

Case report of cervical myelopathy due to ossification of the posterior longitudinal ligament in association with flavum ligament ossification: MRI and CT appearance

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Background. Our objective is to analyze the cervical spinal cord damage and spinal canal stenosis due to OPLL which usually affects the cervical spine and leads to progressive myelopathy in 50–60s in Asian population; to demonstrate the mixed type OPLL and to show OPLL specific dural penetration signs: “double-layer” and “C-sign” on imaging.

Materials and methods. Subacute cord compression developed over a 3-month period in a 43-year-old Japanese patient. Severe spinal canal narrowing was related to the mixed type OPLL at C3–C4 through C6–C7 associated to flavum ligament ossification at T3–T4. Lateral radiograph of the cervical spine showed intraspinal ossification, CT demonstrated specific dural penetration signs, and MRI disclosed spinal cord compression. Laminectomy at C3–C7 was performed and decompression of the spinal cord was confirmed by postoperative MRI.

Conclusions. Absolute cervical stenosis and association with other diseases (like calcification of flavum ligament) predispose the patient to develop more severe deficit earlier in the clinical course. Specific CT signs, “double-layer” and “C-sign”, show dural involvement. MRI is a very useful modality to identify the precise level and extent of the spinal cord injury. OPLL must be included in the differential diagnosis of subacute cervical myelopathy.

Key words: magnetic resonance imaging (MRI), computer tomography (CT), cervical myelopathy, ossification of the posterior longitudinal ligament (OPLL)

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INTRODUCTION

Ossification of the posterior longitudinal ligament (OPLL) is a progressive disorder of the spine that is most prevalent in the East Asiatic countries. It occurs predominantly in the cervical spine. Patients

typically present with the onset of myelopathy. The average age of initial symptoms is during the fifth decade.

We describe specific computed tomography (CT) and magnetic resonance imaging (MRI) features of the mixed form OPLL associated with calcification of the flavum ligament revealed by a subacute onset of upper extremity sensory and motor deficits in a 43-year-old Japanese patient.

CASE REPORT

During a 3-month period, a 43-year-old Japanese male exhibited rapid development of motor impairment of both upper extremities. Neurological examination revealed brisk deep tendon reflexes and Babinski's sign bilaterally. Motor and sensory disturbances were found to be more expressed on the left side. Severe deficit was diagnosed on the left at C4, C5 and C6 and bilaterally at C7 and C8 (grade 3–4/5 muscle strength). Lateral radiographs of the cervical spine showed intraspinal ossification (Fig. 1a). CT showed a hyperdense strip of the cervical canal from C3–C4 through C6–C7 levels due to the OPLL, a specific “double layer” sign corresponding to penetration of dura mater and severe stenosis (50%) with a minimal sagittal diameter of the spinal canal at C4–C5 (Figs. 1b, c).



Fig. 1a. Lateral radiograph of the cervical spine shows heavily calcified posterior longitudinal ligament from C3–C4 through C6–C7 and degenerative changes at C5–C6

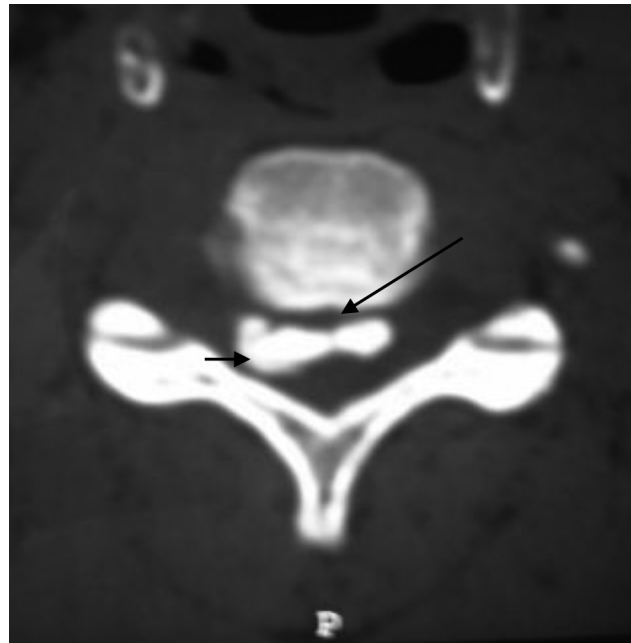


Fig. 1b. Non-contrast enhanced axial CT scan at C3–C4 demonstrates a large mass of ossification, separated from the dorsal aspect of the vertebral body by residual hypodense dura (long arrow): “double-layer” sign specific to dural penetration. “C-sign” (short arrow) is typical for one side cord compression



