



Thoracic ossification of ligamentum flavum without myelopathy: a case report of dorsal pain

Miyelopatinin eşlik etmediği torakal ligamentum flavum ossifikasyonu: Dorsal ağrılı olgu sunumu

Sinan Bahadır,¹ Ahmet Yılmaz²

¹Department of Neurosurgery, Bafra State Hospital, Samsun, Turkey

²Department of Physical Medicine and Rehabilitation, Bafra State Hospital, Samsun, Turkey

Received / *Geliş tarihi:* January 2014 Accepted / *Kabul tarihi:* August 2014

ABSTRACT

Spinal stenosis of the thorax is a less recognized and less common condition than other regions. A variety of disorders such as facet hypertrophy, anterior disc protrusion, spondylotic spurs, ossified posterior longitudinal ligament, ligamentum flavum hypertrophy or ossification may lead to stenosis. Lower thoracic segments (T₉₋₁₂) are most frequently involved. Ossification of ligamentum flavum, which is a major cause of thoracic stenosis in Japanese population, is uncommon in Caucasians. Numbness of the leg, followed by paraparesis, and leg pain are the most common initial symptoms, while a minority of patients have trunkal pain as an initial symptom. In this report, we present an unusual case of thoracic stenosis in a Caucasian patient with trunkal pain as an initial symptom and ossification of ligamentum flavum in the upper thoracic region.

Keywords: Dorsal pain; ligamentum flavum; ossification; spinal stenosis.

ÖZ

Spinal stenoz, diğer bölgelere kıyasla toraksta daha az bilinen ve daha az görülen bir hastalıktır. Faset hipertrofisi, anterior disk protrüzyonu, spondilolitik spurlar, ossifiye posterior longitudinal ligament, ligamentum flavum hipertrofisi veya ossifikasyonu gibi çeşitli hastalıklar stenoza neden olabilir. En sık alt torasik segmentler (T₉₋₁₂) etkilenmektedir. Japon toplumunda torasik stenozun başlıca nedenlerinden biri olan ligamentum flavum ossifikasyonu, beyaz ırkta nadirdir. Paraparezinin izlediği bacaklarda hissizlik ve bacak ağrısı başlangıçta en sık karşılaşılan semptomlar iken, hastaların küçük bir grubunda başlangıç semptomu trunkal ağrıdır. Bu yazıda, beyaz ırka mensup bir hastada başlangıç semptomu trunkal ağrı olan ve üst torasik bölgede ligamentum flavum ossifikasyonu olan olağan dışı bir olgu sunuldu.

Anahtar sözcükler: Dorsal ağrı; ligamentum flavum; ossifikasyon; spinal stenoz.

Spinal stenosis is a well-known disorder both in cervical and lumbar region. However, thoracic spinal stenosis, less common than other regions, is less recognized. Thoracic stenosis may result from facet hypertrophy, anterior disc protrusion, spondylotic spurs, ossified posterior longitudinal ligament, ligamentum flavum hypertrophy or ossification.^[1,2] Symptoms may vary according to the patient's age, cause of stenosis, and level of stenosis.^[3] Most frequently, lower thoracic segments (T₉₋₁₂) are involved, as they are the regions where flexion, extension, and rotation

occurs most.^[4,5] The ossification of ligamentum flavum (OLF), though a major cause of thoracic stenosis in Japanese population, is uncommon in Caucasian people.^[3] The most common initial symptoms are numbness of the leg, followed by paraparesis and leg pain. Only a minority of patients has trunkal pain as an initial symptom.^[6]

In this report, we present an unusual case of thoracic stenosis in a Caucasian patient with trunkal pain as an initial symptom and OLF in upper thoracic region.

CASE REPORT

A 55-year-old female patient was admitted to our hospital with a dull and non-spreading back pain between both scapulae for eight years with a significant worsening for the past two years. She did not experience numbness, tingling or limb weakness. Her complaints aggravated by flexion and extension and also standing. During the course of the complaints, she was admitted to various outpatient clinics at different times and was investigated for cervical, lumbar, and shoulder pathologies. However, no significant pathology was able to be found and, then, she was given various medical therapies and engaged in physical therapies more than once, although we were unable to determine the details of these physical therapies. Meanwhile, her complaints persisted. No traumatic injury was present in her medical history. She had diabetes mellitus treated by dietary modification alone. A written informed consent was obtained from the patient.

