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Case Report

Ossification of the ligamentum flavum as cause of thoracic cord compression: Case report of a Latin American man and review of the literature

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Abstract

Background: Ossification of the ligamentum flavum is a widely described pathology in eastern Asia. Cases have been reported in northern Africa, the Middle-East, India, the Caribbean, Europe, and North America, but no cases from Latin America have been published in the literature. It affects mostly elderly men, with a possible association with obesity and type 2 diabetes.

Case Description: A 38-year-old previously healthy Latin American male presented to the emergency room department with severe functional disability and a 3/5 paraparesis. Blood reports showed no abnormalities. Computed tomography and magnetic resonance imaging showed a ligamentum flavum ossification with myelopathy. The patient underwent a T3-T9 laminotomy. At hospital discharge, the patient remained with a 3/5 paraparesis, mild hypoesthesia in both lower limbs and bladder incontinence. Rectal sphincter was continent. At 6 months, he was able to walk with a cane, with no sphincter or sensory alterations.

Conclusions: Ligamentum flavum ossification is rare. To our understanding, this is the first case reported in the Latin American population.

Key Words: Dorsal stenosis, ligamentum flavum ossification, thoracic myelopathy

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INTRODUCTION

Ossification of the ligamentum flavum is a widely described pathology in eastern Asia and rare in other parts of the world.[11] There have been reported cases in northern Africa, the Middle-East, India, the Caribbean, Europe, and North America, but no reported cases in Latin America.[1,3,7-9] The condition is found most frequently in elderly men, with a possible association with obesity and type 2 diabetes.[2]

The disease process first causes hypertrophy of the ligamentum flavum and subsequent ossification, which narrows the spinal canal and leads to myeloradiculopathy.[7,8,13,16] The lower thoracic spine is most commonly affected. Previous hyperkyphosis and mechanical stress are thought to be predisposing conditions.[11]

This is the first case report of ligamentum flavum ossification in the Latin American population, seen in a healthy middle-aged man with a multi-level thoracic lesion.

CASE REPORT

A 38-year-old male patient with a normal medical history, presented with a 6 month progressive gait disturbance and urinary incontinence. There was no history of previous trauma. Physical examination showed spastic paraparesis (grade 3/5), bilateral positive Babinski sign, with increased deep tendon reflexes. Sensory level at T9 was ascertained. Rectal sphincter was preserved.

Imaging

Dorsal spine magnetic resonance imaging (MRI) was obtained, which showed hypertrophy and ossification of the posterior ligamentous complex from T3 to T10 and underlying myelopathy. No discal pathology was reported. Computed tomography (CT) demonstrated bone density all long the posterior segment of the dorsal spinal canal and a 50% reduction of anteroposterior diameter [Figures 1 and 2]. Imaging of the cervical and lumbar spine showed no abnormalities.

Laboratory

Serum calcium, phosphorous, uric acid, and glucose levels were normal. A discrete elevation of alkaline phosphatase level was observed (485 mIU/ml, normal range up to 306 mIU/ml).

Surgery

Patient underwent a dorsal approach under general anesthesia in a prone position. A midline incision was made and the T3 to T9 lamina were removed. Extensive drilling of the ossification was performed. Thinned laminas were attached to the spinous process of the superior and inferior vertebrae.

Clinical outcome

Patient underwent an intensive care unit stay, complicated with paralytic ileus and respiratory tract infection. At hospital discharge, patient remained paraparetic with a 3/5 muscular strength, mild hypoesthesia in both lower limbs, and bladder incontinence. Rectal sphincter was continent. At 6 months, he was able to walk with a cane, with no sphincter or sensory alterations.

