

Review Article

Asymptomatic Ossification Of The Posterior Longitudinal Ligament, Case Report And Review.

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Abstract

Ossification of the posterior longitudinal ligament (OPLL) is a pathological condition characterized by the abnormal calcification or bone formation within the posterior longitudinal ligament of the spine, most commonly in the cervical region. This process can lead to progressive spinal canal narrowing, resulting in spinal cord compression and associated neurological deficits such as myelopathy or radiculopathy. Although the exact etiology remains unclear, genetic predisposition, metabolic factors, and mechanical stress are considered contributing factors. OPLL is more prevalent in East Asian populations and is frequently associated with other spinal disorders like diffuse idiopathic skeletal hyperostosis (DISH). Diagnosis is typically confirmed via radiological imaging, including X-rays, CT scans, and MRI. Treatment options range from conservative management in asymptomatic patients to surgical decompression in cases with significant neurological compromise. We present a case of a 53-year-old caucasian woman with no relevant medical or surgical history. The patient presented with cervical pain, with several years of evolution, without other associated complaints. There was no history of direct or indirect trauma prior to the onset of symptoms. We perform cervical spine imaging studies such as x-rays and computed tomography (CT) that revealed extensive multilevel stenosis due to OPLL in the cervical and thoracic regions, C2 to T2, contributing this alteration to the rectification of cervical lordosis. Considering the absence of symptoms of myelopathy and/or radiculopathy, we have decided to maintain a conservative attitude. This review aims to summarize the current understanding of the pathogenesis, clinical features, diagnostic modalities, and management strategies of OPLL.

Keywords: spinal ossification disorders, spinal cord compression, myelopathy, spinal stenosis, CT, MRI, surgery.

INTRODUCTION

OPLL is a condition in which bone forms within the posterior longitudinal ligament (PLL) of the spine (heterotopic ossification), causing calcification and ossification of ligament tissue that is normally flexible.¹ From the Histological point of view, this process can involve both endochondral ossification (via chondrocyte proliferation, cartilage intermediate) and metaplastic ossification/fibroblast proliferation and vascular invasion in degenerated ligament.² The cervical spine is the most commonly, as in our case, affected region in OPLL, followed by the thoracic spine, and less often the lumbar spine.³ OPLL may occur in different patterns: continuous, segmental, mixed, or localized ossification on imaging.⁴ Prevalence is highest in East Asian populations (e.g. Japan: about 1.9% to 4.3%) but it is present worldwide, with lower rates in Europe and North America (about 0.1% and 1.7%).⁵ This pathology is more common in middle-aged to older adults, frequently seen in 5th–6th decades of life with slight male predominance has been observed.^{6,7}

OPLL pathology is considered multifactorial, involving genetic factors: with several genes have been implicated (e.g. BMPs bone morphogenetic proteins, collagen gene variants, transcription factors like RUNX2, etc). Also non-genetic factors have been related as age, mechanical stress or strain on the ligament, metabolic factors (e.g. glucose metabolism, obesity), dietary factors, obesity, diabetes mellitus. Inflammation (all related parameters in the inflammatory cascade) and calcium-phosphate metabolism are also related to this pathology.⁸ The OPLL is most commonly found in men, in the elderly, and in Asian patients. The disease can start with mild or no symptoms, but some patients progress slowly to develop symptoms of myelopathy. When OPLL progress may produce myelopathy (chronic compression of the spinal cord), radiculopathy (nerve root involvement), sensory and motor deficits, gait disturbances. In severe cases, sphincter dysfunction or even paralysis may occur.⁹ The OPLL Natural History often progresses slowly, but progression is variable among individuals. Some patients with radiographic OPLL never develop symptoms, others

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Received: 10-Mar-2026, Manuscript No. WNSR - 5480 ; **Editor Assigned:** 11-Mar-2026 ; **Reviewed:** 26-Mar-2026, QC No. WNSR - 5480 ; **Published:** 22-Apr-2026.