Fig. 1c. CT scan confirms stenosis of the cervical canal attributable to calcified PLL on sagittal reconstructions. At the narrowest level the sagittal diameter of the spinal canal is reduced to 5 mm



Fig. 1d. Sagittal T1-weighted MRI demonstrates a hypointense strip on the posterior surfaces of the vertebral bodies and discs through C3–C6 (arrow)

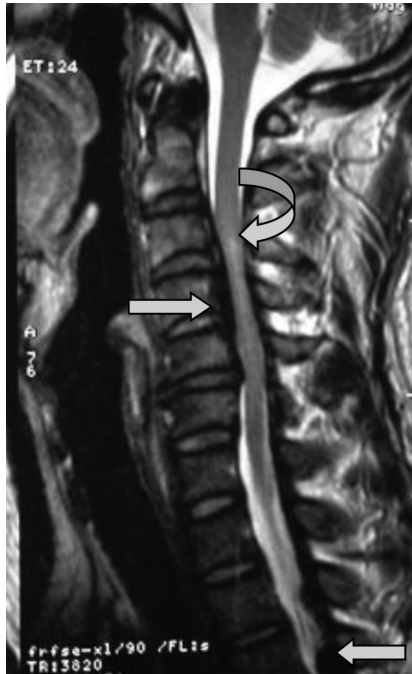


Fig. 1e. Sagittal T2-weighted MRI reveals the spinal cord injury (curved arrow) predominantly at C3–C4 (intramedullary area of high intensity signal). PLL is uniformly hypointense (right arrow) at C3 through C6. Hypointensity at T3–T4 corresponds to the left sided ossified ligament flavum (left arrow)

MRI (Figs. 1d, e) revealed a prominent linear signal void abnormality of the OPLL at the C3–C4 through C6–C7 levels narrowing the anteroposterior diameter of the canal and compression of the sac and cord. At T3–T4 the left sided ligament flavum was hypointense, corresponding to its calcification. Central intramedullary hyperintensity was identified on T2-weighted images at the C3–C4 level (Fig. 1e).

A preoperative diagnosis of the mixed type OPLL was made with signs of penetration of the dura mater and calcification of the ligament flavum. The patient underwent open door expansive C3 to C7 laminectomy. During the operation the most severe stenosis was found at the C4, C5 and C6 levels. After laminectomy the dural sac was expanded. Postoperative MRI (Fig. 2) showed



Fig. 2. Postoperative radiological findings. Sagittal T2-weighted MRI shows decompression of the spinal cord with normal subarachnoid space compartment, hypointensity of OPLL from C3–C4 through C6–C7 and a focal area of central intramedullary hyperintensity at C3–C4 and C6–C7

decompression of the spinal cord with normal subarachnoidal space compartment and hypointense lesion at the C3–C4–C5 levels corresponding the posterior longitudinal ligament position and central intramedullary hyperintensity at the C3–C4 and C6–C7 levels. Two-year follow-up revealed moderate clinical improvement, without changes in MRI appearance.

DISCUSSION

OPLL occurs not infrequently in Japan where the prevalence of 1.5–2.4% has been reported in adults (1). The disease is less common in other Asian non-Japanese populations (0.8%) and in Caucasian population in North America (0.1%) (2). Age at the onset is usually around 50–60 (3). OPLL has a predilection for the cervical spine. Seventy percent of cases occur at C2–C4, 15% at T1–T4, and 15% at L1–L3 (4).

There are 4 types of OPLL (5): 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. The degree of cord compression is more severe in continuous OPLL and degeneration of the disk is frequently associated with both types of OPLL (6).

