Review Article

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Cervical Ossified Ligamentum Flavum Associated With Cervical OPLL Presenting as Compressive Myelopathy : Case Report and Review of Literature

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Abstract

Ossified yellow ligament (OYL) is a disabling cause of extradural compressive radiculo myelopathy, more common in thoracic region and rare in cervical region. Here is a case report of cervical ossified yellow ligament associated with cervical ossified posterior longitudinal ligament from C3 to C7 region causing myelopathy symptoms. Cervical laminectomy from C3 to C7 and excision of OYL flap from the cal sac by blunt and sharp dissection was done .patient's neurological status improved following surgery.This article focuses the metabolic and genetic predisposition and pathogenesis of ossification of ligamentum flavum which is more common in the thoracic region and rare in cervical region.

Keywords: Ossified Yellow Ligament; Cervical Region ; Decompressive Laminectomy; Pathogenesis.

Introduction

Hypertrophy and ossification are well known disorders of the ligamentum flavum of the thoracic and lumbar vertebrae. Polgar in the year 1920 first reported ossified ligamentum flavum or yellow ligament and was later further described by Yamaguchi and Isuruni [1]. Of all documented cases in the literature,the large majority are Japanese subjects (88.8%), with Caucasian patients being the next most prevalent(8.2%) [2]. Reports have also been documented in Afro-american& asian population[3]. Though no major case series has been reported in the literature, Ossified yellow ligament (OYL) is very much prevalent in this part of the world. The author's institution operates on average 5 cases of OYL in a month. Majority of cases operated in the authors institution are located in the dorsal region as has been reported in the literature [4,5,6]. The authors in the present report, described a case of OYL in the cervical thoracic region with clinical characteristics, surgical technique for decompression and reviewed the pathophysiology involved in OYL formation.

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Case Report

A 35 years old farmer presented with neck pain, paraesthesias in all four limbs for 7 months followed by difficulty in walking for past 6 months. On examination patient had hypertonia in all four limbs with exaggerated reflexes. He had spastic gait with Japanese orthopedic association score of 12 [7]. MRI cervical spine has shown thickened Posterior longitudinal ligament from C2 to C7 and thickened Yellow ligament in the cervical spine from C2 to D3 causing narrowing of the cervical spinal canal from C2 toC7 with compression of the cervical spinal cord.(Fig-1). Computed tomography of the cervical spine was suggestive of OYL from C2 to C7 and OPLL in the C6 and C7 region with severe narrowing of spinal canal in the lower part of cervical spine (Fig.2). C2 to C7 laminectomy was done using high speed drill.

Drilling was done on either side just medial to the junction of the lamina and facets till the bone is thinned out. The laminar flap is elevated from C2 to C7 exposing the OYL adherent to the thecal sac from C2 to C7. The OYL is excised from the thecal sac from C2 to C7 as a single flap by dissecting it from the dura using blunt and sharp dissection(Fig.3,4). Following excision thecal sac expanded and good pulsations were noted.Postoperatively patient was relieved of spasticity and paraesthesias. His JOA scores at the time of discharge was 17/17.

Figure 1A: MRI saggittal section of cervical spine showing thickened ossified posterior longitudinal ligament and thickened ligamentum flavum with severe compression of the spinal cord. **Fig.B:** MRI axial sections showing thickened OPLL and OYL with severe compression of the cord. **Fig.C, D:** CT cervical spine sagittal and axial section showing ossified OPLL and OYL showing severe narrowing of the cervial canal.