DOI: 10.52338/wnsr.2026.5480.

Citation: Gonçalo Januário. Asymptomatic Ossification Of The Posterior Longitudinal Ligament, Case Report And Review. World Neurosurgery Research. 2026 April; 16(1). doi: 10.52338/wnsr.2026.5480.

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present deteriorate over time or after minor trauma.⁹ During the diagnosis process the radiographs may show ossification, but may miss early or small lesions.¹⁰ The CT scan is superior to define the extent and type of ossification (pattern, bony vs calcified, involvement of dura).¹¹ The MRI permit to assess spinal cord compression, signal changes in the cord, and related soft tissue changes.^{12,13} The management of this pathology depend of the intensity of symptoms, in cases with mild to moderate symptoms without neurological involvement the nonoperative treatments may include observation, physiotherapy, management of comorbidities, but there is no proven disease-modifying pharmacotherapy yet.¹⁴ The surgical decompression is indicated when the symptoms are severe and particularly the neurological impairment is significant. Approaches can be anterior, posterior, or combined, depending on the type and extent of ossification, spinal alignment, patient condition.¹⁵

CLINICAL CASE

A 53-year-old female, no drug allergies or previous medical or surgical history. The patient presented with left cervicobrachialgia and paresthesia. Onset after intense exertion. Several weeks of evolution. Symptoms are mild and occur sporadically. During the physical examination, Rhomberg and tandem gait tests to identify early signs of gait or balance dysfunction was negative. Brisk reflexes normal, no clonus in upper and lower extremities. The patient don't presented pathologic reflexes such as Hoffman reflex and the inverted radial reflex suggest an upper motor neuron lesion. No dysdiadochokinesia or difficulty with rapid supination and pronation of the hand. The Spurling test was negative. Subsequently, radiography and the computed tomography (CT) study demonstrated OPLL extending from C2 to C7, with mild spinal canal compromise (**Figure 1,2,3**).

Figure 1. A lateral radiograph of the cervical spine shows the presence of ossification along the anterior and posterior aspects of the vertebrae.



Figure 2. sagittal CT slices , OPLL The ossified mass lies anterior to the K-line as demonstrated.