Frequently asymptomatic, OPLL can progress and produce myeloradicular symptoms. The disease usually presents either with dominant motor and sensory disturbances in the lower and upper extremities or with segmental signs like cervicobrachialgia which causes no obvious neurologic deficit but is associated with pain in the neck, shoulder and arm. The occurrence of acute symptoms related to OPLL has been reported previously in the literature (4, 6, 7). We do not believe this is as rare as suggested by the authors. The case reported here shows that absolute cervical stenosis (canal diameter less than 10 mm) predisposes the patient with OPLL to develop more severe deficit earlier in the clinical course.

On lateral plain radiographs of the cervical spine OPLL appears as a dense strip of ossification of variable thickness. CT features of OPLL include a linear dense strip along the posterior vertebral margin, typically separated from the posterior

aspect of the vertebral bodies and intervertebral discs (4, 8). CT is useful to identify extension of OPLL through the dura, characterized by the “single-layer”, “double-layer” and C-sign. The “single-layer” and “C-sign” contribute to one side cord compression and imbrication of the dura, which indicates greater potential for an intra-operative CSF fistula (9). The “single-layer” sign on CT appears as a solid ossified ventral OPLL mass, often accompanied by lateral “C-sign” (Fig. 8 in 10). The “double-layer” is composed of separated ossified rims: the first one along the posterior margin of the hypertrophied posterior longitudinal ligament, the second one located ventrally, where it is continuous with the dorsal aspect of the vertebral body (Fig. 4 in 11). The “double-layer” sign has proven to be the most pathognomonic for absent dura at surgery (10). MRI, especially T2-weighted images are most effective to evaluate both degree and extent of the spinal cord and hyperintensities reflecting edema and / or myelomalacia.

A calcified central sequestered disc is the only condition that can be mistaken for the segmental and retrodiscal forms of OPLL.

Conservative treatment is indicated for patients with mild symptomatology. Surgery is limited to those patients with severe neurologic complications.

CONCLUSIONS

We conclude that absolute cervical stenosis due to OPLL in association with calcification of the ligament flavum predispose the patient to develop more severe deficit earlier in the clinical course. OPLL must be included in the differential diagnosis of subacute cervical myelopathy.

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**CERVIKALINĖ MIELOPATIJA DĖL
UŽPAKALINIO IŠILGINIO IR GELTONOJO
RAIŠČIŲ SUKAULĖJIMO: MRT IR KT
POŽYMIAI. ATVEJO PRISTATYMAS**

Santrauka

Darbo tikslas. Išanalizuoti pasireiškusių stuburo kanalo stenozę ir nugaros smegenų pažeidimą dėl retos patologijos – užpakalinio išilginio stuburo raiščio osifikacijos (OPLL); radiologiškai pademonstruoti mišraus tipo OPLL ir kietojo dangalo pažeidimo požymius.

Darbo metodai. Dėl mišraus tipo OPLL 43 metų japonui per tris mėnesius išsivystė poūminė nugaros smegenų mielopatija. Šoninėje kaklinės stuburo dalies rentgenogramoje buvo matyti intraspinalinė kalcifikacija, KT užfiksavo kietojo dangalo pažeidimo požymius, o MRT – mielopatijos židinių nugaros smegenyse.

Išvados. Absoliuti kaklinės dalies stuburo kanalo stenozė dėl OPLL ir geltonųjų raiščių kalcifikacijos turi įtakos ūmesnei klinikinei eigai ir ankstyvajam neurologiniam deficitui. Pažeisto kietojo dangalo KT specifiniai požymiai – „dvigubas sluoksniš“ ir „C“ požymis. MRT tyrimu nustatoma mielopatija ir nugaros smegenų pažeidimo lygis. OPLL yra viena iš priežasčių, galinčių sukelti cervikalinę mielopatiją.

Raktažodžiai: magnetinio rezonanso tomografija (MRT), kompiuterinė tomografija (KT), cervikalinė mielopatija, užpakalinio išilginio stuburo raiščio osifikacija (OPLL)

