Symptomatic ossification of the anterior longitudinal ligament with stenosis of the cervical spine

A REPORT OF SEVEN CASES

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Seven men with a mean age of 63.9 years (59 to 67) developed dysphagia because of oesophageal compression with ossification of the anterior longitudinal ligament (OALL) and radiculomyelopathy due to associated stenosis of the cervical spine. The diagnosis of OALL was made by plain lateral radiography and classified into three types; segmental, continuous and mixed. Five patients had associated OALL in the thoracic and lumbar spine without ossification of the ligamentum flavum.

All underwent removal of the OALL and six had simultaneous decompression by removal of ossification of the posterior longitudinal ligament or a bony spur. All had improvement of their dysphagia. Because symptomatic OALL may be associated with spinal stenosis, precise neurological examination is critical. A simultaneous microsurgical operation for patients with OALL and spinal stenosis gives good results without serious complications.

Although ossification of the posterior longitudinal ligament (OPLL) is associated with radiculomyelopathy, ossification of the anterior longitudinal ligament (OALL) has not been widely described since it is rarely symptomatic. Resnick et al6 and Resnick, Shaul and Robins7 coined the term diffuse idiopathic skeletal hyperostosis for Forestier’s disease and ossification of the spinal ligaments has been considered as a part of this entity.8,9 They defined diffuse idiopathic skeletal hyperostosis as showing calcification or ossification along the anterior to anterolateral aspect of four contiguous vertebral bodies with relative preservation of the height of the intervertebral disc in the affected areas,9 distinguishing it from degenerative discogenic disease. In the past dysphagia because of OALL has been confused with that caused by degenerative disc disease because of poor recognition of ossification of the spinal ligaments.10-20 The management of symptomatic OALL is still controversial.

Although conservative treatment with anti-inflammatory medication may be effective, aspiration pneumonia has been described11,17,21,22 and there may be myelopathy due to coexisting spinal stenosis.3

We describe the clinical manifestations, radiological diagnosis and surgical treatment of symptomatic OALL in seven patients with special emphasis on simultaneous surgery for OALL and coexisting spinal stenosis.

Patients and Methods

Between January 1987 and December 2002, seven patients with OALL who presented with dysphagia were treated surgically. All were men with a mean age of 63.9 years (59 to 67). The dysphagia was due to oesophageal compression and all had radiculomyelopathy secondary to coexisting stenosis of the cervical spine. Their clinical details are summarised in Table I.

Clinicoradiological characteristics. In all cases, the diagnosis of OALL was made on plain lateral radiographs. There was hyperostosis of the anterior longitudinal ligament at several levels with preservation of the disc height. The degree of anterior projection and the shape of the affected disc were best visualised on transverse CT scans. MRI was effective in evaluating associated compression of the cord. OALL was classified into three types on radiological studies. The segmental type was defined as partial or total ossification over a vertebral body without involving the disc space. The continuous type showed ossification over many disc spaces as well as the vertebral body. In the mixed type there was a combination of the segmental and continuous types (Fig. 1). Of the seven patients, two showed the segmental, three the continuous and two the mixed types. In the continuous and mixed cases OALL was associated with OPLL in the cervical spine and in the segmental cases the spinal stenosis was caused.
by cervical spondylosis. The patients with continuous and mixed disease had coexisting OALL in the thoracic and lumbar regions. Ossification of the ligamentum flavum was not seen.

Operative procedures. A microscopic anterior approach was used for removal of the OALL. A transverse skin incision was employed in six patients in whom the OALL and spinal stenosis were located at the same or at adjacent levels, while a longitudinal incision was made in one patient in whom the OALL and OPLL were at different levels. The platysma was exposed and the operating microscope introduced. Adhesions were present between the oesophagus and the OALL which were separated using a microdissector. After retracting the trachea and oesophagus the ossified mass was incised and the anterior surface smoothed using ronguers and a high-speed drill with a 5-mm burr. A decompressive procedure for stenosis was carried out in six patients. The other patient had a mild left hemiparesis because of previous cerebral infarction and decompression was not undertaken. Compression of the cord was seen at one level in two patients and at two levels in four. Titanium interbody cages were used for fixation after decompression.

Post-operative course. One patient was temporarily hoarse, but otherwise no new symptoms occurred. This patient wore a hard cervical collar for one month. All the patients were able to swallow both liquid and solid food. All were followed up for a mean of 17.1 months (12 to 36); None developed recurrent symptoms or radiological evidence of further ossification.

Illustrative cases

Case 1 (Table I). A 63-year-old man developed severe dysphagia and mild hoarseness three months before admission. He had a mild right hemiparesis after cerebral infarction three years before. The dysphagia gradually deteriorated, and he had difficulty in swallowing solid food. Neurological examination showed a mild hemiparesis, numbness in both upper limbs, severe dysphagia and mild hoarseness. Plain lateral radiography revealed OALL from C1 to C7 with prominent anterior projection of OALL from C2 to C5. Stenosis of the spinal canal was present from C3 to C6 with a small segmental OPLL at C4 to C5. The anterior longitudinal ligament of both the thoracic and lumbar spine was ossified. The disc height in the cervical spine was relatively normal. MRI revealed marked compression of the pre-vertebral soft tissue including the oesophagus and trachea. Resection of the OALL was undertaken from C2 to C5 through an anterior approach using an operating microscope. Decompression of the spinal stenosis was not performed because this patient was hemiparetic on the right side. There were adhesions between the OALL and the oesophagus which required meticulous separation and gentle retraction of the oesophagus. The dysphagia and hoarseness immediately improved after operation (Fig. 2).

