

Case Report



Ossification of the Ligamentum Flavum of the Cervical Spine in an Iranian Woman: Report of a Case With Myelopathy and Review of the Literature

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ABSTRACT

Background and Aim: Ossification of the ligamentum flavum (OLF) is a pathological condition that affects the yellow ligaments, mostly in adult males between the ages of 45 to 70. Ossification is more common in the thoracolumbar region with an accompanying progressive neurological deficit, and its occurrence within the cervical spine is quite rare. Ossification of the yellow ligament is so common in East Asian countries that it is regarded as a frequently occurring aging pathology in the people of this region. Its existence in other ethnic groups is significantly lower.

Case Presentation: A 68-year-old Iranian woman with progressive quadriparesis is presented. Cervical computerized tomography and MRI scans revealed two-level, bilateral, posteriorly located, ossified masses occurring at the C4-C5 and C5-C6 levels, which coincided with the ossification of the yellow ligament. Cervical laminectomy and excision of both masses were undertaken and the patient gradually recovered from his resultant neurological symptoms.

Conclusion: Symptomatic cervical OLF is a markedly rare pathological finding and is typically illustrated as a medical case report. In a careful review of the literature including the presented patient, we could uncover only 77 cases involving a documented symptomatic cervical OLF.

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Highlights

- Calcification of ligamentum flavum (COLF) is less appreciated but is a potentially serious cause of myeloradiculopathy within the aging population.
- Despite the relative rarity of cervical OLF compared to Thoracic ossification of ligamentum flavum (TOLF), it is increasingly being reported in a wide range of ethnic groups.
- Cervical OLFs usually progress slowly and make an early diagnosis very difficult.
- Hopefully, with the increased use of MRI, symptomatic COLFs will be diagnosed in earlier stages.

Plain Language Summary

In cervical MRI, a posteriorly located hypointense mass at the site of the ligamentum flavum means ossification or calcification of the ligamentum flavum. The presence of mature bone within the ligament is the notable difference between the ossification of the ligamentum flavum and the calcification of this ligament. Intraoperative dural tearing is less frequent in cervical OLF in comparison to thoracic ones due to less dural adhesion. Early diagnosis of a cervical OLF is necessary for a favorable outcome.

1. Introduction

The first example of cervical OLF was described in 1962 by Koizumi who reported this pathology in an autopsy of a 55-year-old man who had progressive quadriparesis for the last two years of his life [1]. The second instance was described by Kirita in 1973 in a woman with cervical myelopathy [2]. Since then, with the inclusion of the current presentation, 75 additional cases have been published so far [3-8]. Herein, we present an elderly Iranian woman with symptomatic cervical OLF who was managed by our team's practice.

2. Case Presentation

A 68-year-old old woman was admitted due to an unsteady gait and weakness within all four limbs. Her neurological examination revealed alertness with spastic quadriparesis more prominent within the lower extremities. The tendon reflexes were hyperactive with positive Hoffmann and Babinski signs bilaterally. Her modified Japanese Orthopedic Association (mJOA) score at the time of admission was noted to be 10. MRI revealed a set of 2-level, bilaterally and posteriorly located masses compressing the dural sac at the C4-C5, and C5-C6 levels. Both were hypointense on T1- and T2-weighted images, compatible with the ossification of the ligamentum flavum (Figure 1a). A myelopathic patch was also demonstrated at the C4-C5 spinal level (Figure 1b). Axial computed tomography (CT scan) demonstrated ellipti-

cal, immense, and ossified masses, at both levels (Figure 2). With the diagnosis of a 2-level bilateral OLF, the patient underwent a C4 to C6 laminectomy. After making a longitudinal groove on the side of the lateral masses, laminectomy with a high-speed air drill was undertaken. It was at this time that a level of dural sac compression was noted to be produced by overtly large ossified masses. Subsequently, the masses displayed a mild adhesion to the dura and could be mobilized and then removed. After resection of the mass, the dural sac showed the expected re-expansion and adequate pulsation.

Histological examination of the surgical specimens revealed islands of endochondral ossification within the elastic ligament fibers which were compatible with the diagnosis of OLF (Figure 3). The post-operative course recovery was uneventful and the patient showed some evidence of regained function immediately following the surgical intervention. At one-year follow-up, her mJOA reached a scale score of 15, and surprisingly the mid-cervical myelopathic patch was resolved in the control MRI (Figure 4).

