

# Ossification Of The Posterior Longitudinal Ligament: A Case Report

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## Abstract

Ossification of posterior longitudinal ligament (OPLL) has recently been recognized as a clinical entity. It is a rare condition with a higher incidence in the Japanese population. It is characterized by hyperplasia of cartilage cells with eventual endochondral ossification of the posterior longitudinal ligament. The radiographic signs are characteristic and consist of a linear band of ossification tissue along the posterior margin of the vertebral bodies. OPLL can be associated with mild to serious neurological complication due to spinal cord or nerve root compression or it may be asymptomatic. This paper reviews the radiological, clinical and surgical aspects of this condition.

## INTRODUCTION

Ossification of the posterior longitudinal ligament (OPLL) of the cervical spine has only recently been recognized as a clinical entity. Although it was reported as early as 1839, it was not until 1960 that its association with chronic cervical myelopathy was noted. It was termed OPLL by Terayama in 1964 (5). Since then, numerous cases have been described in the literature, largely, though not exclusively, from Japan. The intention of this case report is to introduce an Iranian patient diagnosed by OPLL and discuss the etiology, pathology, diagnosis and treatment of OPLL.

The etiologies of OPLL remain obscure. Proposed mechanisms include infection, previous trauma, fluoride intoxication, diabetes mellitus, genetic transfer, and an immunologic disorder related to HLA antigens (3-5). Although no single cause has been accepted to date, the predominance of OPLL among the Japanese suggests that it may be the result of hereditary or geographic factors. This higher incidence of OPLL in the Japanese population is reported to be in order of two to three percent (1,2,5). This incidence increases significantly to approximately 30% among family members of second-order relatives of the affected patient.

OPLL has also been seen in up to 50% of patients with diffuse idiopathic skeletal hyperostosis (DISH), raising the question of generalized hyperostosis tendency in these individuals. Of those presenting with OPLL of the cervical spine, the majority tend to be between 50 and 60 years of

age with a male to female ratio of about three to one (1,3,4).

OPLL occurs by the process of endochondral bone formation, beginning with the most superficial layer. The histological findings in surgical specimens are indistinguishable from those of heterotypic bone formation seen in spondylosis or in response to mechanical stress and trauma in other ligaments. Fibroblast hyperplasia and increased collagen deposition are followed by mineralization of the thickened ligament. Cartilage cells and bone matrix proliferate in the vertebral body periosteum, annulus fibrosus and in some cases in dura mater as well as in the PLL. As the ossified mass enlarges and extends longitudinally, lamellar bone eventually matures with well developed Haversian canals and few marrow cavities. This may explain the neurological deficits commonly seen in patient with OPLL (1,3,4).

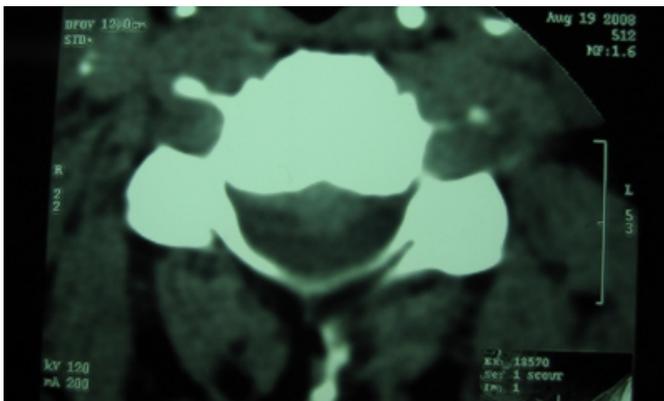
## CASE REPORT

A 54-year-old Iranian female, a known case of diabetes mellitus, presented with a complaint of disability to work, quadriparesia and neck pain since four months ago. Paresia was dominant in the upper extremities especially distal parts. The pain was exacerbated by forward flexion of the cervical spine. There was a history of mid-thoracic laminectomy 5 years ago due to thoracic canal stenosis. On examination, the active range of movement of the cervical spine was painfully decreased by approximately 10 percent of normal. Neurological examination revealed muscle power of 4/5 in proximal and 2/5 in distal parts of the upper extremities.

There wasn't any urinary or fecal incontinency. Radiography of the cervical spine showed an osseous extending from the posterior margins of C2 to thoracic vertebral bodies. Sagittal T2 weighted MR image revealed a hypo-intense signal posterior to vertebral body margin suggestive of ossified posterior longitudinal ligament (Fig. 1). Axial CT image of the cervical spine showed OPLL at C2 to upper thoracic level (Fig. 2). Sagittal CT and MRI image showed canal stenosis , myelopathy and OPLL (Fig. 3).

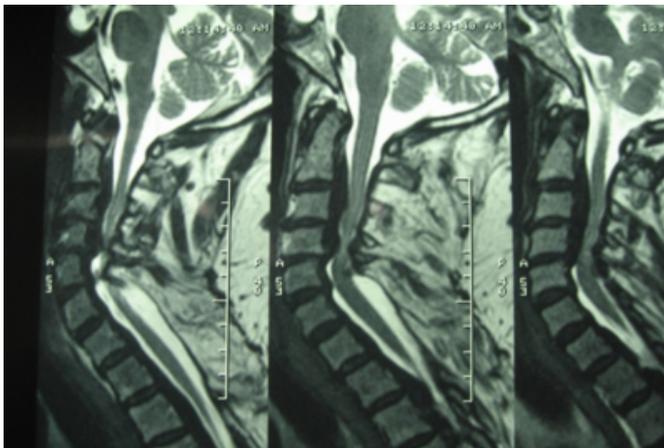
**Figure 1**

Fig.1: Sagittal T2-weighted MR image revealed a hypo intense signal posterior to vertebral body margin



