



Ossification of the Ligamentum Flavum in Cervical and Thoracic Spine. Report of Three Cases

George FOTAKOPOULOS, George A ALEXIOU, Evangelos Mihos, Spyridon VOULGARIS
Department of Neurosurgery, University Hospital of Ioannina, Greece

Abstract

Ossification of ligamentum flavum (LF) is a pathological condition that causes neurological symptoms and usually occurs in the thoracic spine and less frequently in the cervical spine. Symptomatology usually involves occipital headache in the early stage, neck pain and later sensory disturbances of upper and lower extremities, gait disturbance, restricted cervical movement and tetraparesis. Herewith, we report on 3 cases of ossification of the LF in the cervical and thoracic spine that had a favourable outcome after surgical treatment.

Key words: ligamentum flavum; ossification; cervical spine; thoracic ossification.

Introduction

Ossification of the ligamentum flavum (OLF) is a pathological condition that causes neurological symptoms (radiculopathy and/or myelopathy) and usually occurs in the thoracic and less frequently in the cervical spine. Mechanical stress, growth factors and trauma, are the most common reason for the development of ossified ligamentum (Li *et al.* 2006, Wang *et al.* 2007). This pathological condition is usually encountered in East Asian populations and exceptionally reported in Caucasian people (Inamasu *et al.* 2006). Herewith, we report on three cases of OLF in Caucasian people that were treated in our institute.

Case report

Case 1

A 84-year-old female presented in our institute complaining of gait imbalance and numbness of the left hand and foot that gradually worsened over a period of ten months. On admission her gait was

spastic and hypaesthesia at the dermatome C6 in both hands was revealed. The reflexes were hyperactive in the lower extremities. Deep tendon reflexes were increased at and below the bilateral biceps with myoclonus bilaterally. There was no urinary or bowel dysfunction. Plain x-rays of the cervical spine failed to demonstrate any specific abnormality. Magnetic resonance imaging (MRI) (Fig. 1A, B) of cervical spine that ensued, showed posterior compression of the dural tube, in the area corresponding to the ligamentum flavum, with cord compression at C4-6 levels. These hypointense lesions had similar signal intensity with the bone marrow in the T1- and T2-weighted images. The patient was operated and the lamina of the C4 and C5 vertebra and the nodular masses were removed en bloc. Besides the depression on the undersurface, there was no bony pedicle arising from the lamina. These masses were connected with the tissue of the ligament to a globular epidural masses beneath the C4 and C5 lamina, with no fixation of the bony masses, causing compression of the cervical cord and myelopathy. Histopathology revealed osteochondral metaplasia and the presence of elastic fibers. Postoperative the spasticity in the

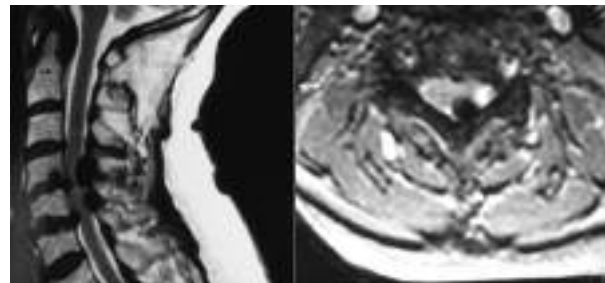


Fig. 1. — A preoperative sagittal (A) and axial (B) T2-weighted images of the cervical spine show the cervical stenosis and the ossification of the ligamentum flavum at C4-C5 and C5-C6 levels. This ossification is seen as a hypointensity in the dorsal aspect of the spinal canal caused myelopathy.