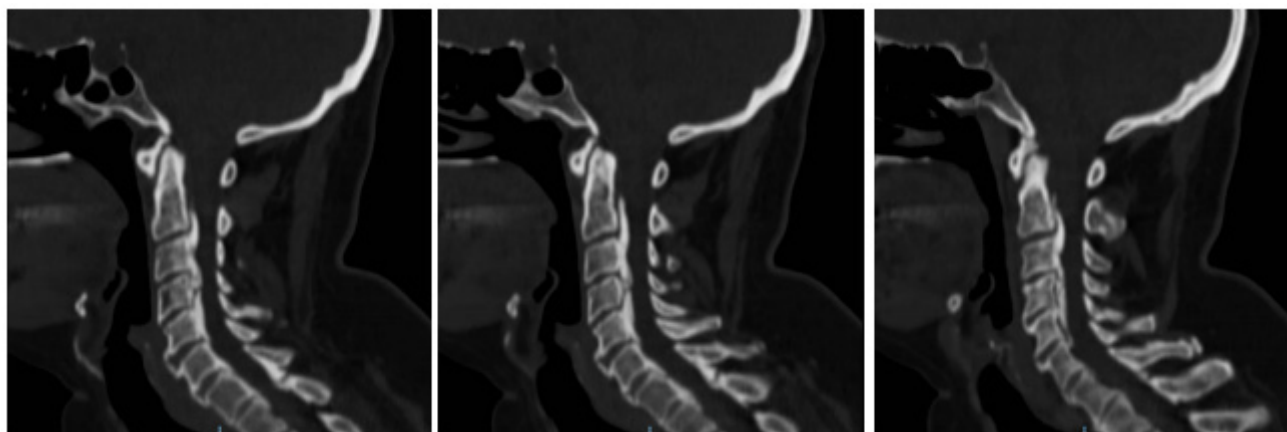
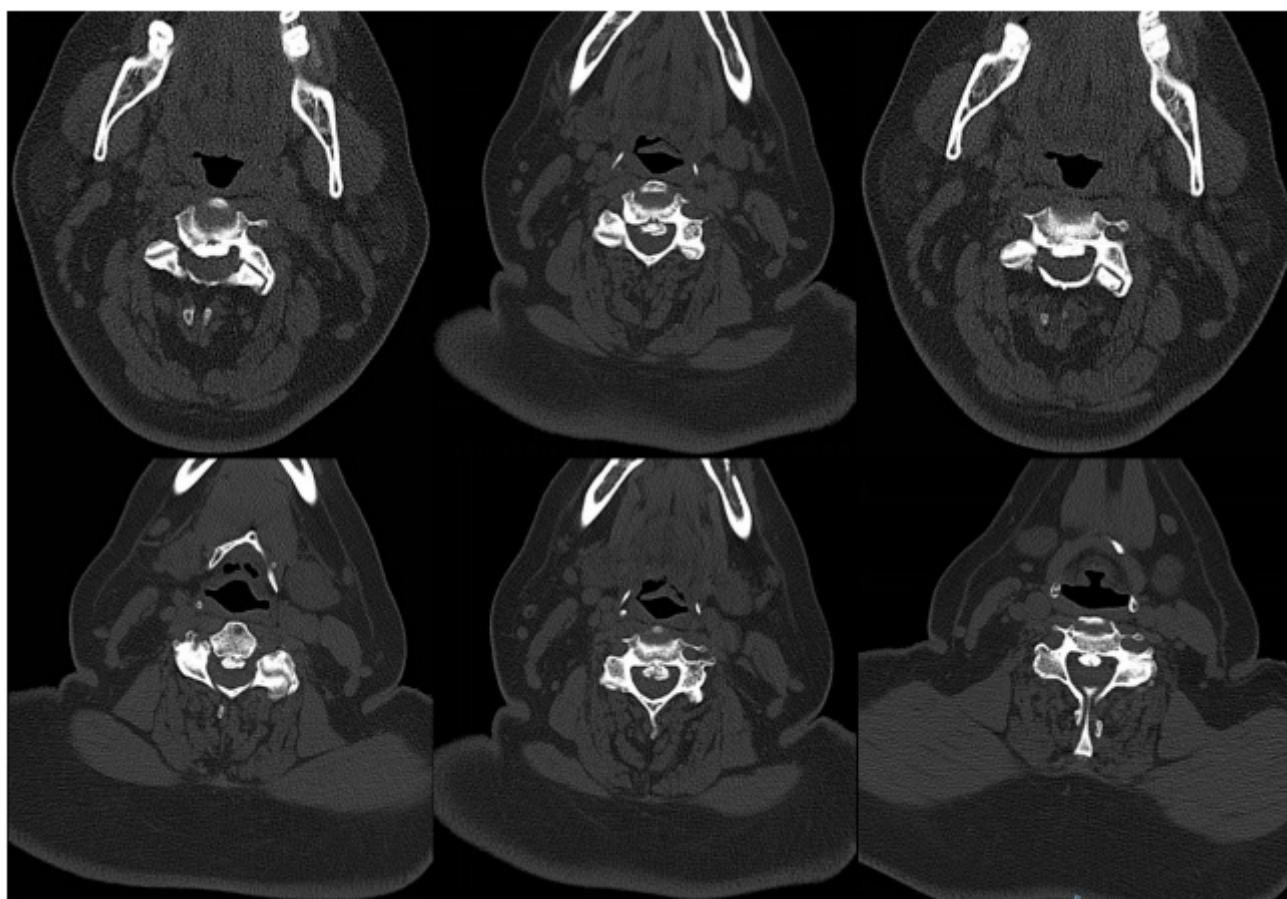




Figure 3. axial CT slices , showing ossified posterior longitudinal ligament decreasing the space available for the cord.