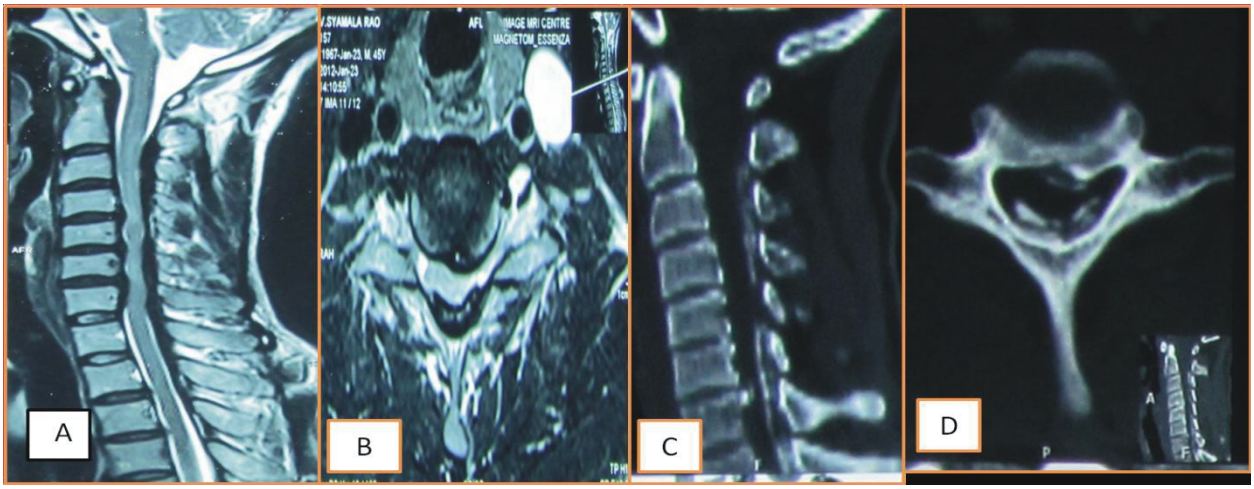
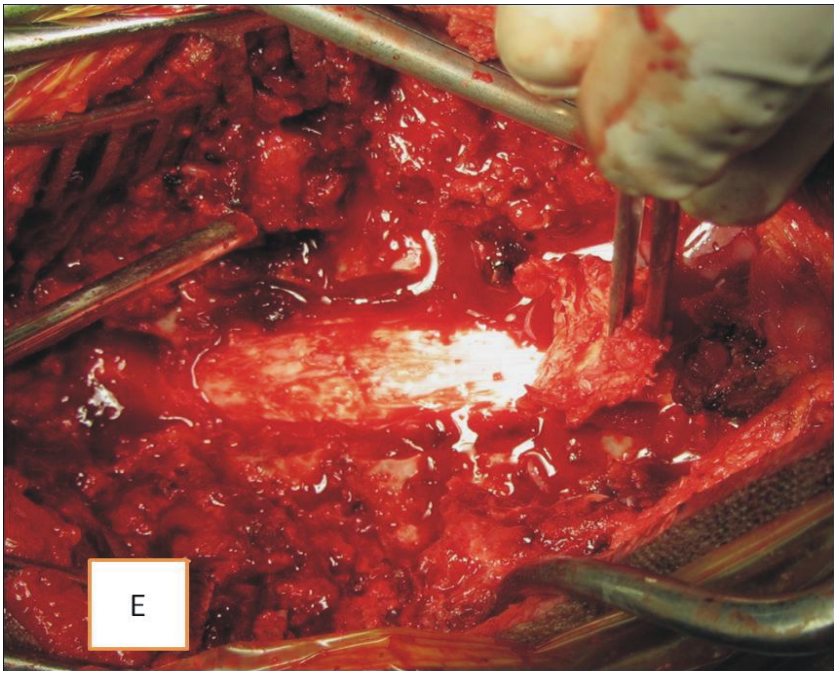


Figure.E: Intraoperative photograph showing OYL flap being stripped and elevated from the thecal sac.



Discussion

Ligamentum Flavum Ossification

The incidence of ossified ligamentum flavum is more in the thoracic region,the ossified segments are mainly located in the lower thoracic spine, rarely in the upper thoracic spine [5,6,8,9]. Cervical thickening of OYL due to calcification is common while the same due to ossification is rare with only few reported cases in the literature [10,11,12,13,14,15]. The OYL in thoracic spine is seen frequently but for unknown reasons the cervical OYL is rarely encountered. Probably cervical flexion and extension enables the yellow ligament to become lax frequently and thus maintain its elastic fibres [16]. Many of cervical OPLL cases are surprisingly associated with dorsal OYL but not with cervical OYL [17]. The mechanisms underlying this selective ossification of ligaments appear to be multifactorial [18,19,20,21] .

