Clinics in Diagnostic Imaging (55)

M Y S Soo

Fig. 1a Lateral radiograph of the cervical spine radiograph.

Fig. 1b Sagittal reformatted CT image of the same patient as Fig. 1a (Reproduced with permission from Soo MYS, Rajaratnam S. Symptomatic ossification of the posterior longitudinal ligament of the cervical spine: pictorial essay. Australas Radiol, 2000; 44:15)

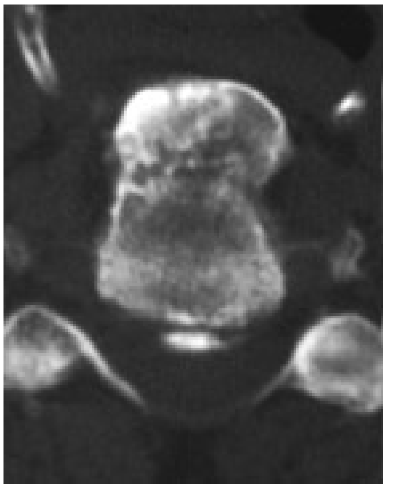

Fig. 1c Axial CT scan taken at C5 level. (Reproduced with permission from Soo MYS, Rajaratnam S. Symptomatic ossification of the posterior longitudinal ligament of the cervical spine: pictorial essay. Australas Radiol 2000; 44:15)

CLINICAL PRESENTATION

A 70-year-old Caucasian man presented with rapid progression of paraesthesia in both upper and lower extremities over a four-month period. He also noticed limb weakness, especially in the upper limbs, and experienced pain in both shoulders for two months. The positive findings on clinical examination included global upper limb weakness with increased reflexes and spasticity in all four limbs. Lower limb weakness was less pronounced. There was bilateral extensor response. The clinical impression was that of a cervical myelopathy. Radiographs and computed tomography (CT) of the cervical spine were performed (Figs. 1a-c). What are the imaging features? What is the diagnosis?

Department of Radiology
Westmead Hospital
PO Box 533
Wentsworthville NSW 2145
Australia

M Y S Soo, MBBS, DMRD, FRACR
Consultant

Correspondence to:
Dr M Y S Soo
Tel: (61) 2-9845-6522
Fax: (61) 2-9687-2109
Email:rmso@imag.wsahs.nsw.gov.au

IMAGE INTERPRETATION

The lateral cervical spine radiograph (Fig. 1a) shows diffuse idiopathic spinal hyperostosis (DISH) extending from the second to the seventh cervical vertebrae. Linear calcifications measuring between 1 and 2 mm in thickness are present along the course of the posterior longitudinal ligament between the second and third cervical vertebrae. A similar finding is noted along the posterior borders of C5 and C6 vertebrae. There is moderate posterior osteophytosis at C6/C7 level. The sagittal diameter of the cervical spinal canal is narrowed to 15mm (normal range 16-24mm) at C2 level.

These radiographical findings are confirmed by CT. The sagittal reformatted CT image (Fig. 1b) shows ossification of the posterior longitudinal ligament (OPLL) at C5/C6 levels as well as the changes of DISH. In both the sagittal and axial CT images, a thin radiolucent band separates the posterior vertebral margin from the OPLL (Fig. 1c). At both C2/C3 and C5/C6 levels, the sagittal diameters of the cervical canal are significantly narrowed.

DIAGNOSIS

OPLL of the cervical spine causing spinal canal stenosis.

CLINICAL COURSE

After neurological consultation, opinion was that the patient's myelopathic syndrome was more in keeping with a high cervical cord compression rather than a multilevel canal stenosis, with the upper limb weakness being due to stenosis of the nerve roots exit foramina at the affected levels. A C1 laminectomy with foramen magnum decompression, followed by a posterior occipitocervical fusion, was performed. Post-operatively, the patient's symptoms improved markedly. He continued to improve at follow-up six months later and the occipitocervical fusion has remained stable.