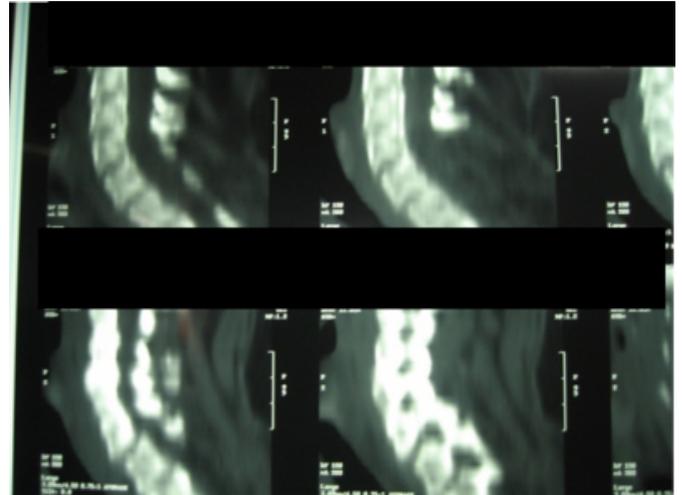
**Figure 2**

Fig. 2. Axial CT image of cervical spine shows ossified posterior longitudinal ligament



**Figure 3**

Fig.3. Sagittal CT image shows canal stenosis and ossification of posterior longitudinal ligament



## DISCUSSION

The clinical syndromes of OPLL consist of slowly progressing compressive myelopathies with or without radiculopathy. They are localized neurologically to the levels of spinal involvement, unless both cervical and thoracic lesions are present. Symptoms described in addition to neck pain (42%) and arm pain or dysesthesia (48%) include arm weakness (19%), leg weakness (15%), urinary incontinence (10%), arm and leg clumsiness or weakness, arm numbness or paresthesia, and gait difficulties each occurred in at least 80% of patients. Limited cervical spinal motion, hyperreflexia, extensor plantar response, and arm or leg weakness were found in 65% of patients in the literature. Hypalgesia below segmental levels on the torso were present in 60% and radicular sensory loss was found in 55%. The treatment of spinal stenosis due to OPLL is based on standard principles of managing neurospinal diseases. Acute episodes of pain and the onset or in-mase of neurological deficit are treated with rest, external spinal immobilization with a collar or brace, and analgesics, antiinflammatory or antispasmodic medications when indicated (1,3,4).

Acute cervical or thoracic spinal cord contusion can be treated with high-dose steroids. Surgery is indicated for patients with acute or chronic progression of neurological deficits or recurrent temporary disability from reversible neurological deficits including impairment of manual dexterity and leg coordination (4,5).

The choice of operation depends on the anatomy of the lesion causing spinal cord and nerve root compression. In most cases of cervical and thoracic myelopathy due to

OPLL, anterior decompression and arthrodesis are indicated. Only in the rare cases of focal segmental lesions that do not extend above or below the disk, does a simple interbody discectomy and fusion suffice (2,5,6).

### IMAGING DISCUSSION

Morphologically, four forms of OPLL have been described. The continuous and segmental forms account for up to 95% of all cases. The continuous form extends over several contiguous vertebral bodies while the segmental form is situated fragmentally behind each vertebra. The mixed form includes the combination of continuous and segmental forms. The fourth or retrodiscal form is situated exclusively behind the intervertebral disc and radiologically may be indistinguishable from a posterior osteophyte.

CT scan is the method of choice for detecting the presence and extent of ossified mass (Fig. 3) (1,3-5).

Sagittal reformations are invaluable in showing the exact extent of the disease. On axial sections, OPLL is seen as a 'mushroom', a 'hill', and a 'square' or a mixture of these shapes and a characteristic sharp radiolucent line separates OPLL from the posterior vertebral margin representing the unossified deep component of PLL (5,6).

MR imaging is helpful in depicting the nature of cord compression such as myelomalacia, edema, demyelization or cyst formation and root sleeve involvement. The extent of myelomalacia and edema of the cervical cord is best seen in sagittal and axial T2 weighted sequences. OPLL is seen as a constant signal void in both T1- and T2- weighted images (1,3,4).

### TREATMENT

Surgical treatment by anterior approach, C3-C6 corpectomy, removal of ossified PLL, and canal decompression was done. Then strut graft harvested from iliac crest was replaced to the bodies of vertebrae C3-C6 and then fixed with anterior cervical plate (Fig. 4). The Patient had no new deficit postoperatively and gradually showed improvement with respect to motor function within one month and almost achieved normal daily activities.

### Figure 4

Fig.4. Postoperative cervical x-ray shows C3-C6 corpectomy, graft replacement and anterior cervical plate fixation.



### CONCLUSION

Ossification of posterior longitudinal ligament is a relatively rare finding, but it is important to recognize it as a cause for cervical canal stenosis. It may result in compression of the spinal cord or nerve root, and can cause permanent neurological damage. The radiographic sign of PLL is diagnostic and can consist of a linear band of ossified tissue along the posterior margin of the vertebral bodies.

A thorough history and examination will determine the plan of management, which can vary from immobilization to rest and possibly surgical decompression. Careful conservative management of these cases may be beneficial in relieving pain of myofascial and joint origin. Early surgical consultation however is emphasized when symptoms progress.

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