The Journal of Turkish Spinal Surgery
2008; 19 (4): 447-452

CASE REPORT / *OLGU SUNUMU*

OSSIFICATION OF LIGAMENTUM FLAVUM CAUSING SPINAL STENOSIS: A CASE REPORT

LİGAMENTUM FLAVUM OSSİFİKASYONUNA BAĞLI SPİNAL STENOZ : OLGU SUNUMU

Ulaş YENER* , Serdar ÖZGEN, Arzu GERÇEK***, Deniz KONYA******

SUMMARY:

Ossification of the ligamentum flavum is a rare entity observed exclusively in Japanese people. We report a new case in a 46-year old man with ossification of the ligamentum flavum that was located at the thoracic (Th) 2 vertebra level with cervical disc herniation at the cervical (C) level 5-6. The patient showed myelopathy symptoms due to the compression on the spinal cord. A decompressive Th 2 laminectomy was performed. The cause of the compression was found to be an ossified ligamentum flavum, which was confirmed by histopathological examination. The postoperative course was uneventful and signs, symptoms, and functional status markedly improved after decompressive surgery.

Key words: *Ligamentum flavum ossification, spinal stenosis, surgical treatment*

Level of Evidence: *Level IV*

ÖZET:

Ligamentum flavum ossifikasyonu çoğunlukla Japonlarda görülen nadir bir antitedir. Bu çalışmada, 46 yaşında erkek hastada torakal (Th) 2. vertebra seviyesinde bulunan ligamentum flavum ossifikasyonu ve buna eşlik eden servikal 5-6 disk hernisi bulunan bir olgu sunulmaktadır. Spinal kord basısına bağlı myelopatik semptomlarla başvuran hastaya Th 2 laminektomi uygulanmıştır. Ligamentum flavum ossifikasyonunun spinal kord basısına neden olduğu intraoperatif görülüp; yapılan örnekleme ile de tanı histopatolojik olarak desteklenmiştir. Sorunsuz bir ameliyat geçiren hastanın şikayet ve bulgularının dekompresif laminektomi sonrası belirgin şekilde gerilediği görülmüştür.

Anahtar Kelimeler: *Ligamentum flavum ossifikasyonu, spinal stenoz, cerrahi tedavi*

Kanıt Düzeyi: *Düzey IV*

(*) *Resident of Neurosurgery, Department of Neurosurgery, Marmara University Faculty of Medicine, Istanbul, Turkey*
(**) *Associate Professor of Neurosurgery, Department of Neurosurgery, Marmara University Faculty of Medicine, Istanbul, Turkey*
(***) *Instructor of Anesthesiology, Clinic of Anesthesiology, Institute of Neurological Science, Marmara University, Istanbul, Turkey*
(****) *Assistant Professor of Neurosurgery, Department of Neurosurgery, Marmara University Faculty of Medicine, Istanbul, Turkey*

INTRODUCTION:

Ossification of the ligamentum flavum (OLF) and posterior longitudinal ligament (OPLL) is a well recognized cause of spinal canal stenosis resulting in myelopathy or radiculopathy [6, 12]. Ossified ligamentum flavum was first described by Poglar [14] in 1920 and the first case causing myelopathy was reported by Yamaguchi [24]. This condition was reported in Asia, mostly in Japanese people, and therefore it has been termed as "Japanese disease". A few cases of OLF have been reported in whites [7, 12]. Although OLF and OPPL accompaniment is common and found mostly in Asian people, our report presents a case of spinal cord compression by isolated OLF at the upper thoracic level with cervical disc herniation at the same time, which has rarely been described in whites. [8]

CASE REPORT:

A 46-year-old Caucasian man was admitted to the hospital with a 3 months history of numbness, initially developing in the lower limbs and then involving the thoracic T-4 dermatome level. The arms were asymptomatic, and he had no back pain. There were no sphincter disturbances. The patient was a former worker in marble industry. There was no family history of metabolic or neurologic diseases. The sensory examination revealed hypoesthesia below the T-4 dermatome level. Pinprick test and temperature sensation were normal. The patient's gait was wide based and unsteady, requiring bilateral help. Plain radiographs of the cervical spine revealed osteophytic degenerations, but thoracic spine showed no abnormality. Magnetic resonance imaging (MRI) of the cervicothoracic spine showed cervical disc herniation at the Cervical (C) level 5-6 and spinal canal stenosis from T-2 down to T-12, with spinal cord compression on T-2 caused by OLF (Figure-1). Axial MRI and computed tomography (CT) scan demonstrated the presence of a spinal stenosis caused by a bone apposed intraspinally at the level of the lamina, most severe at T-2 (Figure-2). Hematologic and biochemical investigations were within normal range.