In the clinical context presented, we requested an electromyogram (EMG) of the upper extremities and somatosensory evoked potentials (SSEP), which were normal. An MRI was not performed because the patient refused the test. She is claustrophobic even in an open MRI machine and refused sedation for the procedure. The patient was sent for cervical spine rehabilitation treatment, showing a favorable evolution with residual cervical pain, without radiation to upper extremities or presence of paresthesias. Taking into account the symptomatic improvement with conservative treatments and their mild intensity, in addition to the fact that the results of the spinal cord and nerve root function test, this is a case for which there is currently no indication for surgery. However, it was made clear to the patient that, considering the natural evolution of the disease, it is likely that at some point in the future she will need surgical intervention. Our patient presents with continuous OPLL that needs monitoring the progression of the disease, follow-up appointments are scheduled every 3 months. A CT scan will be repeated, and an MRI will continue to be recommended. The repetition of the EMG and SSEP will be taken into account according to the presence or absence of radiculopathy or paresthesias.

DISCUSSION

OPLL is uncommon pathology in the world but occurs not infrequently in Japan where the prevalence of 2–4% has been reported in adults, compared with 0.01 to 2% in non-Asian populations.^{16,17} The prevalence of OPLL is higher in patients with myelopathy, 27% in Japan and 20 to 23% in the United States.¹⁸ This disease is occurring more frequently around 50–60, with a predilection for the cervical spine. Seventy percent of cases occur at C2–C4, 15% at T1–T4, and 15% at L1–L3.^{19, 20} OPLL commonly presents in the 5th–6th decades, and a male-to-female ratio of 2:1 has been reported.²¹ Ossification of the posterior longitudinal ligament of the cervical spine is a subtype of degenerative cervical myelopathy (DCM), characterized by pathological ligament ossification.²²

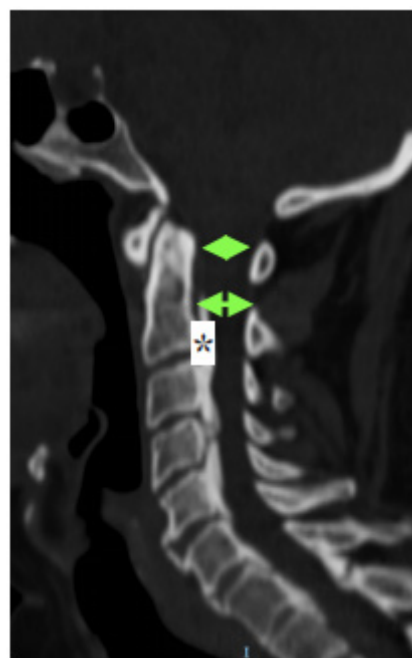
This pathology is related with the proliferation and differentiation of fibrocartilage-like cells and osteoblasts, along with the formation of new blood vessels within the ligament.²³ A genome-wide association study identified 6 susceptibility loci associated with OPLL in cervical spine, the genes *RSPH9* and *STK38L* are implicated in membranous ossification, and others as *HAO1*, *RSPO2*, *CCDC91* linked to endochondral ossification pathways.²⁴ For diagnosis OPLL it's recommended a careful history and physical examen. Although 5% of diagnosed patients are asymptomatic, varying degrees of neurologic symptoms can be present including radiculopathy and myelopathy or both.²⁵ A importante point is the rate of progression of symptoms in OPLL, linked to the age at the momento of the diagnosis or presentation. In fact the presentation in the 4th decade or younger is associated with progressively symptomatic OPLL and subsequent myelopathy. Presentation in the 5th–6th decades is less frequently associated with progression to myelopathy.^{26, 27, 28}