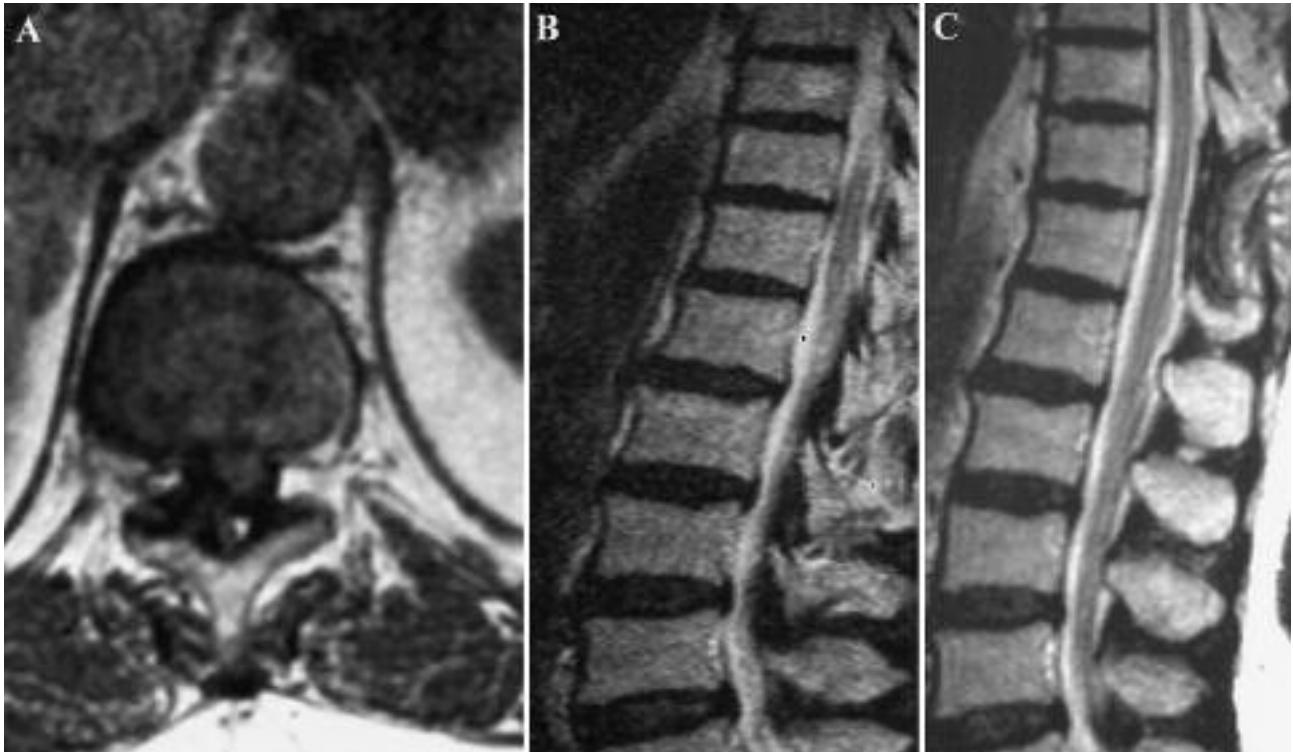


Fig. 2. — A preoperative axial (A) and sagittal (B) T2-weighted MR image showing a hypointense signal at T11-T12 level. C. Post-operative sagittal T2-weighted MR image showing the decompression at the T11-T12 level.

lower limbs improved and the patient managed to walk without major difficulty. At the 1-year follow-up, there was total recovery of neurological functions.

Case 2

A 72-year-old woman complained of pain at the cervical area and numbness in her left hand gradually worsened over the last six months. On neurological examination there was hypaesthesia at the dermatome C5 and an increase at the deep tendon reflexes below the level of the triceps without myoclonus or Babinski reflexes. MRI that ensued showed enlargement of the ligamentum flavum at the C4-5 level and stenosis in the cervical canal. T1- and T2- weighted MR imaging disclosed a hypointense mass posteriorly and dorsal in the anatomical place of the ligamentum flavum, and signs of cervical spinal cord compression. The patient underwent a posterior decompression via a laminectomy of the C4-C5 level. The ossified ligament was removed carefully en bloc under microscopic visualization. Histological examination revealed the presence of a fibrochondral tissue with degenerative elements and without signs of malignancy. One year postoperative

there was gradually improvement of neurological symptoms.

Case 3

A 66-year-old woman complained of gait imbalance that gradually worsened over a period of 2 months. On neurological examination the deep tendon reflexes were increased at and below the T12 level, with bilaterally positive myoclonous and Babinski, without urinary or bowel dysfunction. CT revealed narrowed spinal canal resulting from a dorsolateral V-shaped hyperdensity. MRI that ensued showed (Fig. 2A, B) hypointense signal at the anatomical place of the ligamentum flavum, with cord compression at T11-T12 level. The patient underwent posterior decompression and laminectomy (Fig. 2C). The ossified ligament was slightly connected by dura and removed en bloc. Postoperatively, the patient's neurological status started to improve gradually.

Discussion

OLF is a pathological condition that causes neurological symptoms and usually occurs in the

thoracic and less frequently in the cervical spine. It is more common in East Asian countries and usually affects adults 40 to 60 years of age (Inamasu *et al.* 2006). The mechanism of hypertrophy and progression of ossification is confined to ligament flavum (LF) only and does not extend to the neighbouring spinal bony arch (Kyongsong *et al.* 2008). The significant role of hypertrophy and ossification of the LF, is not clearly understood, but has been found an association with ossification of the posterior longitudinal ligament (OPLL), or diffuse idiopathic skeletal hyperostosis (Wang *et al.* 2007). Furthermore, bone morphogenetic proteins (BMPs), vascular endothelial growth factor (VEGF) (Aizawa *et al.* 2006), transforming growth factor- (TGF) (Wang *et al.* 2007) and especially the cartilage derived morphogenetic protein-1, a key molecule regulating cartilage formation and member of the TGF superfamily have been related to the development and growth of ossification of the ligament (Wang *et al.* 2007).