DISCUSSION

Resnick[1] credited Key (1839) to be the first person to report compromise of the spinal canal due to OPLL. However, recognition of OPLL as a distinct clinical entity did not occur until the early 1960s. The incidence amongst Japanese is 1.5 to 2%[2] and 0.8% among non-Japanese Asians[3]. A recent report revealed an incidence of up to 0.7% in North Americans[4]. Symptomatic OPLL affects males more frequently than females[5,6,7]. The aetiology of OPLL is probably multifactorial. The high incidence of DISH in patients with this disease suggests a hereditary diathesis of the spinal ligament ossification. The pathogenesis of OPLL is thought to involve inheritance of OPLL-related human leukocyte antigen (HLA) haplotype genes from both parents. The condition may thus occur in the presence of this genetic predisposition and with the interaction of various other environmental factors, including repeated minor trauma and regressive degeneration due to ageing[8]. A gene abnormality on chromosome 6 has also been implicated[9]. Ikegawa et al[10] found an increase of serum growth hormone (GH) binding protein, which is in turn associated with an increased number of GH receptors in patients with OPLL. Bone-seeking hormones, such as parathyroid hormone and calcitonin, may also play a role in OPLL.

Fig. 2 Lateral cervical spine radiograph of a 45-year-old Chinese man who presented with compressive myelopathy shows a typical OPLL extending from C2 to C5 levels. At C3 level, the ossified ligament occupies more than 50% of the sagittal canal diameter (arrowhead).

The majority of subjects with cervical OPLL are asymptomatic. Symptoms are more likely to occur when the OPLL occupies more than 40% of the normal canal diameter, with development of cord compression when it reaches 60%[11]. The disease usually presents between the fifth and seventh decades of life. Ono et al[11] divided the abnormal neurological findings into three groups, the commonest being cord signs, manifested by dominant motor and sensory disturbances in the lower extremity, followed by segmental signs, manifested by predominantly motor and sensory symptoms in the upper extremity. Cervical brachalgia without neurological signs is a rarer but recognised presentation. Symptoms are precipitated by trauma in up to 20% of cases. Other less common symptomatology includes urinary and faecal incontinence, and loss of libido. Physical examination may reveal muscle atrophy,

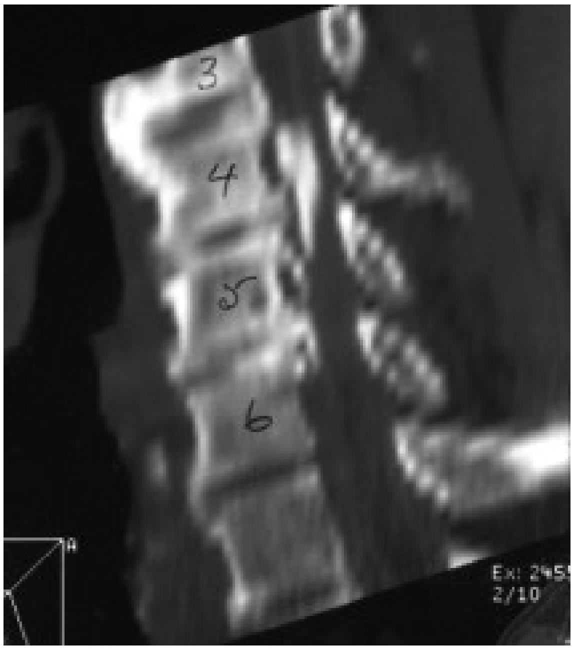

Fig. 3a Sagittal reformatted CT image of a 39-year-old Vietnamese man who presented with a three-week history of lower limb weakness and spasms. A combination of continuous and segmental forms (mixed form) of OPLL is shown. Note that the OPLL has produced narrowing of the canal to more than 60% and is causing marked cord compression. The patient subsequently underwent anterior surgery. (Reproduced with permission from Soo MYS, Rajaratnam S. Symptomatic ossification of the posterior longitudinal ligament of the cervical spine: pictorial essay. Australas Radiol 2000; 44:15)

fasciculation, hyperreflexia and sensory loss. Laboratory investigations are usually unrewarding. An association with diabetes mellitus has been reported[2].

The diagnosis of OPLL is established by its characteristic radiographical and CT appearances. In the cervical spine, a dense ossified strip or plaque of variable thickness (1 to 5 mm) is evident along the posterior margins of the vertebral bodies and the intervertebral discs. It is most common in the midcervical region (C3 to C5) although any cervical level may be affected (Fig. 2). On axial CT sections, the OPLL is depicted as a mushroom - or "hill"- shaped ossified mass. A thin radiolucent line representing the non-ossified deep layer of the PLL is present between the ossified superficial layer and the posterior vertebral border. This sign is typically found in about 50% of patients with OPLL (Figs. 3 and 4)[11,12]. OPLL may be confined to one or several vertebral levels, but without involving the disc spaces (segmental form). Most commonly it extends in an uninterrupted fashion spanning several vertebral levels (continuous form). A mix of the segmental and continuous forms can occur in some patients (Figs. 3 and 5). A rare retrodiscal OPLL, sometimes indistinguishable from posterior osteophytosis, has also been described[13].