For the diagnosis the radiographs are helpful in the diagnosis of OPLL, particularly when located in the cervical region. However, low inter- and intraobserver reliability for diagnosing OPLL with radiographs has been demonstrated as compared with computed tomography (CT). CT scan sagittal sequences can help classify the type of OPLL and have been associated with higher intraobserver reliability than radiographs.²⁹ The axial slices define the lesion as central or paracentral, which can aid in the preoperative planning. As described in this study some groups have used three-dimensional CT to quantify the volume of OPLL as well as for classification purposes.³⁰ In cases of also dural ossification can be detected using a bone window on CT scans. Dural defects are more common in nonsegmental type of OPLL.³¹

The double-layer and single-layer signs on CT indicate OPLL penetration into the dura. The single-layer sign describes a mass of uniformly hyperdense OPLL, and the double-layer sign describes anterior and posterior rims of hyperdense regions separated by a hypodense area.³²

The double-layer sign is more predictive of a dural defect than the single-layer sign.³³ Magnetic resonance imaging (MRI) actually is useful in detecting myelomalacia as a result of compression from OPLL as well as detecting the extent of spinal cord compression. This technique is also useful to show disc herniation or foraminal stenosis that could be contributing to radiculopathy.³⁴ There are imaging risk factors that may be associated with possible symptom progression. The occupancy ratio, defined as the anterior-to-posterior ratio of the OPLL (*) in the Figure 4) to the spinal canal, can be calculated on lateral radiographs or CT sagittal images (Figure 4).

Figure 4. CT sagittal bone slices, occupancy ratio (double arrow, b-double head arrow), defined as the anterior-toposterior ratio of the OPLL (*) to the spinal canal.



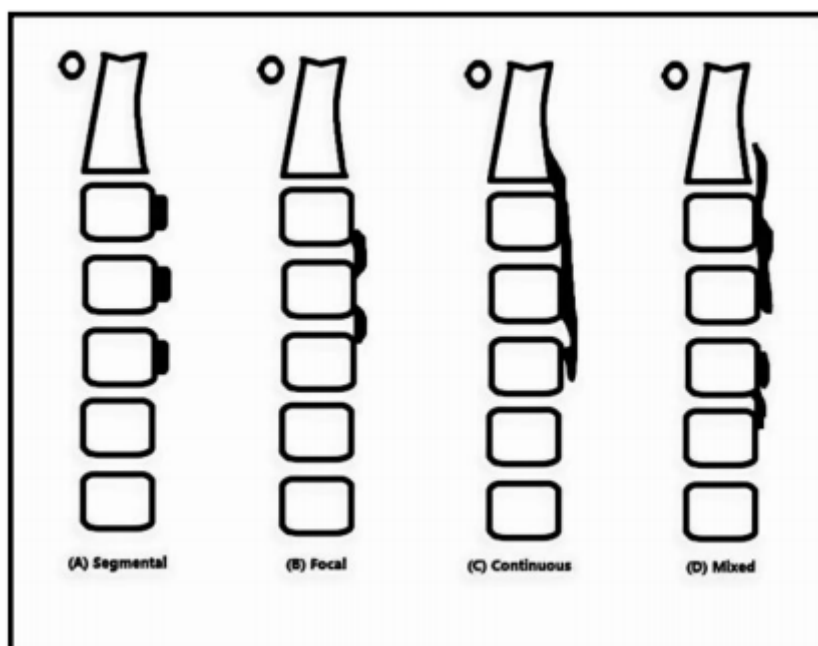
Historically are considered to exist 4 types of OPLL: the more common form are the segmental form (39%) found behind the vertebral body, the continuous form (27%) extending from vertebra to vertebra, the mixed form (29%) including both continuous and segmental elements, and the localized form (5%) opposite to the disc space with rostral and caudal extension.³⁵

The degree of cord compression is more severe in continuous OPLL and degeneration of the disk is frequently associated with both types of OPLL.^{36,37}

We present an illustration to better understand and visualize the 4 different types of OPLL; (A) Segmental: ossification occurs at multiple distinct sites behind the vertebral bodies, potentially representing an early-stage form that could originate from intervertebral disc degeneration or herniation. (B) Focal: ossification is restricted to a specific region directly behind the intervertebral space. (C) Continuous: ossification

extends over multiple vertebral bodies, spanning several intervertebral spaces. (D) Mixed: this type combines features of both segmental and continuous ossification patterns (**Figure 5**).³⁸

Figure 5. illustration of the 4 different types of OPLL.