Most cases of OLF (Inamasu *et al.* 2006) occur in the thoracic spine or the thoracolumbar spine and rarely in the cervical spine (Kruse *et al.* 2000). Thoracic OLF is most frequently noted in patients with diffuse idiopathic skeletal hyperostosis, fluorosis, diabetes mellitus and ankylosing spondylitis (Li *et al.* 2006) and occur in only 2% of all spines surgeries (Aizawa *et al.* 2006). The number of OLF-affected segments varies. Forty to 60% has single-segment interlaminar disease and in 10 to 25% the OLF is multisegment. 30 to 50% of OLF patients coaxed with ossification of the posterior longitudinal ligament (OPLL) (Inamasu *et al.* 2006, Li *et al.* 2006). Asymptomatic OLF may be a relatively common condition in the elderly population (6.2% in male and 4.8% in female patients) (Inamasu *et al.* 2006) and occur in 38,5% in thoracic areas, 26,5% in lumbar and only 0,9% in cervical regions (Kyongsong *et al.* 2008). According to the distribution pattern of ossification in MRI three types of OLF have been described, namely isolated, continuous and non-contiguous. Another classification by CT-scan is lateral, diffuse and thickened nodular 55% (Li *et al.* 2006).

Symptomatology usually involves occipital headaches in the early stage, neck pain and later sensory disturbance of upper and lower extremities, gait disturbance, restricted cervical movement and tetraparesis (Tatsuhito *et al.* 2000). OLF in the thoracic area is associated with back pain and numbness (Aizawa *et al.* 2006). On radiological investigation CT shows a bean-like high density mass inside the LF, with no continuation to the neighbouring lamina. On MRI there is usually a dorsolateral low signal mass appearance on both T1 and T2-weighted

images (Tatsuhito *et al.* 2000, Kruse *et al.* 2000). Posterior decompression with laminoplasty or an bloc laminectomy combined with lateral fusion is the mainstay treatment (Li *et al.* 2006). For thoracic OLF fenestration or en bloc laminectomy are usually performed (Aizawa *et al.* 2006). The ossified ligament should be removed carefully because there might be an ossification of the dura matter. In our series we removed en bloc the ossified ligament despite strong adhesion to the dura matter and without dural tear.

The overall prognosis may not be optimal because of the long-standing compression of the ossified LF at the spinal cord (Inamasu *et al.* 2006). Preoperative symptom's duration has been reported to significant related with patients outcome (Inamasu *et al.* 2006). The surgical outcome depended on the severity of myelopathy, thus the earlier it is operated the better the outcome (Aizawa *et al.* 2006, 7).

Conclusions

To conclude, ossification of the ligamentum flavum has been widely recognized as a primary cause of thoracic myelopathy, even though the pathogenesis is still poorly understood. Furthermore, although OLF in the cervical spine segment is rare, the interesting finding in our series was that we encountered two patients with OLF in the cervical spine. Further research and studies of OLF cases are necessary for early and correct diagnosis and therapy, in order to avoid a poor clinical outcome.

REFERENCES

- Aizawa T, Sato T, Sasaki H, Kusakabe T, Morozumi N, Kokubun S. Thoracic myelopathy caused by ossification of the ligamentum flavum: clinical features and surgical results in the Japanese population. *J Neurosurg Spine.* 2006;5:514-9.
- Inamasu J, Bernard G. A review of factors predictive of surgical outcome for ossification of the ligamentum flavum of the thoracic spine. *J Neurosurg Spine.* 2006;5:133-9.
- Kruse JJ, Awasthi D, Harris M, Waguespack A. Ossification of the ligamentum flavum as a cause of myelopathy in North America: report of three cases. *J Spinal Disord.* 2000;13:22-25.
- Kyongsong K., Toyohiko I. Cervical ligamentum flavum ossification - Two case report. *Neurol Med Chir.* 2008;48:183-7.
- Li F, Chen Q, Xu K. Surgical treatment of 40 patients with thoracic ossification of the ligamentum flavum. *J Neurosurg Spine.* 2006;4:191-197.
- Naffziger HC, Inman V, Saunders JB. Lesions of the intervertebral disc and ligamenta flava: clinical and

- anatomical studies. *Surg Gynecol Obstet.* 1988;66: 288-99.
- Otani K, Aihara T, Tanaka A, Shibasaki K. Ossification of the ligamentum flavum of the thoracic spine. *Int Orthop.* 1986;10:135-9.
- Takafumi Y, Kenzo U. Thoracic ossification of the human ligamentum flavum: histopathological and immunohistochemical findings around the ossified lesion. *J Neurosurg Spine.* 2007;7:184-93.
- Tatsuhito Y, Nobuyuki K, Hiromi N. Calcification of the ligamentum flavum-case report. *Neurol Med Chir.* 2000;40:234-8.
- Wang W, Kong L. Ossification of ligamentum. *J Neurosurg Spine.* 2007;6:96.
- Yamazaki A, Homma T, Uchiyama S, Takahashi H. Ossification of the ligamentum flavum and disc degeneration: spinal disorders in growth and aging. Springer, Tokyo; 1995: pp 249-53.

G. Alexiou
PO BOX 103, Neohoropoulos
Ioannina, 45500
Greece
Email: Alexiougrg@yahoo.gr
alexiougr@gmail.com