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Case Report

Extensive ossification of the ligamentum flavum treated with triple stage decompression: a case report

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Abstract

BACKGROUND CONTEXT: Concurrent ossification of the ligamentum flavum (OLF) in the cervical, thoracic, and lumbar spine is a rare occurrence often associated with rheumatologic abnormalities. Although the pathology may be asymptomatic and discovered incidentally on routine imaging, compression of the cord and surrounding nerve roots can produce myelopathic or radiculopathic symptoms that are best treated with surgical decompression. There is limited evidence to support the use of single versus multistage decompression for tandem ossification at multiple levels, although several factors including duration of symptoms have been associated with a worse prognosis.

PURPOSE: To describe the presence of extensive symptomatic tandem OLF with concurrent ossification of the posterior longitudinal ligament (PLL) and its treatment using multistage decompression.

STUDY DESIGN: Case report and literature review.

METHODS: The authors describe a case of a 35-year-old woman with OLF extending from the cervical to lumbar spine and tandem ossification of the cervical PLL. Her initial presentation was significant for symptoms consistent with thoracic myelopathy in the absence of radiculopathic findings, and initial imaging also demonstrated disc herniation at L4–L5 and L5–S1.

RESULTS: The patient was first treated with a thoracic laminectomy and fusion from T7 to T11, given her back pain and thoracic myelopathy. Persistence of myelopathic symptoms necessitated further surgical intervention with a posterior cervical decompression and fusion from C3 to T1. Finally, after the appearance of radiculopathic findings, she underwent a microscopic L4–L5 laminectomy with improvements in her symptoms and ambulation.

CONCLUSIONS: Symptomatic OLF in non-East Asian population is a rare occurrence. Its etiology is likely multifactorial, involving both biomechanical and genetic factors. Although early detection and management are necessary, multistage decompression can be an effective intervention for extensive multilevel ossification.

Keywords: Ossification ligamentum flavum; Tandem ossification; Posterior longitudinal ligament; Myelopathy; Decompression; Fusion

Introduction

Ossification of the ligamentum flavum (OLF) is caused by an enthesopathy involving the replacement of the ligamentum flavum, with calcifications likely secondary to a combination of intrinsic and extrinsic factors. First described by Polgar in the 1920, OLF has been reported primarily in East Asian population and particularly the Japanese, where its prevalence is as high as 63.9% [1,2]. Additional studies have demonstrated cases in Caucasians, African Americans, and other Asian populations, but the reported rates are far less [3]. The disorder can exist in the general population, but those affected are often asymptomatic and are discovered incidentally on routine imaging. When symptoms occur, the most common pathology is thoracic

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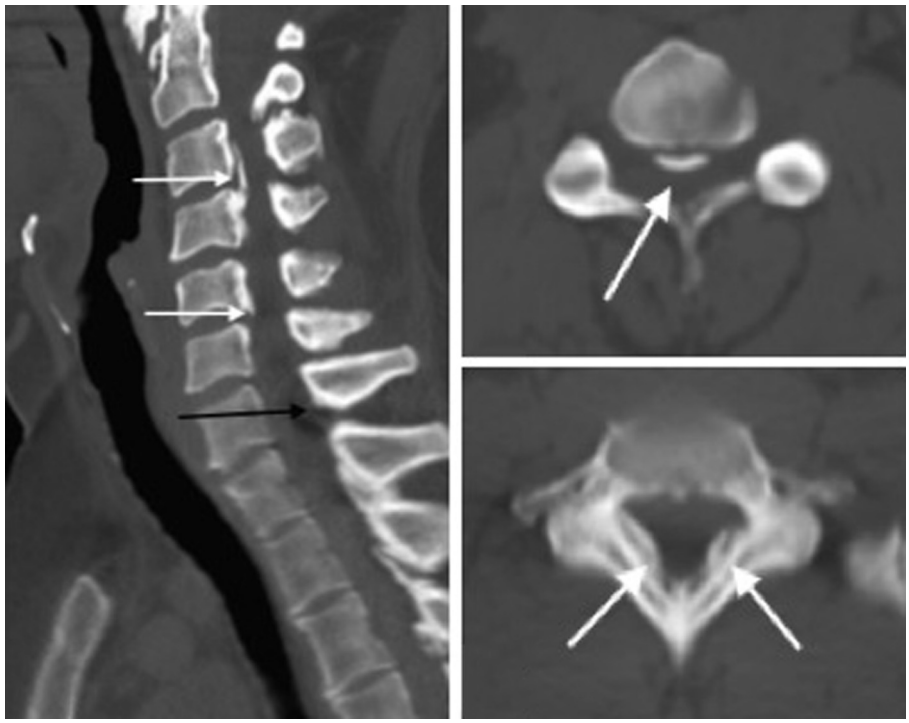