There is a possible association between the focal retrodiscal form of OPLL and disc herniation. Hanakita et al suggested that OPLL causes weakness of the

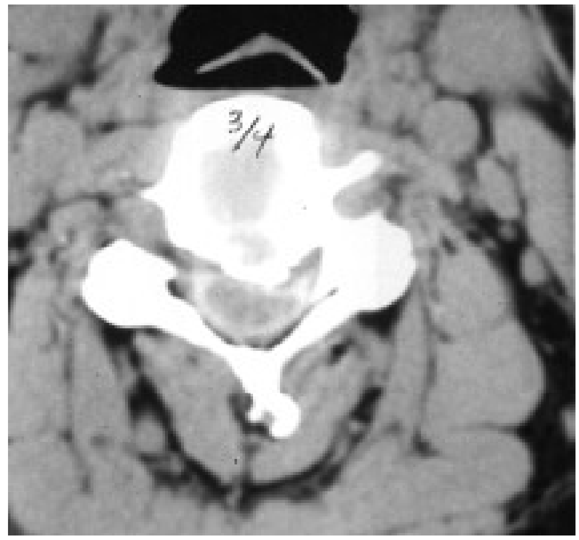

Fig. 3b Axial CT myelogram of the same patient as Fig 3a. At the most cephalad portion of the OPLL at C3/C4 level, the lesion is almost similar in appearance to an osteophyte.

Fig. 3c Sagittal T1-weighted MR image of the same patient as Figs. 3a-b shows a marked signal void posterior to C4 vertebra (arrowheads), corresponding to the OPLL seen on CT. Cord compression is maximum from C4 to upper C5 levels. The C4/C5 and C5/C6 discs show loss of height and signal intensity, indicative of degeneration. The C4/C5 disc is also herniated posteriorly.

Fig. 3d Axial T2-weighted MR image taken at C4/C5 level of the same patient as Figs. 3a-c. The cord compression has resulted in intramedullary areas of high signal intensity. Note stretching of the C5 roots (arrows).

Fig. 4 Axial CT scan taken at C4 level shows OPLL. A radiolucent line (arrowheads) separates the vertebral margin from the OPLL, which has a characteristic "mushroom" appearance.

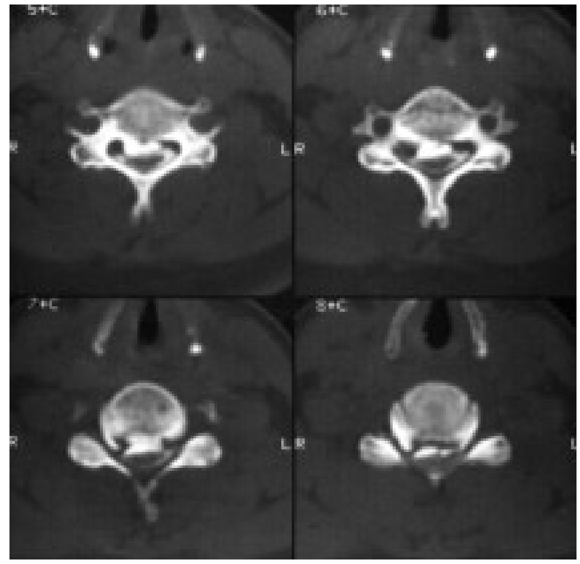

Fig. 5 Series of axial CT myelogram scans through the mid-cervical spine of a middle-aged Fijian woman presenting with progressive myelopathy. The OPLL causes cord compression. A radiolucent line is not present as both layers of the PLL have ossified. The OPLL has a "hill" shape which is another common appearance on CT. The patient required anterior surgery.

Fig. 6a 63-year-old Indian woman with a long history of left C7 brachalgia. Axial CT scan at C6/C7 level shows a moderate-sized ossified lesion encroaching posteriorly into the spinal canal (arrows). The absence of a radiolucent line and a similar bone texture to the adjacent vertebra favour a posterior osteophyte. She was managed conservatively.

