Research Article



Journal of Spine & Neurosurgery

A SCITECHNOL JOURNAL

Ossification of the Ligamentum Flavum of the Lumbar Spine in Caucasians: Case Series

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Abstract

Ossification of the ligamentum flavum or OLF is a disease with heterotopic ossification in this spinal ligament. The key of OLF pathogenesis is the differentiation of fibroblasts into osteoblasts.

The majority of cases of ossification of the yellow ligament occur at the lower third of the thoracic or the thoracolumbar spine. However, the literature contains only few reports of patients with OLF of the lumbar spine. The clinical presentation of lumbar OLF is radiculopathy or intermittent neurogenic claudication, although some might remain asymptomatic.

Herein, we present 8 Caucasians with symptomatic lumbar OLf. The age and gender as well as the clinical picture, characteristic radiological features, the number of the affected levels will be described. The tips and tricks for achievement of a successful decompressive laminectomy, as the treatment of choice, done at one level in three and at two levels in five cases will be described. Furthermore, the degree of adherence of the lesions to dura and ultimate outcome will be discussed.

Keywords

Canal stenosis; Lumbar spine; Ossification of ligamentum flavum; Ossification of yellow ligament

Introduction

Ossification of the ligamentum flavum (OLF) was first reported in the 1920s by Polgar in lateral radiographs. This was further described by Yamaguchi and Isuruni as thickened or ossified ligamentum flavum this pathology is now appreciated as an important cause of thoracic myeloradiculopathy in Japanese population [1-4]. With regard to prevalence, this ligamentous disease is reported to occur in up 20% of the people older than 65 years among Japanese and many reports viewed it as a normal feature of aging in this ethnic group [2-4]. For more than 3 to 4 decades, it was believed that ossification of the ligamentum flavum occurs exclusively in Japanese people. However, this belief was ruled out, once the disease was found increasingly in other ethnic groups, in particular in Chinese and Koreans in the past few years [5-8].

Received: October 26, 2017 Accepted: November 08, 2017 Published: November 13, 2017



In a prospective study done by Guo et al. in 2009, of total of 1736 Chinese volunteers for whom whole spine MRI had been done, the prevalence of OLF was 3.8% [5]. Surprisingly, in another study done by Lang et al the prevalence rate of thoracic OLF in 993 Chinese who had done chest spiral C.T scan reached to 63.9%.

Other than the East Asian people, there have been some scattered case reports among whites, Indians, Africans, Arabs and Caucasians [9-17]. However, there have been no case series reports among Iranian population.

Most of the OLFs occurs at the thoracic spine mainly at lower thoracic levels, but lumbar affection is regarded a rare entity [2-8]. In the survey of Guo et el. evaluating whole spine MRI of 1736 outpatients cases, only one subject with lumbar OLF with prevalence of 0/1% could be found [5]. In 1988, Kurihara et al. analyzed 2403 plain lumbar roentgenograms and found a prevalence rate of 8.4% for lumbar spine OLF, with frequent involvement of the upper and middle lumbar spine [18]. However, it should be noted that most of the patients harboring ossification of yellow ligaments were asymptomatic.

However, lumbar OLF as a distinct paper was first reported by Weiss and Spencer in 1970. Since then only a few papers with detailed information have been published so far. Surprisingly, majority of the cases with lumbar OLF were among east asian population. Furthermore, although with increased use of new imaging modalities, it was expected that more reports among Non-East Asian people emerge, but the number of the cases with the symptomatic lumbar OLF did not increase in the literature. Therefore, published papers about lumbar OLF remained scarce and confined to a few case reports.

Herein, a retrospective survey of 8 Iranian patients with lumbar OLF will be reported. Age, sex, medical history, clinical picture, interesting imaging features and family history of ossified spinal lesions are noted. Surgical treatment of choice and ultimate outcome after surgery will be described.

Material and Methods

We retrospectively reviewed the charts, imaging studies, and pathological reports obtained in 8 Iranian patients with symptomatic lumbar canal stenosis secondary to OLF treated at our hospital. There were four males and four female patients. The mean age at presentation was 61.75 years (range 37 to79 years). The clinical picture in three cases was lower limb radiculopathy and in the remaining five was neurogenic claudication. Duration of the symptoms from the onset before initial consultation to surgery ranged from 4 to 32 months with an average of 10.8 months. Sphincter dysfunction was not seen in any of these patients. Obesity was observed only in one patient and diabetes mellitus was not found in any. But, no other concomitant systemic diseases such as, flurosis, hemochromatosis and calcium metabolism disorders were found in the patients of this series. No family history of lumbar canal stenosis or lumbar laminectomy could be found in close relatives of the patients.