Fig. 1. Preoperative computed tomography (CT) of the cervical spine. (Left) Sagittal CT demonstrating prominent ossification of the posterior longitudinal ligament (OPLL) at C3–C4 and C5–C6 (*white arrows*) and ossification of the ligamentum flavum (OLF) at C7–T1 junction (*black arrows*). (Top Right) Axial CT at C3–C4 demonstrating OPLL (*white arrow*). (Bottom Right) Axial CT at C7–T1 junction demonstrating OLF (*white arrows*).

spondylotic myelopathy resulting from compression of the spinal cord. As such, patients often present with symptoms such as muscle weakness, numbness of the lower extremity and torso, and back and lower extremity pain [1,4,5].

Diagnosis of OLF occurs through a combination of plain radiograph and computed tomography (CT) imaging. On plain radiograph, calcification of the posterior elements of the spinal canal may indicate OLF; however, further investigation with CT is often needed for confirmation. On sagittal CT, thickening and calcification of the laminar components can be identified and used to accurately diagnose OLF [5–7]. Once diagnosed, the goal of surgical intervention is decompression by removing the calcific areas that are responsible for the spinal canal narrowing and subsequent cord compression [4,5,7].

Although OLF occurs most commonly in the lower thoracic spine, it can present with concurrent OLF in the cervical or lumbar spine or ossification of the posterior longitudinal ligament (OPLL) at other spinal levels [1,4,5,8]. Although several cases of thoracic OLF-induced myelopathy have been reported, we are aware of few cases with such diffuse involvement of the spinal column. This article reports a case of extensive OLF involving the cervical, thoracic, and lumbar spine with coexisting OPLL at the cervical spine.

Case report

A 35-year-old African American woman presented with increasing difficulty in gait, a right foot drop, and severe

Fig. 2. Sagittal T2-weighted magnetic resonance images of the cervical spine on presentation demonstrating hypertrophy of the posterior longitudinal ligament at C3–C4 and C4–C5 (*arrows*), with resultant cervical stenosis at C3–C4 and mild stenosis at C4–C5 and C5–C6.

Fig. 3. Preoperative computed tomography (CT) of the thoracic spine. (Left) Sagittal view shows ossification of the ligamentum flavum (OLF) extending from C7 to T12, most prominent at caudal levels. (Top Right) Axial CT at T2–T3 demonstrating moderate bilateral OLF (*arrows*). (Bottom Right) Axial CT at T9 demonstrating severe OLF with significant central stenosis (*arrow*).

thoracic back pain. One year before presentation, the patient was able to ambulate with a normal gait and had worked as a bank teller. She noticed progressive difficulty in walking over the course of a year and was seen previously by an outside spine surgeon who according to the patient had diagnosed her with OLF. Comorbidities included asthma and hypertension, both well-controlled with medical management. She denied any major difficulty using her hands or an inability to grasp objects. She had no upper motor neuron signs in her upper extremities but demonstrated a Babinski reflex in her lower extremity.

Preoperative cervical CT revealed linear OPLL at C3–C4 and C5–C6, along with mild OLF at C6–C7 and C7–T1 (Fig. 1). Cervical magnetic resonance imaging demonstrated PLL and ligamentum flavum hypertrophy consistent with CT findings and resultant cervical stenosis at C2–C3 and C3–C4 (Fig. 2). Thoracic CT showed extension of the OLF throughout the thoracic spine, worst at the caudal levels (Fig. 3). Again, magnetic resonance imaging of the thoracic spine demonstrated diffuse ligamentum flavum hypertrophy, consistent with the ossification on CT and severe stenosis at T9–T10 and T10–T11 (Fig. 4). Magnetic resonance imaging of the lumbar spine showed disc desiccation and central posterior disc bulges at L4–L5 and L5–S1 associated with lumbar stenosis. The ligamentum flavum hypertrophy exhibited in the thoracic spine was also found to be contiguous at all levels in the lumbar spine (Fig. 5).

