field in Fig. 5A. Elastic fibers are regularly arranged in the ligament. There is no evidence of degeneration in the elastic fibers (hematoxylin and eosin staining).

Although the pathogenesis of OLF remains almost unknown, several investigators have described the possible contribution of mechanical, [7] metabolic, [8] genetic [9] and cell biological [10,11] factors. A polygenetic autosomal dominant mode of inheritance has been offered for OPLL, [9] whereas OLF and spondylosis correlate better with diffuse mechanical stress and degenerative changes [5,12]. Repeated mechanical stress on the ligamentum flavum associated with intervertebral disc and facet joint degeneration has been reported to promote hypertrophy and ossification of the ligament [12]. However, in the present case, the patient reported no trauma to his lumbar spine and there were no degenerative changes to the discs or facet joints. Generally, photomicrographs of the ligamentum flavum from patients with OLF show degenerative change in elastic fibers.

Interestingly, there were no such findings in our case. Therefore, in the present case, mechanical stress might not have contributed to the development of OLF.

Although OLF is reported to involve patients predominantly in their 60s and 70s, [1,13] our patient was only 37 years old. It is known that the incidence of OLF is higher in patients with diffuse idiopathic skeletal hyperostosis and with ankylosing spondylitis [14,15]. However, in the present case, there were no findings of skeletal hyperostosis or ankylosing spondylitis. A detailed family medical history obtained from his parents revealed that the patient had no blood relatives within a second degree of relationship who had any medical history of treatment for ossification of spinal ligaments. Therefore, in the present case, the influence of genetic factors seems unlikely.

Patients with ossification of the spinal ligament are reported to have a higher frequency of diabetes mellitus, obesity, hyperinsulism, hemochromatosis and abnormalities in calcium metabolism [2,14]. Some systemic hormones, such as calcium regulating hormones, insulin, leptin and local growth factors, such as transforming growth factor-beta and bone morphogenetic protein, have been studied and are thought to be involved in the initiation and development of ossification of the spinal ligament [11,16]. In the present case, the patient did not have any previous generalized disorders except for hyperthyroidism. Thyroid hormones are essential for normal skeletal growth and the maintenance of bone mass in adult hood, although their mechanism of action in the bone is poorly understood [17]. Excessive amounts of thyroid hormone induce increased activity of osteoblasts [18]. Zhong et al. [19] investigated the phenotypic characterization of ligamentum flavum cells from patients with OLF. They suggested that OLF cells have phenotypic characters of chondrocytes. These metaplastic cells in the ligament eventually transform into osteoblasts, which contribute to the early development of OLF. They hypothesized that specific osteogenetic cytokines and certain metabolic disorders might induce such cellular proliferation. Although there is no report that describes a relationship between hyperthyroidism and ossification of spinal ligaments, it is possible that superabundant thyroid hormone in this patient induced osteoblastic differentiation of the ligamentum flavum cells followed by heterotopic bone formation at the ligamentum flavum. The reason why bone formation was located at the L5-S1 level is still not clear.

Decompressive laminectomy and removal of the OLF is the most commonly performed surgical procedure [3,6]. Because the ossification existed laterally in the capsular portion of the ligamentum flavum and preoperative L5 nerve root blockage was effective for pain relief, we resected the L5–S1 facet joints to remove the OLF completely. There was iatrogenic destabilization after the sufficient decompression. To avoid remain of the low back pain and recurrence of the ossification due to postoperative instability, instrumented fusion was performed. The patient's intolerable low back pain and leg pain disappeared completely after surgery.

As a limitation, we need additional rationale for the pathology investigation. It would be beneficial to provide additional data for pathology screening. Since tetracycline is absorbed into bone, it has been widely used in bone histomorphometry to label new bone formation. Unfortunately, we didn't use tetracycline labeling in this case. As a future investigation, if the patients recurred and needing operation to another segment, we will consider applying tetracycline labelling before operation to estimate the ossification process.

Conclusion

This case is different from previously reported cases. Differences were: the chief complaint was intolerable low back pain; the patient was a young adult; the ossification was located at the L5–S1 level and existed laterally in the capsular portion; it did not involve degeneration of the ligamentum flavum and facet joint; and did not involve coexisting ossification of other spinal ligaments. Kurihara et al. [6] reported an 8.6% incidence of OLF in the upper and middle lumbar spine. Although long-term follow-up is needed to confirm this assumption, it is possible that OLF of the lower lumbar spine causes severe low back pain and is treatable by surgery.

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