Radiological assessment included plain radiographs, standing dynamic lateral radiographs, magnetic resonance imaging (MRI) and axial and reconstructed computed tomography (CT) scan which were done in all cases.

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doi: 10.4172/2325-9701.1000283

Whole-spine MRI was also performed in all cases for exclusion of tandem involvement.

The upper lumbar region was the common site of involvement by the disease. The pathology was isolated in four and multiple in four all continuous. In all cases, the lesions were bilateral, affecting one level in three, double-level in five (Figures 1-5).

However, radiographic features of ankylosing spondylitis and diffuse idiopathic skeletal hyperostosis (DISH) were not detected in the present study.

Surgical intervention

All patients underwent decompressive laminectomy. In total, 13 segments were decompressed. Surgery was started with a midline incision over the involved levels, with subsequent resection of the spinous processes followed by laminectomy. Laminectomy was

done with the aid of high speed burr being started laterally near the medial facet joints with making a groove till dura could be exposed bilaterally. Drilling with burr was continued till the remaining lamina became paper thin. Thereafter, OLF dissection from the dura was done microscopically in seven and macroscopically in one. Adherence of OLFs to the dura was mild or moderate in 7, where in one was tight and severe.

Result

All patients showed good recovery except one who was readmitted because of low back pain in straining and coughing. In this case, further evaluation showed pseudomeningocele formation which was cured after a redo surgery.

The patients were followed from two to 82 months and no case with signs of recurrence of OLF could be detected. With classification



doi: 10.4172/2325-9701.1000283



sagittal reconstructed CT showing a rather large OLF, (c) a few months after surgery showing a pseudomeningocele at L2-L3 level.

doi: 10.4172/2325-9701.1000283



Figure 3: 66-year-old woman with intermittent claudication for about a year. (a) T2-weighted sagittal MRI at L3-L4 and L2-L3 levels.(b) reconstructed sagittal CT scan demonstrating ossification at the site of normal yellow ligaments,(c) Axial view at L3-L4 level.



Figure 4: 62-year-old man with radiculopathy. (T2-weighted sagittal MRI showing hypointense mass compressing the theca at L2-L3 level, (b& c) axial and reformatted CT scan demonstrate an ossification at the site of ligamentum flavum.,(d) intraoperative photograph shows the patch of ossified yellow ligament after its removal.

doi: 10.4172/2325-9701.1000283



Figure 5: 79-year old man with neurogenic claudication for about a year,(a) T1 weighted sagittal MRI showing isointense mass at the site of yellow ligament at the the posterior aspect at L2-L3 level,(b) T2-weighted MRI wher the mass is hypointense compatible with OLF. (c) Axial CT showing bilateral type OLF, (d) reconstructed sagittal images showing a large OLF at the posterior aspect of canal.

doi: 10.4172/2325-9701.1000283

No	Sex	Age	Clinical Picture	Affected Levels	Treatment	Outcome
1	М	67	Intermittent claudication	L2-L3+L3-L4	Laminectomy	excellent
2	F	37	Radiculopathy	L2-L3	Laminectomy	Good
3	F	66	Intermittent claudication	L2-L3-L3-L4	Laminectomy + Screw rod fixation	Good
4	М	62	Radiculopathy	L2-L3	Laminectomy	excellent
5	М	79	Intermittent claudication	L2-L3	Laminectomy	excellent
6	F	64	Intermittent claudication	L1-L2+L2-L3	Laminectomy	excellent
7	F	58	Radiculopathy	L2-L3	Laminectomy	Excellent
8	М	61	Intermittent claudication	L2-L3+ L3-L4	Laminectomy	Excellent

Table 1: showing the age, sex, clinical picture, the number of the affected levels, treatment and ultimate outcome of 8 patients.

of outcome to excellent, good, fair and poor, 6 patients were excellent and 2 were good. The distribution of lumbar OLF by age and gender and the level and the number of the affected levels are shown in Table 1.

Discussion

Ligamentum flavum is a well-defined elastic structure. About 80% of this ligament is composed of elastic fibers and 20% of collagen fibers. The function of this ligament is preserving intrinsic stability of spine.

Pathology

Ossified ligamentum flavum (OLF) is characterized by ectopic bone formation in the ligamentum flavum which is normally composed of fibrous tissue. Development of OLF begins with hypertrophy of this ligament (LF) gradual loss of elastic fibers, differentiation of the fibroblast into chondrocytes followed by transformation of the chondrocytes into osteoblasts.