MANAGEMENT OF OPLL

A)Non-Surgical Treatment or conservative management is appropriate for patients who are asymptomatic or have mild, non-progressive myelopathy without significant neurological deficit.³⁹

Indications

- Asymptomatic OPLL
- Mild myelopathy without progression
- Radiculopathy without cord compression
- High surgical risk patients

Treatment Options

1.Pharmacological therapy

- Nonsteroidal anti-inflammatory drugs (NSAIDs)
- Analgesics
- Neuropathic agents (e.g., gabapentinoids)
- Short-term muscle relaxants

2.Physical therapy

- Cervical stabilization exercises
- Postural training
- Gait training in mild myelopathy

3.Cervical orthosis

- Temporary immobilization in selected cases

4.Activity modification

- Avoidance of high-risk trauma activities

Limitations of Conservative Treatment

Conservative therapy does not halt the progression of ossification. Patients remain at risk of neurological deterioration, particularly those with high canal occupying ratios or dynamic instability. Close clinical and radiological follow-up is therefore essential.

B)Surgical Treatment

- Surgical decompression is the treatment of choice for patients with:
- Progressive myelopathy
- Moderate to severe neurological deficits
- Significant spinal cord compression on imaging
- Failure of conservative therapy

Early surgical intervention is associated with improved neurological recovery.⁴⁰

Different surgical approaches

The choice of approach depends on:

- Number of involved levels
- Cervical alignment (lordosis vs kyphosis)
- Canal occupying ratio
- K-line status
- Surgeon expertise

1. Anterior Approaches

- Anterior cervical discectomy and fusion (ACDF)
- Anterior cervical corpectomy and fusion (ACCF)
- Direct resection of OPLL

Indications

- Focal or segmental OPLL
- Canal occupying ratio > 60%
- Kyphotic cervical alignment
- K-line negative OPLL

Advantages

- Direct decompression of spinal cord
- Better neurological recovery in high canal occupancy cases

Disadvantages

- Technically demanding
- Higher risk of complications
- Dural tear and cerebrospinal fluid (CSF) leak

Anterior decompression has demonstrated superior neurological outcomes in selected patients with severe canal compromise.^{41,42}

2. Posterior Approaches

- Laminoplasty
- Laminectomy with fusion

Indications

- Multi level OPLL
- Preserved cervical lordosis
- K-line positive OPLL

Advantages

- Indirect decompression via posterior cord shift
- Lower risk of anterior approach-related complications

Disadvantages

- Postoperative axial neck pain
- Risk of C5 palsy
- Potential postoperative kyphosis (especially without fusion)

Posterior decompression techniques have shown good long-term outcomes in multilevel disease with maintained cervical lordosis. The choice depends on cervical alignment, patient age, instability, and surgeon preference.⁴³

Laminoplasty vs Laminectomy with Fusion

- Laminoplasty: Preserves motion but may allow continued OPLL progression.

- Laminectomy with fusion: Provides stability and reduces postoperative kyphosis but sacrifices motion segments.

CONCLUSIONS

Cervical OPLL is a potentially disabling but manageable condition. Early recognition, appropriate imaging, and individualized treatment strategies are crucial to preventing irreversible spinal cord injury and optimizing patient outcomes. Non-surgical treatment is reserved for mild or asymptomatic cases and requires close monitoring. Surgical decompression remains the definitive treatment for progressive or moderate-to-severe myelopathy. Posterior approaches are generally preferred for multilevel disease with preserved lordosis, while anterior approaches are indicated for focal disease, high canal occupancy, or kyphotic alignment.

Acknowledges

For the editorial manager for the invitation to do this manuscript. To my family for all the support. Also and specially for all the patients and colleagues.

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