Case 7 (Table I). A 65-year-old man slowly developed dysphagia over a period of two years. There was also some clumsiness in both hands for one year, but he was otherwise healthy. Neurological examination showed clumsiness of the hands and hyperaesthesia in C7/8 with severe dysphagia. A plain lateral radiograph revealed OALL from C4 to C6 and a posterior spur at C6/7. CT confirmed marked anterior projection of the OALL compressing the pre-verte-
bral soft tissue. MRI showed compression of the cord due to cervical stenosis. A simultaneous anterior procedure combined with removal of the OALL and decompression of the spinal canal was performed. There were slight adhesions between the OALL and the oesophagus. He made a good post-operative recovery (Fig. 3).

**Discussion**

The patients in our series were all men in the sixth or seventh decade as is seen in OPLL. Both the anterior and posterior longitudinal ligaments run longitudinally along the vertebral body from the cervical spine to the sacrum. Because of the frequent association of OALL and OPLL, the same mechanism may enhance ossification of these spinal ligaments, but none had ossification of the ligamentum flavum, which may be explained by the anatomical difference between this structure and the anterior or posterior longitudinal ligament. Because the ligamentum flavum is attached to the lamina segmentally it may be affected by extension, flexion and rotational forces more than the longitudinal ligaments. Overstretching of the ligamentum flavum by neck movements may be required in addition to a systemic hyperostotic factor for the initiation and progression of ossification, whereas dynamic stress would not be of importance in OALL and OPLL.

Neurological complications from OALL are rare. Myelopathy or radiculopathy in our patients was caused by stenosis of the cervical spine due to coexisting OPLL or cervical spondylosis. The most common symptoms of OALL are compression of the oesophagus and trachea. Although fewer than 10% of patients require surgical decompression of these structures, aspiration pneumonia and suffocation by aspiration of food have been described. Dysphagia due to OALL which is severe or resistant to conservative treatment should be treated by decompression of the protruding ligamentous mass.
Figure 3a – A lateral cervical radiograph with barium contrast showing severe compression of the oesophagus because of segmental ossification of the anterior longitudinal ligament (OALL) at C4/5. Figure 3b – T2-weighted MRI showing compression of the pre-vertebral soft tissues, including the trachea and the oesophagus from C4 to C6 with mild compression of the cord by a spur at C6/7. Figure 3c – CT showing anterior projection of the OALL. Figure 3d – Post-operative plain radiograph showing resection of the OALL and fusion of C6/7.
Radiological evaluation should include plain lateral radiography and CT. MRI cannot differentiate OALL from an anterior spur, although compression of the pre-vertebral soft tissues or spinal cord is well demonstrated. A barium meal and endoscopy may be undertaken for further evaluation of the oesophagus. The radiological patterns of the segmental, continuous and mixed types in OALL are similar to those of OPLL. All continuous and mixed cases were associated with thoracic and lumbar OALL, while segmental OALL was isolated in our patients. This suggests that radiological evaluation should be performed in the thoracic and lumbar spine as well as in the cervical region because compression of the trachea and oesophagus can occur at the upper thoracic level. Stuart and Underberg-Davis and Levine described oesophageal compression by the thoracic spine. Segmental OALL and an anterior spur may be confused because of their clinical and radiological similarity. However, a decrease in disc height, a posterior spur, although compression of the pre-vertebral soft tissues or spinal cord is well demonstrated. A barium meal and endoscopy may be undertaken for further evaluation of the oesophagus. The radiological patterns of the segmental, continuous and mixed types in OALL are similar to those of OPLL.

Although mild dysphagia due to OALL may be treated with anti-inflammatory medication, surgical decompression should be performed when the symptoms are severe. The use of an operating microscope for resection of the OALL helps avoid complications such as oesophageal injury. None of our patients had experienced mortality or recurrent symptoms. Some advise cervical fusion after resection of OALL, but it can be treated by simple resection of the ossified mass without fixation.

Six of our seven patients had simultaneous resection of the OALL and decompression for spinal stenosis. A routine anterior procedure enabled us to remove both the OALL and coexisting OPLL or cervical spondylolisthesis using a single incision. Laminectomy or laminoplasty by a posterior approach for spinal stenosis coupled with anterior resection of OALL may be an alternative procedure in cases of OALL and spinal stenosis at three levels. Spinal fusion is required after decompression because instability may produce further symptomatic osteophytes causing dysphagia. Hirano et al. and Suzuki et al. have each described a patient with recurrent dysphagia due to an anterior osteophyte.

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References