Ligamentum flavum ossification, calcification and calcium pyrophosphate dehydrate crystal deposition disease (CPPDcdd) which are quite different entities, sometimes are confused due to lack of inadequate histological examination. However, it is now proved that calcification of the ligamentum flavum and CPPDcdd belong to a single category. Initially, basic changes in these two entities are similar and begin with retrograde degeneration. But, later their difference will become apparent with respect to the corresponding histologic findings.

Ossification initially develops in the cephalic and the caudal attachments of the ligamentum flavum to lamina, at the junction of the bone and ligament (enthesis) and is actually described as endochondral ossification or enthesopathy. Characteristic features of endochondral ossification are demonstration of the foci of wellformed woven bone, lamellar bone structure and bone marrow formation being surrounded by chondroid cells.. These findings indicate that factors other than aging are considered to be involved in formation, development and progress of the ossification. In calcification, CPPD crystals are deposited within degenerated ligamentous fibers calcified granules

Eventually, mature bone is not demonstrated inside the degenerated ligament.

However, calcification is associated with increased CPPD crystal deposition being demonstrated as calcified granules inside the degenerated ligament.

Ossification of the yellow ligament mostly occurs at the lower third of the thoracic or the thoracolumbar spine, from T9 to T12 [2-8]. However, only a few authors have published surgically treated OLF of the lumbar spine with detailed clinical and radiological features [18-

25]. Most of the cases with lumbar OLF have been observed in east Asian people, with only a few case reports from non-East Asian areas, from North America, Greece and Turkey have been published to date [19,21,23,24]. Therefore, this is the first lumbar OLF case series from non-east Asian countries and the first case series in Caucasian if we accept that Iranians are dissident of pure Aryans.

Pathogenesis

Despite numerous basic science studies, the pathogenesis of OLF has not been conclusively established. However, several investigators have described the possible contribution of repeated mechanical stress, metabolic diseases such as diabetes mellitus, hyperinsulism, hemochromatosis, calcium metabolism abnormalities, genetic and cell biological factors it has also been reported that patients with OLF have a higher frequency in the patients with obesity and in the cases with higher bone mineral density [26-35]. Besides these factors, dietary habits may constitute an independent risk factor for OLF formation. Moreover, it is known that the incidence of OLF is higher in patients with diffuse idiopathic skeletal hyperostosis, fluorosis, and ankylosing spondylitis [36].

In contrast to thoracic OLF which is reported to involve patients predominantly in their 60s and 70s, the reported lumbar OLF has been found to occur in younger people with mean of 53, 25 unpublished case reports. The mean age of the reported cases is less than our series with 61,75. However, review of the published papers about LOLF shows almost equally affection in males and females correlating with our case series [18-24].

Clinical feature

The slowly progressive course of lumbar OLF makes its early diagnosis difficult. Nonetheless, the symptoms in patients with OLF in the lumbar spine are similar to the cases with lateral recess syndrome or degenerative lumbar canal stenosis where it can be presented with radiculopathy or intermittent claudication respectively. Because of the insidious behavior of lumbar OLF, it is not unusual to depict the pathology once the disease has progressed with more sever root damage or cauda equina syndrome. Our patients were diagnosed in a rather early course of the disease, because of easy and direct accessibility of the Iranian patients to neurosurgeons, the low price for MRI performance and increased number of MRI centers both in private and governmental centers,

Association with diabetes insidious, or calcium metabolism should be carefully evaluated.

Imaging

A good quality AP and lateral lumbar plain radiographs might show OLF as a small ossified lesion between the bases of two spinal processes. Lateral dynamic lumbar X-ray is necessary to rule out instability.

doi: 10.4172/2325-9701.1000283

However, the exact location, the number of the affected levels and the degree of cord or cauda equina compression can be best assessed with magnetic resonance imaging (MRI) [37,38]. A typical OLF is visualized in MRI as a hypointense signal intensity mass at the level of the posterior margin of the spinal canal, both in T1 and T2-weighted images. Besides, showing the degree of canal compromise [37-40]. In order to detect coexisting or tandem OLF at other locations, whole spine MRI is quite necessary. With the aid of MRI, ossification of the yellow ligaments has been classified with respect to the number into two types; isolated and multiple types. With regard to the continuity of the lesions, multiple type itself is divided into continuous and noncontinuous sub-types.