Physical examination showed no major postural anomaly except for a small degree of kyphosis. Although she did not report myofascial pain symptoms, she was examined for similar pathologies and no muscle tensions or trigger points were detected. Neurological examination findings were unremarkable. Both upper and lower limbs had normal muscle strength and no sensory deficit was detected. Deep tendon reflexes were symmetrical on both sides, despite reduced in lower extremities. Since location of the pain suggested either cervical or thoracic etiology and neurological examination findings were normal, plain radiographs of cervical and thoracic spine was taken initially. Vertebral alignment and intervertebral space distances

were normal and no sign of an osteoporotic vertebra fracture was present. There was a minor kyphosis in thoracic region. The patient was diagnosed with mechanical back pain and given non-steroidal anti-inflammatory agents and myorelaxants. As she had no relief from the drugs, further diagnostic measures were needed. First, cervical and thoracic magnetic resonance imaging (MRI) was performed. Although no significant pathology was present in cervical MRI, thoracic MRI revealed a 9 mm lesion, both hypointense on T₁ and T₂-weighted MRI, located anterior to the left lamina of T₃ vertebra, compressing the spinal cord from left posterior and considered as focal hypertrophy/OLF. Then, computed tomography (CT) showed ossification in the lesion at T₃ level, thus suggesting the final diagnosis as OLF. The width of the anteroposterior spinal canal was 13.7 mm at the midline, 9 mm at the medial border of the right superior articular process, and 5.1 mm at the medial border of the left superior articular process (Figure 1). Although there was no sign of myelopathy in our case, the patient was advised for surgical therapy due to persistent pain. However, she refused surgery for unknown reasons.

DISCUSSION

There are various degenerative conditions which may lead to thoracic stenosis such as thickening or OLF, facet joint hypertrophy, ossification of the posterior longitudinal ligament, ventral disc protrusion or osteophytic changes.^[6] Additionally, systemic metabolic diseases such as acromegaly, achondroplasia, or Paget's disease may lead to thoracic stenosis, where

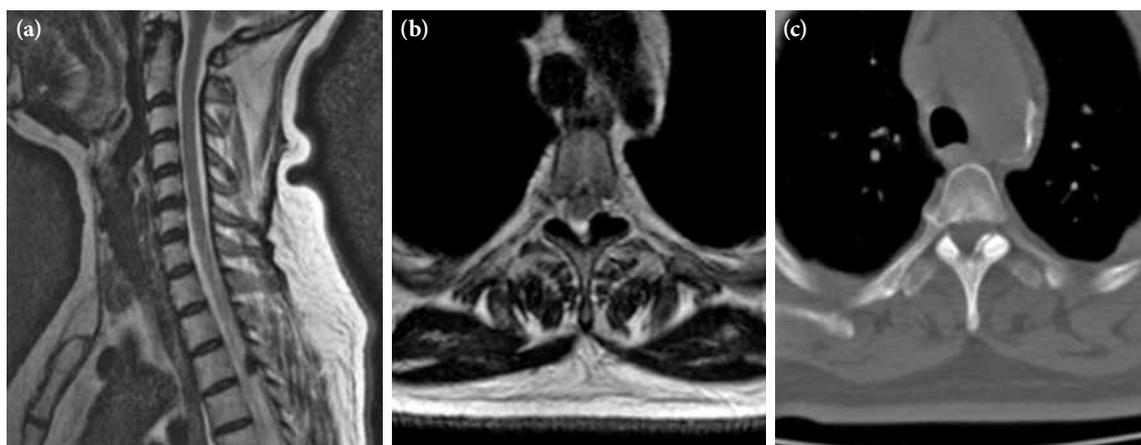


Figure 1. (a) Sagittal magnetic resonance imaging showing beak-like ossified ligamentum flavum protruding toward the spinal canal. (b) On axial magnetic resonance imaging, compression of spinal cord by laterally located of the ligamentum flavum can be seen. (c) Computed tomography scan showing calcification along the border of ligamentum flavum.

the spinal canal becomes narrow circumferentially and extends to the multiple segments.^[7] Degenerative thoracic stenosis is most commonly seen in the lower thoracic segments which are under local mechanical stress due to the repetitive motions.^[5] Symptoms may vary according to the patient's age as well as the cause and level of the stenosis.^[3]

Ossification of ligamentum flavum, although commonly seen in aging Japanese population, is rare in Caucasian people and Americans.^[8,9] It is characterized by hypertrophy and endochondral ossification of ligamentum flavum which leads to stenosis by progressively extending into the spinal canal.^[7] As lower thoracic spine sustains greater forces due to frequent motion, severity of OLF increases as thoracolumbar junction is approached. Although the exact etiology is unknown, the incidence of thoracic OLF is reported to be higher in patients with diffuse idiopathic skeletal hyperostosis, fluorosis, diabetes mellitus, ankylosing spondylitis, and ossification of the posterior longitudinal ligament.^[10] Our case had diabetes mellitus; however, had a single level OLF. Since most systemic diseases may cause multi-segment OLF, it is controversial whether OLF is due to diabetes mellitus in our case. Possible biomechanical factors in the etiology include limited range of motion, transitional area of spinal curvature, and mechanical stress caused by facet joints and ligamentum flavum including micro-motion.^[11] The disease begins with thickening of ligamentum flavum and, then, ossification occurs along the superficial layer of the thickened ligamentum flavum.^[12] This ossified segment usually compresses the spinal cord, leading to thoracic myelopathy or radiculopathy.^[13] The most frequent initial symptoms are tingling or numbness, while the major complaint is paraparesis, usually spastic among patients. Other symptoms are back pain, gait disturbances, lower limb pain, and bladder dysfunction.^[6]