DISCUSSION

Although several studies have tried to explain the chemical mechanism that leads to the ossification in this pathology, it remains unclear.[13] At least two phenomena must be present for the disease to manifest: Hyperplasia of the ligament and its subsequent ossification. If ectopic bone formation is limited to the original thickness of the ligamentum flavum, it would never compress the spinal cord. Patients suffering from ligamentum flavum ossification that leads to spinal cord compression have been known to have preossification tissue alterations including a neovascularization that promotes infiltration of mesenchymal cells and alters matrix mineralization, particularly in the area of the ossification. Cell proliferation and matrix hyperplasia is stimulated primarily by transforming growth factor beta and bone morphogenetic proteins.[8,13,17]

This pathology is found most commonly in the thoracic region.[11] It is thought that the hyperkyphosis of this region of the spine is associated with mechanical stress, which makes ligaments in this region more prone to ossification.[11] The mechanism of ligament ossification is classified into five types: Type I, laterally at the origin of the ligamentum flavum at the articular processes; Type II, from the lateral origin of the ligamentum flavum to the interlaminar portion of the ligamentum flavum; Type III, protrudes into the canal posterolaterally but is not fused in the midline; Type IV, consists of bilateral

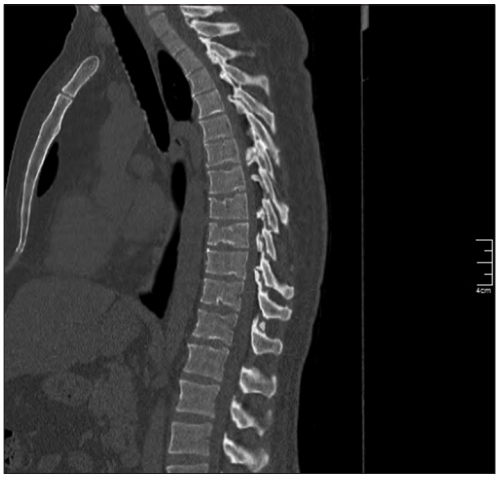

Figure 1: Sagittal view, CT scan showing hyperdense images of a reduction in the anteroposterior diameter of the dorsal spinal canal

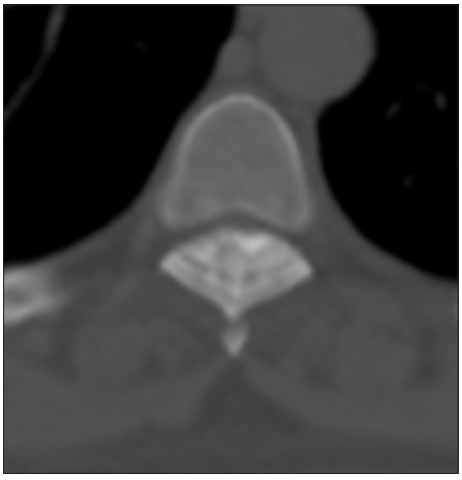

Figure 2: Axial CT scan at the level of the T-6 neural foramen demonstrating significant posteromedial compression

ossified ligaments that are fused at the midline with a groove at the fusion in midline; and Type V, the tuberous type, occurs when the fused ossified ligamentum flavum forms a "tuberous" mass posteriorly in the midline, which protrudes into the spinal canal. According to this classification only types IV and V would develop a myelopathy, and most of types I, II, and III would remain asymptomatic.[1,12]

MRI and CT scans are necessary for diagnosis as both studies provide necessary information for surgical planning. CT scan delineates precisely the anatomy of the spine and shows the ossification of the ligament more precisely than MRI, and can show if there is dura matter ossification, which is essential to planning the surgical approach.[12] MRI is helpful for showing spinal cord involvement, especially when, as in our case, compression is multi-segmental.[14]

The surgical treatment for this pathology depends on which level is involved and whether or not there is multi-level involvement. For most cases, laminectomy is the chosen technique as it provides a wide decompression of the medullar canal.[18] An exception to this technique can be done, as in our case, when compression involves multiple levels of the spine. A multiple level laminectomy can result in further kyphosis. Laminoplasty with detailed drilling of the ossificated ligamentum flavum is also an acceptable option when multiple levels are involved.[6] The most common complication with this approach are dural tears, which are present in about 30% of procedures, especially when dura mater is involved in the ossification process.[4,10]

An independent factor in functional outcome is the time from initial symptom presentation to the spinal decompression. Additionally, clinical factors that can predict postoperative recovery include sex, age, level of ossification of the lesion, number of levels affected, ossification of the lateral ligament, dura mater calcification, and MRI signs of myelopathy.[5,15] Close follow-up of these patients is critical as they are at increased risk for subsequent calcification of the posterior longitudinal ligament, new ossifications at different levels of the spine, and/or thoracic kyphosis.