Figure-1. (a) T2-weighted sagittal cervical MRI shows C5-C6 disc herniation, (b) T2-weighted sagittal thoracic MRI shows ossified ligamentum flavum at the T-4 vertebra level.

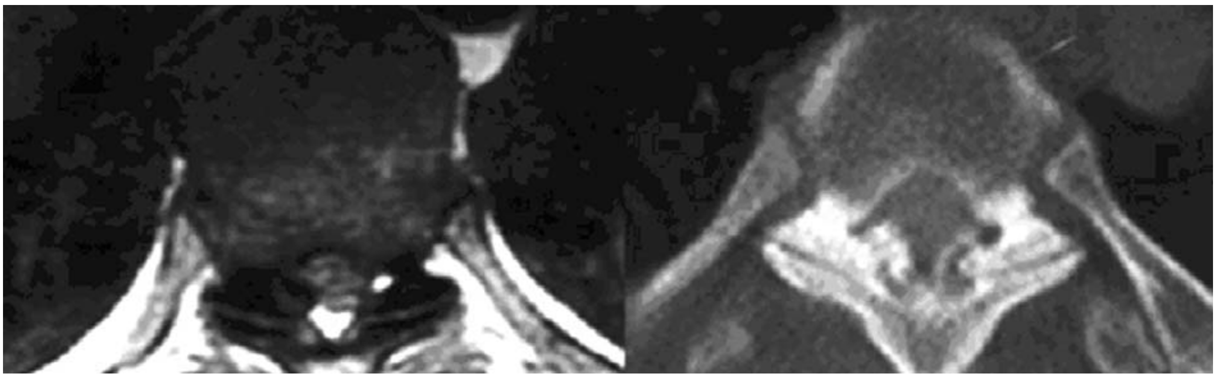

Figure-1.(a) T2-weighted axial thoracic MRI shows narrowing in the spinal due to ossified ligamentum flavum, (b) axial sectioned thoracic CT shows ossified ligamentum flavum.

Decompressive surgery was scheduled. Cervical C5-C6 discectomy was performed and fusion was carried out using allograft material (Tutogen Medical GmbH, Neunkirchen am Brand, Germany), an anterior cervical plate, and screws (DePuy Spine Inc, Raynham, MA, USA). Then, in the same session, decompressive laminectomy was performed at the T-2 level with the patient in prone position. The tissue was carefully excised by using high-speed pneumatic diamond fraise starting from rostral part of the T-2 lamina, which caused spinal cord compression with its exophitic part towards the spinal canal. There were no adhesions of this exostosis to the dura mater. Histopathological examination of the specimen confirmed the presence of ossification with lameller bone formation, cartilage, and fibrous tissue (Figure-3-4).

Figure-3. Tissue samples serially sectioned documented isolated nodules of micro calcifications. (Hematoxylin-eosin, x40 original magnification).

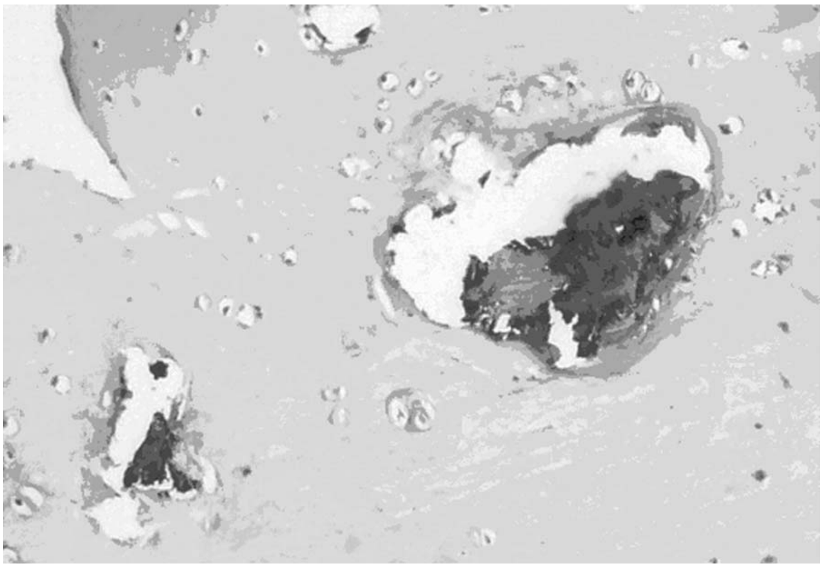

Figure-4. High magnification revealed only tiny mineralization foci formed by calcium salts unassociated with osteoid or bony tissue. (Hematoxylin-eosin, x400 original magnification).