Ossification results from progressive replacement of the hypertrophied ligamentum flavum by lamellar bone through endochondral ossification of the vascularized fibrocartilaginous tissue starting at the

densely adherent ligamentous-osseous junction (enthesis), then extending along the ventral aspect of the ligament[2,21,22,23]. Microscopic examination of the ossified ligamentum flavum shows the presence of endachondral ossification, showing the presence of mature lamellar bone with well developed haversian system and the ligamentum replaced by proliferating cartilage[21,22]. Calcification of the ligamentum flavum(CLF) is another rare entity which characteristically occurs in the cervical spine of older women contrasts from ossification which is more common in the lower thoracic of the younger individuals with no sex prevalence. In CLF,calcified granules are characteristically deposited within the degenerative ligamentum fibers,with no mature bone formed within the ligament ,which is the main difference between CLF and OLF[11,25] . On imaging ,CLF will have nodular or diffuse calcification. Calcification will be discontinuous with the lamina while ossification would be continuous pattern, whereas OYL will have nodular or V-shaped ossification[11,25]. On MRI, CLF is hypointense on T1 and T2 weighted images. Same will be the imaging findings of OYL,but is seen at the intervertebral level[11].

The cause of OYL remains unclear, despite its identification a century before. There is an association with hyperostotic syndromes including OPLL, DISH, and ankylosing spondylosis. OPLL is generally associated with ossified ligamentum flavum, rare with CLF[17, 22]. it has also been associated with metabolic derangements including diabetes mellitus, hyperinsulinism, impaired glucose tolerance, obesity, Paget disease, hemochromatosis, fluorosis, X-linked hypophosphatemia, and hypoparathyroidism. [2,11,20] Additionally, mechanical stress, trauma have been suggested as contributing factors associated with OLF [11, 18, 26].

The major diagnostic difference between OYL and OPLL is that in the former , the histopathological calcification allows the ligamentum flavum to fuse with adjacent lamina and extend form the ventral aspect of the upper lamina to the dorsum of the lower lamina , whereas for the latter, the posterior longitudinal ligament joins the posterior aspects of the vertebral bodies and the intervertebral discs to extend from the body of the axis to the sacral canal. But similarities exist between OYL and OPLL at the molecular and possibly the genetic level[2,11,20]. Ono et al identified that growth factors-bone morphogenic protein-2(BMP-2), and transforming growth factor-b(TGF-b) played an important role in the matrix hyperplasia and ossification of the spinal ligament of both OYL and OPLL[22]. BMP-2 and TGF-b were demonstrated to stimulate progenitor cells within the ligament, causing them to proliferate,form cartilage and ossify. BMP-2 initiates new cartilage and bone formation whereas TGF-b stimulates chondroprogenitor and osteoprogenitor cells to form cartilage and bone [2,19,27,28].

Very recently ,linkage and linkage disequilibrium analyses identifies genetic relation in patients of OPLL and OYL with genes like COL6A1 and COL11A2 have been identified.[27,29,30]. Most of the cases of ligamental ossification, the authors encounter at their institution, come from flurosis affected areas and areas where coal and other mineral deposits are mined. But the patient under study comes from Krishna river belt implies the fact that more than environmental factors strong genetic factors must be resulting in such pathologies. GUO Jiong-jiong, YANG Hui-lin, et al designed two methods of classification of tandem ossification . The first classification divided the OPLL and OFL based on distributional patterns.

The other classification divided OPLL and OFL into concurrent, nonconcurrent, and mixed types, based on the pattern of whether OPLL and OFL were at the same levels[17]. Muthukumar et al classified cervical OYL into 5 types, based on extent / location of the ossification[31). 93 case series by GUO Jiong-jiong etal doesn't include single case of cervical OYL implying rarity. [17]

Management

Once myelopathy develops, the only treatment is surgical decompression[26,32]. The surgical technique is described by the authors allows good surgical decompression with minimal manipulation or insult to the cord resulting in good outcomes. Similar technique has been described by Lana D.Christiano previously [11].

Conclusion

Cervical OYL is a rare entity. It is associated with ossification of ligaments in other regions of spine. The genetic factors are likely to have greater role in addition to the environmental and metabolic factors as exemplified by the case described by the authors. The surgical technique described by the authors can be used for effective decompression of spinal cord in these cases.

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