Because of the severe thoracic back pain and myelopathic symptoms, the decision was made to decompress her thoracic spine initially with an intention to decompress her cervical spine at a later date. She underwent a thoracic laminectomy and fusion from T7 to T11 to decompress the posterior thoracic canal (Fig. 6). Her ambulation improved significantly, but she still required an assistive device and continued to endorse myelopathic symptoms at her 6-month follow-up, including gait and balance difficulties. Given the cervical stenosis seen on initial imaging and the presence of ongoing myelopathic symptoms, we proceeded with a posterior cervical decompression and fusion

Fig. 4. Preoperative sagittal T1-weighted magnetic resonance images of the thoracic spine demonstrating extensive hypertrophy of the ligamentum flavum, consistent with the ossification of the ligamentum flavum demonstrated on computed tomography. Also shown is the resulting spinal canal stenosis most severe at T9–T10 and T10–T11 and moderate stenosis at T8–T9.

Fig. 5. Preoperative sagittal T2-weighted magnetic resonance images of the lumbar spine on presentation demonstrating disc desiccation and posterior disc protrusion at L4–L5 and L5–S1, consistent with lumbar degenerative disc disease. The ligamentum flavum hypertrophy consistent with ossification of the ligamentum flavum shown in previous images is also seen throughout the lumbar spine at all levels.

from C3 to T1 (Fig. 6). The patient's recovery was uneventful and she reported improvement in hand function within 2 weeks and improvement in ambulation within 1 month.

Four months after her cervical decompression, the patient complained of new onset of right-sided radicular symptoms in an L5 and S1 distribution. Her previous presentation of a foot drop had improved after her thoracic procedure, but her radicular symptoms had persisted so we proceeded with a microscopic L4–L5 laminectomy. Postoperatively, the patient reported improvements in her pain and ambulation and was able to walk without an assistive device.

Discussion

Although symptomatic thoracic OLF is a relatively rare condition, its association with coexistent cervical OPLL (tandem ossification) is being reported with increasing frequency [8]. However, the incidence of concurrent symptomatic paraspinal ossification is still poorly established. Park et al. [6] reported the presence of tandem ossification in 33.8% of symptomatic cervical OPLL patients, whereas Gao et al. [9] found coexistent disease in 54.7% of patients with multilevel OLF, of which 14.7% had cervical OPLL. First described by Koizumi [10] in 1962, the presence of hyperostotic lesions in the lumbar spine in addition to cervical and thoracic enthesopathies is an even rarer occurrence with few reported cases in the literature.

Many of the reported cases of coexistent ossification at all three levels have been associated with rheumatologic abnormalities, such as diffuse idiopathic skeletal hyperostosis [10–12]. Given this possibility, we referred our patient to a rheumatologist for further evaluation and her symptoms appeared to be limited to the spine. Current theory regarding the etiology of OLF and OPLL has implicated various transcription and growth factors, hormones, and genetic polymorphisms that result in fibrocartilage proliferation and endochondral bone formation [13,14]. Mechanical stress is also believed to play a role in thoracic OLF, with tensile forces playing a large role given the limited mobility of the thoracic spine relative to the cervical and lumbar regions [15]. This is complicated by concurrent OPLL, which may limit the normal range of motion in the cervical spine, resulting in hypermobility of the cervicothoracic junction and upper thoracic region [6]. Both of these mechanical forces, a tensile component in the