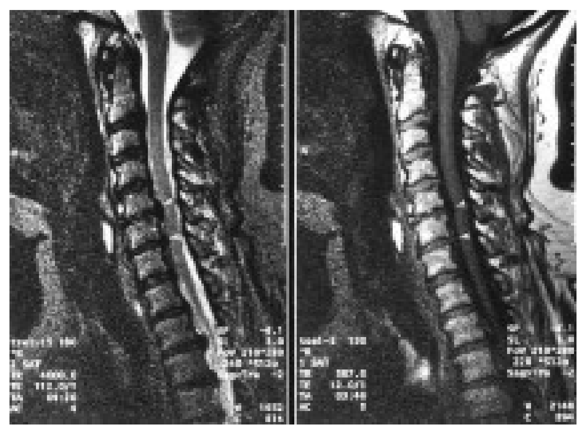

Fig. 6b Sagittal T2- (left) and T1-weighted (right) MR images of the same patient as Fig. 6a. The areas of very low signal intensity indenting the thecal sac at C5/C6 and C6/C7 levels in both sequences are due to cortical bone (osteophytes) (arrowheads). Note that the T1-weighted image shows more morphological details, including a small posterior disc protrusion at C6/C7.

ligament and a resultant increased incidence of disc herniation (up to 79%) in this subset of OPLL[14]. Others have postulated an increase in posterior osteophytic build-up with extension into the PLL. When posterior osteophytic formation becomes excessive, the osteophytes may be indistinguishable from focal OPLL (Fig. 6)[15]. A focal posterior midline calcified sequestrated disc can also mimic the retrodiscal form of OPLL[16]. A distinctive feature that characterises fused posterior osteophytes is their horizontal growth orientation (Figs. 7 and 8). This contrasts with the vertical spans of OPLL (Figs. 2 and 3).

On magnetic resonance (MR) imaging, OPLL is seen as a band of low signal intensity between the bone marrow of the vertebral body and the dural sac on both T1- and T2-weighted sequences (Fig. 3c-d). An area of increased signal intensity, corresponding to fat within the ossified ligament, is visible in up to 50% of cases of the continuous type of OPLL. The segmental form of OPLL is less distinctive. OPLL needs to exceed 3 mm in thickness to be unequivocally discerned on MR imaging (Fig. 9)[17]. Concomitant cord compression and its attendant complications, such as myelomalacia and oedema, are clearly depicted as areas of high signal intensity on T2-weighted images (Fig. 3). The neurological deficits observed in patients with intramedullary high intensities on T2-weighted sequences are more severe than those with normal signal, implying a poorer surgical prognosis[18]. The ossified ligament can extend laterally to the neural foramen to stretch the exiting nerve roots (Fig. 3d). In many respects, MR imaging has supplanted CT myelogram in the surgical work-up of OPLL[18].

Fig. 7 Coronal (left) and sagittal (right) reconstructed cervical CT images of a 50-year-old Caucasian man presenting with right radiculopathy. At C5/C6 level, the virtually fused posterior osteophytes have a horizontal orientation.

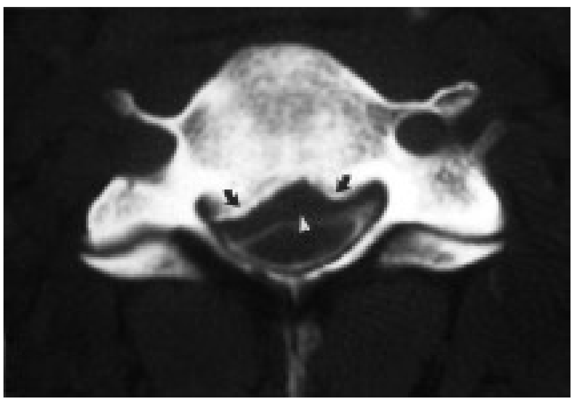

Fig. 8 Axial CT myelogram of a 56-year-old Caucasian man presenting with a progressive cervical myelopathy taken at C5 level shows paracentral posterior osteophytes (curved arrows). Note the marked cord compression caused by a concomitant herniated disc at the midline (arrowhead). These findings were confirmed at anterior surgery.