Diagnostic methods include MRI and CT. While CT shows contours of ossification and additional ossification of dura more precisely, MRI shows the degree of canal compromise and myelopathy more accurately.^[14] Ossification of ligamentum flavum is also classified into five groups according to the CT findings. These are lateral (type 1), extended (type 2), enlarged (type 3), fused (type 4), and tuberous (type 5). Only type 4 and 5 would develop myelopathy, whereas others tend to be asymptomatic.^[15] Those having myelopathy symptoms and asymptomatic patients who are not relieved by conservative treatment are often treated with surgery. Although

our case had type 1 OLF, she was symptomatic and failed to respond conservative treatment. Therefore, we recommend surgery; however, she refused. Furthermore, various surgical techniques are recommended such as decompressive laminectomy, laminoplasty/laminectomy with lateral fusion, and total laminectomy in addition to resection of OLF.^[10-12,16,17] In general, the surgical outcomes are favorable.^[16,17] Of note, our patient will be followed to see the course of the disease.

In conclusion, we present an unusual case of thoracic OLF in a diabetic Caucasian female, single level OLF in the upper thoracic region who was investigated for cervical, lumbar and shoulder pathologies previously. In addition to presenting unusual characteristics of our case, we suggest that retractile pain on the thoracic region should be investigated further, if other more common pathologies are ruled out.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Marzluff JM, Hungerford GD, Kempe LG, Rawe SE, Trevor R, Perot PL Jr. Thoracic myelopathy caused by osteophytes of the articular processes: thoracic spondylosis. *J Neurosurg* 1979;50:779-83.
2. Epstein NE, Schwall G. Thoracic spinal stenosis: diagnostic and treatment challenges. *J Spinal Disord* 1994;7:259-69.
3. Yonenobu K, Ebara S, Fujiwara K, Yamashita K, Ono K, Yamamoto T, et al. Thoracic myelopathy secondary to ossification of the spinal ligament. *J Neurosurg* 1987;66:511-8.
4. Panjabi MM, Takata K, Goel V, Federico D, Oxland T, Duranceau J, et al. Thoracic human vertebrae. Quantitative three-dimensional anatomy. *Spine (Phila Pa 1976)* 1991;16:888-901.
5. Barnett GH, Hardy RW Jr, Little JR, Bay JW, Sybert GW. Thoracic spinal canal stenosis. *J Neurosurg* 1987;66:338-44.
6. Chang UK, Choe WJ, Chung CK, Kim HJ. Surgical treatment for thoracic spinal stenosis. *Spinal Cord* 2001;39:362-9.
7. Schmidt RF, Goldstein IM, Liu JK. Ossified ligamentum flavum causing spinal cord compression in a patient with acromegaly. *J Clin Neurosci* 2013;20:1599-603.
8. Nakagawa H. Pathophysiology and surgical strategies in the management of the ossified spinal ligaments. *World Neurosurg* 2011;75:443-4.
9. Amato V, Giannachi L, Irace C, Corona C. Thoracic spinal stenosis and myelopathy: report of two rare cases and review of the literature. *J Neurosurg Sci* 2012;56:373-8.

10. Li F, Chen Q, Xu K. Surgical treatment of 40 patients with thoracic ossification of the ligamentum flavum. *J Neurosurg Spine* 2006;4:191-7.
11. Yoon SH, Kim WH, Chung SB, Jin YJ, Park KW, Lee JW, et al. Clinical analysis of thoracic ossified ligamentum flavum without ventral compressive lesion. *Eur Spine J* 2011;20:216-23.
12. Okada K, Oka S, Tohge K, Ono K, Yonenobu K, Hosoya T. Thoracic myelopathy caused by ossification of the ligamentum flavum. Clinicopathologic study and surgical treatment. *Spine (Phila Pa 1976)* 1991;16:280-7.
13. Sagar H, Fernandez-Madrid F, Kupsky W. Rare cause of thoracic myelopathy: ossified ligamentum flavum. *J Clin Rheumatol* 2010;16:326-9.
14. Hanakita J, Suwa H, Ohta F, Nishi S, Sakaida H, Iihara K. Neuroradiological examination of thoracic radiculomyelopathy due to ossification of the ligamentum flavum. *Neuroradiology* 1990;32:38-42.
15. Muthukumar N. Dural ossification in ossification of the ligamentum flavum: a preliminary report. *Spine (Phila Pa 1976)* 2009;34:2654-61.
16. 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.
17. Yu S, Wu D, Li F, Hou T. Surgical results and prognostic factors for thoracic myelopathy caused by ossification of ligamentum flavum: posterior surgery by laminectomy. *Acta Neurochir (Wien)* 2013;155:1169-77.