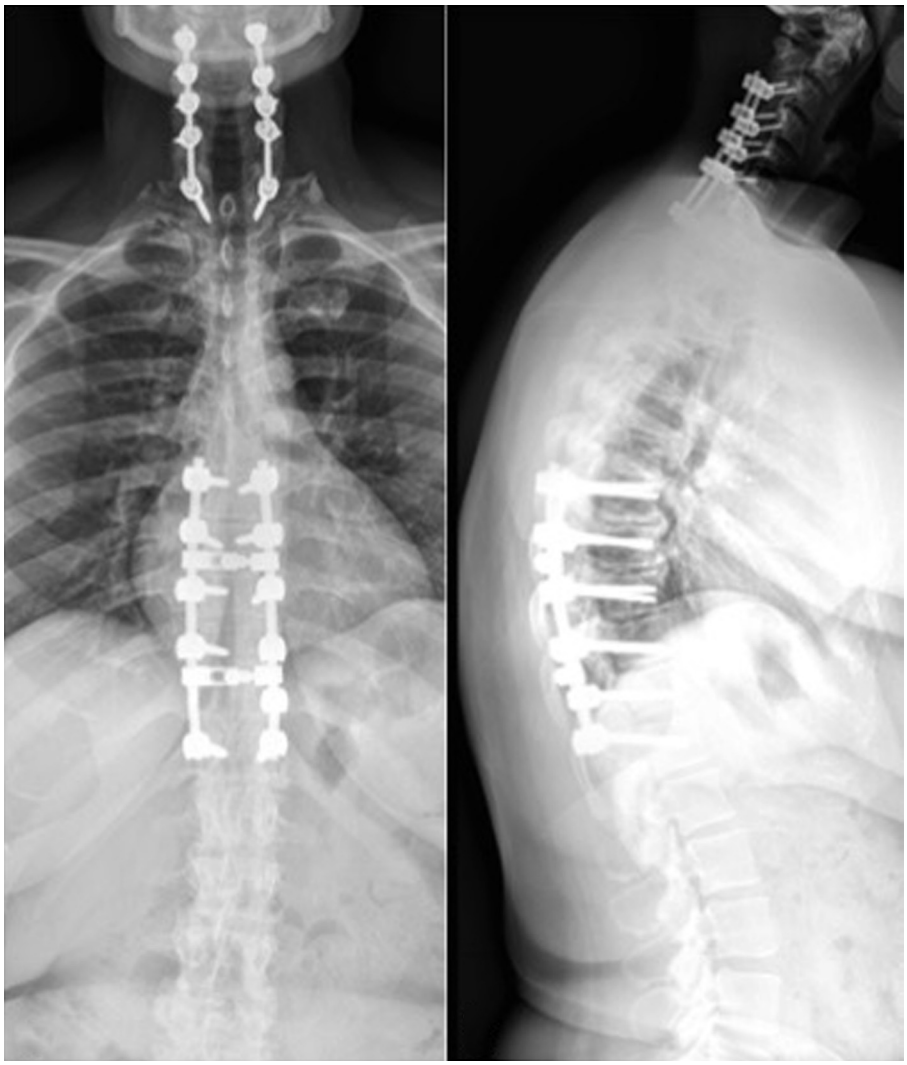

Fig. 6. Postoperative (Left) anteroposterior and (Right) lateral plain film radiographs of the thoracic and cervical spines after the first and second stage decompressions with fusion from T7–T11 and C3–T1, respectively.

thoracolumbar region and hypermobility at the C7–T1 junction, would help to explain the contiguous OLF seen throughout this patient's thoracic spine, rather than the typical concentration in the lower thoracic vertebrae that is most commonly reported.

The diagnosis and management of symptomatic tandem ossification can be challenging, especially in cases where paraspinal ossification occurs at multiple levels. In these instances, differentiating between incidental and causative lesions is often difficult. Current management stresses surgical decompression, however, there is no consensus regarding the optimal surgical strategy when faced with coexistent ossifications at multiple spinal regions, including the decision whether to fuse after decompression [7]. Previous reports have demonstrated recurrence after decompression alone, possibly secondary to persistent micromotion at the affected levels [12]. When there is a concurrent symptomatic ossification, surgical options include multistage intervention, with cervical, thoracic, or lumbar decompression occurring first depending on the severity and clinical findings, or single-stage intervention with the combined decompression of all involved regions. Chen et al. [4] reported comparable clinical outcomes for single-stage combined decompression of the cervical and thoracic spine; however, such procedures carry an increased incidence of complications and are highly invasive [5].

Here we proceeded with a triple-stage decompression, with symptom improvement after lumbar decompression. This patient's postoperative recovery was excellent, although she had two poor prognostic factors; the first being a relatively prolonged period of symptoms before surgical intervention, which has been correlated with poor outcomes after surgical decompression of thoracic OLF [16–18]. The second factor was the presence of coexistent ossification, which has also been associated with worse clinical outcomes [5,19]. However, the identification of additional spinal pathology may be due to the slowly progressive nature of OLF, such that by the time symptoms occur,

secondary areas in the paraspinal ligaments are already involved but have little clinical relevance. This has been demonstrated in numerous studies that have failed to show the presence of concurrent disease as a prognostic indicator of postoperative outcomes [3,17,18].

Conclusions

This article reports a case of extensive tandem ossification involving cervical OPLL and OLF, thoracic OLF, and lumbar OLF-induced myelopathy and radiculopathy. Symptomatic OLF in non-Asian populations at each of these sites individually is a relatively rare occurrence and few cases have been reported in the literature involving their extensive coexistence as seen in this patient. In such cases, the etiology is likely multifactorial involving both mechanical and intrinsic factors. Although the diagnosis and choice of intervention can be difficult, early detection and management are critical to postoperative success.

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