3. Discussion

Incidence

OLF most often affects the thoracic and lumbar regions whereas effect of the cervical spine leading to a resultant myeloradiculopathy remains a much infrequent occurrence [9-12]. The prevalence of asymptomatic OLF in East

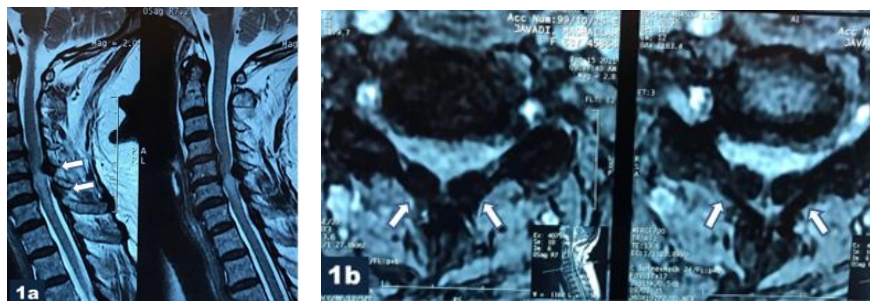


Figure 1. Cervical MRI

(a) T2-weighted axial images demonstrate bilateral hypointense massing levels at the corresponding levels. (b) T2-weighted sagittal image showing a hypointense epidural mass at the posterior aspect of the spinal cord at the C5-C6 level, a smaller one is visualized below this level at C6-C7 white arrows. A large myelopathic patch is demonstrated at the corresponding levels.

Asian populations is 38.5% and 26.5% in the thoracic and lumbar regions respectively, where its occurrence within the cervical spine is less than 1% [10, 13-16].

In a study conducted by Hiraoka et al., the incidence of OLF was found to be 34% in the postmortem autopsies of a large group of aged patients [17]. Accordingly, all discovered OLFs were overwhelmingly confined to the thoracic or lumbar spinal regions and not a single patient possessed such an anomaly within the cervical spine [17].

Fujimori et al. conducted a research study involving 1500 Japanese patients utilizing a multi-detector computed tomography scan [13]. While the prevalence of asymptomatic OLF within the thoracic spine was found to be 12%, its prevalence within the cervical spine was 0.3% [18].

The high rate of OLF within the thoracic spine is the direct result of the constant tension placed on the ligamentum flavum induced through the presence of thoracic kyphosis [9-14]. Contrarily, the exposure of the ligamentum flavum at the cervical spine to tension is shown to be less than that displayed within other regions of the vertebral column. Moreover, lordotic curvature of the cervical spine and the difference in the thickness of the yellow ligaments compared to the thoracic segments may also help to explain the overall low prevalence of COLF [3-8].

Ethnicity

Most of the published cases with cervical OLF are of East Asian ethnic descent [1-8]. However, its occurrence in Caucasians is not as infrequent as it was once supposed [3].

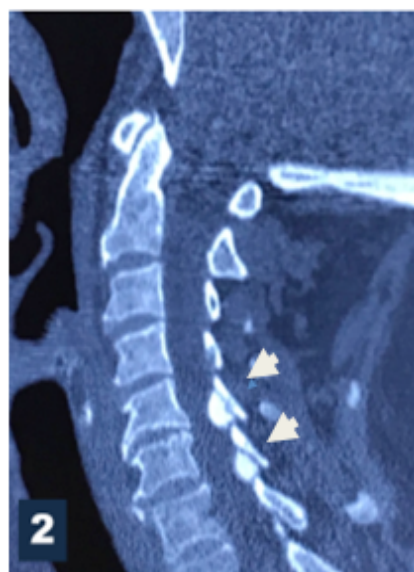


Figure 2. Sagittal reconstructed cervical computed tomography scan (CT scan) showing two calcified masses between C5-C6 and C6-C7, at the site of the ligamentum flavum (white arrowheads).