CONCLUSION

Ligamentum flavum ossification is a rare entity. To our understanding, this is the first case reported in the Latin American population.

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REFERENCES

1. Christiano LD, Assina R, Goldstein IM. Ossification of the ligamentum flavum: A unique report of a Hispanic woman. Neurosurg Focus 2011;30:E15.
2. Fan D, Chen Z, Chen Y, Shang Y. Mechanistic roles of leptin in osteogenic stimulation in thoracic ligament flavum cells. J Biol Chem 2007;282:29958-66.
3. Fong SY, Wong HK. Thoracic myelopathy secondary to ligamentum flavum ossification. Ann Acad Med Singapore 2004;33:340-6.
4. Hur H, Lee JK, Lee JH, Kim JH, Kim SH. Thoracic myelopathy caused by ossification of the ligamentum flavum. J Korean Neurosurg Soc 2009;46:189-94.
5. Inamasu J, Guiot BH. 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.
6. Iwasaki M, Kawaguchi Y, Kimura T, Yonenobu K. Long-term results of expansive laminoplasty for ossification of the posterior longitudinal ligament of the cervical spine: More than 10 years follow up. J Neurosurg Spine 2002;96;2 Suppl:180-9.
7. Jayakumar P, Devi BI, Bhat D, Das B. Thoracic cord compression due to ossified hypertrophied ligamentum flavum. Neurol India 2002;50:286-9.
8. Kim HN, Min WK, Jeong JH, Kim SG, Kim JR, Kim SY, *et al.* Combination of Runx2 and BMP2 increases conversion of human ligamentum flavum cells into osteoblastic cells. BMB Rep 2011;44:446-51.
9. Kim TJ, Kim TH, Jun JB, Joo KB, Uhm WS. Prevalence of ossification of posterior longitudinal ligament in patients with ankylosing spondylitis. J Rheumatol 2007;34:2460-2.
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. Maigne J, Ayral X, Guerin-Surville H. Frequency and size of ossifications in the caudal attachements of the ligamentum flavum of the thoracic spine. Role of rotatory strains in their development. Surg Radiol Anat 1992;14:119-24.
12. Muthukumar N. Dural ossification in ossification of the ligamentum flavum: A preliminary report. Spine 2009;34:2654-61.
13. Ono K, Yonenobu K, Miyamoto S, Okada K. Pathology of ossification of the posterior longitudinal ligament and ligamentum flavum. Clin Orthop Relat Res 1999;359:18-26.
14. Sonntag VKH. Ossification of the Ligamentum Flavum (OLF): An increasing cause of cervical myelopathy. World Neurosurg 2011;75:445-6.
15. Sun C, Chen Z, Liu Z, Liu X, Qi Q, Guo Z, *et al.* Longterm outcome after the decompressive surgery for thoracic myelopathy due to the ossification of the ligamentum flavum. Zhonghua Wai Ke Za Zhi 2012;50:426-9.
16. Takeuchi Y, Matsumoto T, Takuwa Y, Hoshino Y, Kurokawa T, Shibuya N, *et al.* High incidence of obesity and elevated serum immunoreactive insulin level in patients with paravertebral ligamentous ossification: A relationship to the development of ectopic ossification. J Bone Miner Metab 1989;7:17-21.
17. Uchida K, Yayama T, Cai HX, Nakajima H, Sugita D, Guerrero AR, *et al.* Ossification process involving the human thoracic ligamentum flavum: Role of transcription factors. Arthritis Res Ther 2011;13:R144.
18. Yamazaki M, Koda M, Okawa A, Aiba A. Transient paraparesis after laminectomy for thoracic ossification of the posterior longitudinal ligament and ossification of the ligamentum flavum. Spinal Cord 2005;44:130-4.