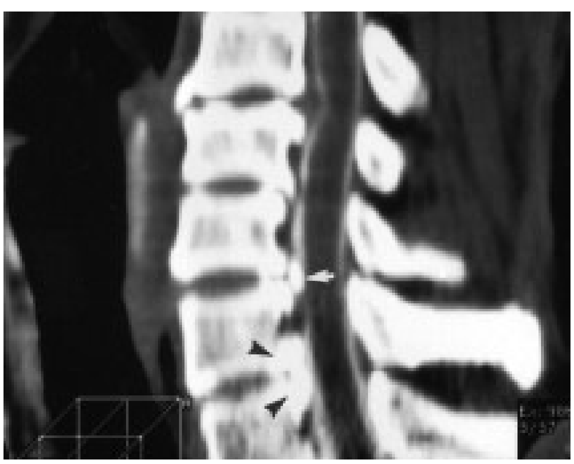

Fig. 9 42-year-old Chinese man who presented with a one-month history of decreasing sensation in all four extremities. Sagittally-reconstructed CT myelogram shows posterior osteophytosis at C4/C5. At C6/C7 level, a calcified sequestrated disc was recovered at anterior surgery (arrow). This was mistaken to be a retrodiscal form of OPLL pre-operatively. Another ossified lesion thought to be retrodiscal OPLL is seen at C7/T1 level (arrowheads). This measures 2 mm thick and was not distinctive on MR images.

Conservative measures should be initially tried for those patients with less disabling symptoms. When a patient presents with recent rapid onset of compressive myelopathy as in our case, then surgical decompression is indicated. There is no agreement in the literature as to the preference of either the anterior and posterior approaches in terms of surgical outcome[19]. Anterior surgery aims at removal of the offending calcified ligament which compromises the blood supply to the cord. This technique decompresses the cord and widens the subarachnoid space[20]. It includes a multilevel vertebrectomy, removal of osteophytes and OPLL, interbody fusion with a bone graft and metal plate fixation. This approach is used when disease is confined to less than three levels that extend between C3 and T1 vertebrae. There is a relatively longer convalescent period as fixation with a halo vest is required. The potential complications include transdural cerebrospinal fluid leak, injury to the vertebral arteries, vertebral haematoma, recurrent laryngeal nerve palsy and infection at the site of halo insertion. Fracture and dislodgment of the bone graft and failure of the graft to unite are remote complications requiring re-exploratory surgery.

Posterior decompression is preferred for the continuous type of OPLL that spans more than three or more vertebral levels[21]. The basic principle of posterior surgery is to widen the cervical canal and consists of laminectomy or its modification, expansive open-door laminoplasty. The latter procedure aims at decompression with preservation of the posterior facet joints[13]. With severe canal stenosis, laminectomy may be extended laterally to include foramenotomy which aims at relieving pressure on the existing nerve roots. The major objection to posterior surgery is its inability to adequately expose and safely remove the compressive structures anterior to the cord[21]. Occasional post-operative spinal instability leads to a kyphus deformity and thus increased tension on the blood vessels of the cord. Scar tissue formation can constrict the adjacent nerve fibres resulting in more pain. Complications related to laminoplasty are stated to be lower than that of the anterior approach and the results of the recovery from the myelopathy in a two-year follow up are about equal in one series[22].

In conclusion, OPLL is not an uncommon cause of cervical radiculomyelopathy. It occurs mainly in middle-aged male individuals, irrespective of racial origins. OPLL has distinctive imaging features. Modern surgical treatment is safe and offers relief for those with disabling symptomatology.

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ABSTRACT

Ossification of the posterior longitudinal ligament (OPLL) of the cervical spine associated with diffuse idiopathic skeletal hyperostosis is described in a 70-year-old Caucasian man presenting with a rapidly progressive myelopathy. The acute nature of his myelopathic symptoms and cervical canal stenosis necessitated posterior decompressive surgery. Four other patients with OPLL are presented to illustrate the spectrum of imaging findings. The computed tomographic features of OPLL are distinctive. A 2-5 mm thick linear ossified strip along the posterior vertebral margin usually at mid cervical (C3 to C5) level characterises the condition. Magnetic resonance (MR) imaging is valuable in excluding possible cord damage and associated disc lesions prior to surgery. A calcified central sequestrated disc is the only condition that may be mistaken for the segmental and retrodiscal forms of OPLL. In a clinical setting of compressive myelopathy, it is pertinent to distinguish between these two conditions since a sequestrated disc has a more favourable surgical prognosis. The merits and relevance of anterior and posterior surgery together with their possible complications are outlined.

Keywords: Cervical myelopathy, computed tomography, ligament ossification, magnetic resonance imaging, posterior longitudinal ligament

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