For detection of the shape and the extent of ossified lesions, computerized tomography is the diagnostic tool of choice. On axial CT scans, the lesions are seen as ossified mass from the facet joint to the base of the spinal process either unilaterally or bilaterally and in sagittal reconstructed images, OLF is seen as an ossified mass at the posterior aspect of the spinal column at the site of ligamentum flavum besides, the shape, contour and the extent of OLF, the type of the pathology can be obtained by this diagnostic tool [37-40].

With respect to CT images, OLF is classified into five types: The lateral type, Type I, is located only laterally at the origin of the ligamentum flavum at the articular processes. The extended type, Type II, extends from the lateral origin of the ligamentum flavum to the interlaminar portion of the ligamentum flavum. The enlarged type, Type III, protrudes into the canal posterolaterally but is not fused in the midline. The fused type or type IV consists of bilateral ossified ligaments that are fused at the midline with a groove at the fusion site in the midline. 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.

Another categorization is based on sagittal configuration of OLF into round and beak type, being demonstrated both in sagittal MRI and reconstructed CT scans, according to Kuh et al. in 2006.

Associated OPLL can also be suggested in sagittal MRI, but can be confirmed with computerized tomography. Unlike the patients with OLF in the thoracic spine with a high incidence of ossification of the other spinal ligaments, such as ossification of the posterior longitudinal ligament (OPLL), ossification of this ligament remains an infrequent event in the, the OLF of the lumbar.

Treatment

Where asymptomatic cases can be closely followed, decompressive laminectomy and excision of the OLF are the main stay of surgical procedure [41-43]. The indications for surgery are cauda equina syndrome, radiculopathy or progressive neurological impairment. This can be done after induction of general anesthesia with the patient in the prone position. After exposure, intraoperative fluoroscopy should be used for correct localization of the affected levels.

The spinal process can be removed with rongeur. Subsequently, laminectomy with high speed burr should be done. Laminectomy is better to be started from the lateral side of lamina with making a longitudinal groove on both sides. The grooves should be deepened till the lateral dura mater can be identified. Laminectomy with burr should be continued till the laminas become paper thin. After piece meal removal of sound lamina, a patch of thinned lamina and OLF remains on either side of the midline. These patches should be lifted, dissected and detached from the dura with a sharp dissector. At this moment, great care is necessary not to tear the dura [41-43]. However, in some instances, dural tearing is inevitable and in infrequent situation the dura might be absent at the site of OLF. However, the incidence of dural tears and CSF leakage in OLF patients is relatively frequent and has been estimated to be from 10% to 32%. The dural tears should be closed with continuous silk and for dural defects; duraplasty with a range of materials is required, usually with placement of fascia or artificial dura. Once, tight closure was accomplished, Valsalva maneuver is necessary to confirm the integrity of the repair. Subsequently, the wound should be closed tightly in 3 layers.

Post-operatively, therapeutic lumbar puncture or short term lumbar cerebrospinal fluid drainage are recommended. Leaving the small tears unrepaired may result in continued CSF leakage pseudomeningocele formation and even meningitis [44].

Laminoplasty as an alternative option has been used in a few patients by Kawaguchi et al. in 2005, with acceptable results [22]. Laminoplasty should be done only in the patients with lumbar lordosis.

Outcome

The outcome after surgery depends on the duration of the symptoms, degree of anteroposterior diameter of the lesion and type of OLF.

As has been documented, longer the duration of the symptoms, tuberous shape OLF with marked protrusion into the spinal canal and fused type of OLF are associated with poor prognosis [45-49].

However, unlike the thoracic spine, lumbar OLFs have better prognosis. The difference in outcome between lumbar and thoracic OLF can be explained with higher resistance of the cauda equina and the roots to gradual compression, their less susceptibility to intraoperative forces derived during laminectomy, more rapid response of the cauda to decompression, higher resistance of the cauda equina to perioprative fluctuation of the blood pressure, and finally more familiarity of the neurosurgeons to lumbar laminectomy

Conclusion

Lumbar OLF (LOLF) remains one of the rare causes of intermittent claudication or lower extremity radiculopathy. It seems that OLF is under reported in Iran, despite lack of a case series among the Iranian population.

The present case series is the first retrospective study in Iranians with ossified ligamentum flavum. Our survey showed that the clinical and radiological features of lumbar OLF in the Iranian population appear to be similar to other ethnic societies. Early diagnosis with shorter duration of the symptoms probably due to easy availability of MRI have been the cause of relatively good outcome in Iranian patients.

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doi: 10.4172/2325-9701.1000283

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