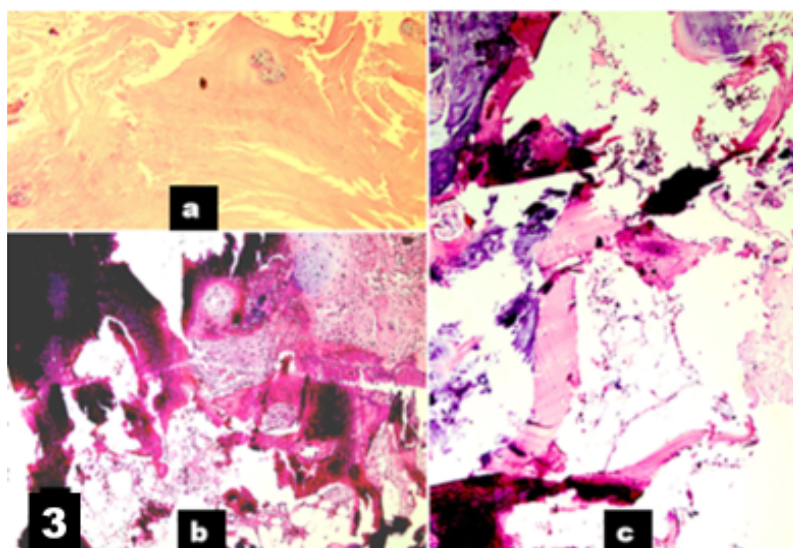


Figure 3. Histological examination of the surgical specimen



(a) Dense fibrotic tissue of ligamentum flavum having 3 small chondroid islands. (b) (Hematoxylin & Eosin stained tissue section; extensive areas of amorphous calcifications at the left side (dark purple), along with chondroid and osseous metaplasia are seen and fatty bone marrow is also present at the bottom of the field (c) Higher magnification shows fatty and hematopoietic bone marrow elements surrounded by pinkish mature bone trabeculae.

Clinical picture

Typical clinical features of COLFs at the time of diagnosis include an unsteady gait, decreased muscle strength, sensory disturbances, and urinary dysfunction. Such findings are noted to be indistinguishable from cord compressions seen within cervical spondylotic myelopathies [3-8]. The interval from the onset of the disease to the correct diagnosis is usually very lengthy and can range from one to several years.

Imaging

The exact location and shape of the OLF, the number of affected levels, as well as the degrees of cord compression and the associated myelopathies can be best demonstrated by utilization of MRI [3-8]. A typical cervical OLF is visualized as a hypointense signal intensity mass at the level of the posterior margin of the spinal canal when viewed using T1-weighted sagittal MR images. A patient with cervical OLF myelopathy is observed as an intramedullary hyper-intense spot within T2-weighted sagittal views at the time of diagnosis [3-8].



Figure 4. Postoperative sagittal MRI after at 1-year follow-up



(a) T1-weighted image shows appropriate laminectomy. (b) T2-weighted image, note the disappearance of the myelopathic patch.

Most cervical OLFs are located within the subaxial segments and their occurrence in the upper cervical region is rare [3-8].

It is important to assess the exact size, shape, and extent of the ossified lesions before surgical intervention. To accomplish this goal, computerized tomography should be performed. On axial CT scans, the lesions are visualized as ossified masses from the facet joint to the base of the spinal process and can be either unilateral or bilateral [19, 20]. Reconstructed images of an OLF are primarily shown as an ossified mass at the posterior aspect of the spinal column at the site of the ligamentum flavum [3-8, 18-20].

Treatment

Symptomatic cases with cervical OLF should be followed closely with periodic examinations and MRI scans. A decompressive laminectomy and excision of the ossified masses are the mainstay of treatment in symptomatic sub-types [3-8]. The indications for surgery are cervical radiculopathy, myelopathy, or myeloradiculopathy. Bilateral occurrences usually display a tight adherence to the dura. Unilateral nodular OLF sub-types typically show a minimal amount of adhesion to the dura mater. A dural tear is not considered a frequent complication during the surgical removal of a cervical OLF.

Differential

Cervical OLF and cervical ligamentum flavum calcification (CLF) which are separate unique entities are sometimes confused with one another due to the many similarities they present [20, 21]. Imaging features and intraoperative findings will likewise mirror each other [20, 21]. The calcification of the ligamentum flavum, pseudogout, and calcium pyrophosphate dehydrate crystal deposition disease (CPPD), all belong to a single sub-category and can only truly be differentiated from OLF histopathologically [21, 22].

Outcome: The outcome after surgery depends on the duration of the symptoms, the anteroposterior diameter of the lesion, the degree of cord compression observed, the presence of myelopathy, and the specific type of OLF discovered. Our review has shown that surgical intervention for a cervical OLF is associated with a better prognosis when compared to thoracic subtypes.

4. Conclusion

COLF, although rare within non-East Asian populations, should be considered a possible cause of cervical myeloradiculopathy. Surgical excision of COLF is quite easier in the cervical spine with less adhesion and dural tear compared with the thoracic and lumbar OLFs, probably due to the rarity of concomitant dural ossification. Detection of this illusive condition requires a careful and concise neurological exam, the utilization of specific imaging features, a high level of adequate suspicion, and a solid working knowledge of the unique pathological features of this clinical presentation. As a whole, this rare phenomenon is markedly different from a simple calcification of the yellow ligament itself.

Ethical Considerations

Compliance with ethical guidelines

Written informed consent was obtained from the patient for publishing this report and the corresponding images.

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Authors' contributions

All authors contributed equally to preparing this report.

Conflict of interest

The authors declare no conflict of interest.

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