DISCUSSION:

OLF is frequently but not always found in association with OPLL. Furthermore, it can be found in association with diffuse idiopathic skeletal hyperostosis, and metabolic diseases such as diabetes mellitus, Paget's disease, X linked hypophosphatemia, hypoparathyroidism, calcium metabolism abnormalities, ankylosing spondylitis, and Forestier disease [16, 17, 22]. A recent study has also confirmed that induction of bone morphogenic protein-2 causes the differentiation of spinal ligament fibroblast into chondrocytes [5].

In Caucasian individuals, myelopathy caused by isolated OLF is a rare condition. Although its true incidence has not been determined, this disorder is common in Japan and North Africa, and the osseous proliferation occurs in 20 % of Japanese individuals. In the literature, OLF predominantly affects males. Spine involvement is most commonly in the upper thoracic in contrast to OPLL, which generally involves the middle and lower thoracic spine [3, 11, 15]. It must be kept in mind that OLF can cause spinal cord compression in the lumbar spine and rarely in the cervical spine. OLF in the cervical region is most commonly seen in women in contrast to men. The ossification is in most cases multileveled [12, 18]. Age predominance is not well known but most patients are reported to be more than 50 years of age [17].

The most common clinical picture presents with progressive myelopathy, resulting in spastic paraparesis, with or without sphincter dysfunction. Symptoms vary depending on the level of the spinal cord and degree of the cord compression caused by OLF. In addition, radiculopathy may be seen if the lesion is located toward the facet. Walking impairment is generally reported to be the main complaint. OLF can also lead to loss of balance [12, 25].

Plain films are not useful for the diagnosis of OLF. On cervical radiographic imaging, OLF shows radio dense "hook" images located in the posterior part of the foramina [10] and the position does not change by the extension of the neck. CT is the imaging modality of choice. It appears as a radio dense line highlighting the laminae bilaterally in most cases and creates a characteristic V shape with anterior concavity in the epidural space [13, 20]. OLF is found at the superior and inferior extremities of the 2 adjacent laminae, to which they are joined [9-11, 23] MRI is the modality of choice to demonstrate the neural elements and the cord. OLF appears as a hypointense signal on T1 and T2-weighted sequences. Moreover, MRI has another advantage of documenting the degree of cord compression. When gadolinium is used, cord edema is also seen [19]. Hence, for precise diagnosis of OLF, a combination of MRI and CT must be used [2, 12]. Consequently, the ideal radiologic work-up of OLF should combine sagittal CT reconstruction and MRI. The only differential diagnosis of the OLF is the calcification of the ligamentum flavum and sagittal CT reconstruction is useful in distinguishing two similar pathologies.

When neurologic symptoms develop, decompression of the spinal cord is achieved using laminectomy, which must be performed with great care because of dural adhesions to the bone. En bloc dissection of the lamina using a high-speed drill is advised.

Laminectomy is followed by resection of the ligamentum flavum. In the literature, numerous cases of dural injury have been reported [1, 8, 20, 22]. A sharp dissector must be used to separate OLF from the dura. A neurological deficit usually persists even after successful decompression, but when diagnosed early, the outcome of surgery is generally good, providing satisfactory neurologic recovery. For patients with both OPLL and OLF, complet circumferential spinal decompression is recommended [21]

Histologically; hypertrophy, calcification, ossification may be seen in the spectrum of the ligamentum flavum pathologies, and endochondral ossification, which leads to lamellar bone formation, is the pathogenesis for the development of ossification in the ligament [4]. The course generally appears to be slow. Calcification and ossification must be differentiated by the pathologist. Calcified ligamentum flavum reveals calcified granules within the ligamentous fibers and no mature bone as in ossified LF [6, 12] Adequate histological examination is, therefore, essential for accurate diagnosis.

Although OLF is a rare pathology, in our case, after the diagnosis was established, complete recovery was achieved through surgical treatment. The disease is described almost exclusively in Japanese patients, and surgery is the only treatment that can adequately address compression of neurologic structures. Nowadays, BMP-2 protein which was found in the genetic research for the noninvasive treatment of OLF, is in daily use to catalyze the osteofusion process[8]

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