Case report

Cervical myelopathy, ossification of the posterior longitudinal ligament, and diffuse idiopathic skeletal hyperostosis: problems in investigation

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SUMMARY This report describes a patient presenting with a spastic quadriplegia who was found to have both diffuse idiopathic skeletal hyperostosis (DISH) and ossification of the posterior longitudinal ligament (OPLL) in the cervical spine. There was a dramatic worsening of his symptoms during a myelogram examination of the neck. It is suggested that computed tomographic imaging of the neck is the preferred investigative procedure if OPLL is suspected as a cause of cervical myelopathy.

Key words: spastic quadriplegia, myelography, CT scan.

Cervical myelopathy is not uncommon in rheumatology clinics and most frequently is due to cervical spondylosis. Ossification of the posterior longitudinal ligament (OPLL) in the cervical spine is uncommon, and most series have originated in Japan.1-3 The association of OPLL and diffuse idiopathic skeletal hyperostosis (DISH) is recognised, particularly in Caucasians.4,5 The patient described had both DISH and OPLL and presented with a severe cervical myelopathy. There was a dramatic deterioration in symptoms and signs during myelography.

Case report

The patient was a 55 year old white man who presented in September 1985 with a two year history of paraesthesia in both hands in a non-dermatone distribution. For six months he had experienced increasing difficulty in walking because of ‘heaviness’ of the legs. His walking distance was 10 metres at presentation. He had a previous history of gout and hypertension, and was taking allopurinol 300 mg daily, atenolol 100 mg daily, and nifedipine 20 mg daily at presentation. On examination he weighed 107 kg. The salient positive findings were of restricted cervical spine movement and a spastic quadriplegia with an absent jaw jerk. There was sensory loss to all modalities below the level of T5. There was no clinically apparent bladder or bowel involvement. Routine laboratory investigations including full blood count, erythrocyte sedimentation rate, urea and electrolytes, fasting blood sugar, calcium, phosphate, alkaline phosphatase, and serum uric acid were all normal. Organ and non-organ specific autoantibodies were not detected.

Plain radiographs showed evidence of DISH with ‘flowing’ anterior osteophytes in both the lumbar and lower cervical spine (Figs 1 and 2). Pelvic radiographs showed a rim of osteophytes around the acetabular margin with preservation of joint space (Fig. 3). There was no evidence of sacroiliitis. Careful inspection of lateral views of the cervical spine showed calcification of the posterior longitudinal ligament extending from C3 to C7. He was referred to the neurosurgical unit, where he underwent a lumbar route myelogram and post-myelographic computed tomographic (CT) examination of the cervical spine. During myelography there was a sudden deterioration in his clinical state with the development of a flaccid paresis in the left arm, probably due to the prone...
Cervical myelopathy, ossification of the posterior longitudinal ligament, and DISH

Fig. 1 Lateral view of cervical spine; with 'flowing' anterior osteophytes and ossification of the posterior longitudinal ligament (arrowed).

Fig. 2 Lateral view of lumbar spine; with typical features of DISH.

Fig. 3 Pelvic radiograph showing 'rim' of acetabular osteophytes and ossification of the left sacrotuberous ligament.

position with extension of the neck compromising further the compressed cord. The myelogram showed a complete block to the passage of contrast media at C5. The CT images showed dense calcification of both the anterior and posterior longitudinal ligaments (Figs 4 and 5). At C3 the posterior longitudinal ligament obliterated more than half the spinal canal. No contrast media was present in the subarachnoid space at this level.

He underwent a posterior cervical laminectomy from C2 to C7. There has been a slight clinical improvement compared with his postmyelogram condition, but he is wheelchair bound, able to transfer only with help, and continues to have a flaccid paresis of the left arm.

Discussion

OPLL was first described in the English literature in 18 Japanese subjects, six of whom had a significant cervical myelopathy. These cases were identified from a total group of 1800 patients seen with cervical spine related disease. No association with DISH was observed. OPLL with cervical myelopathy was reported in two Caucasians in 1969. Nakanishi et al described eight Japanese patients with OPLL and cervical myelopathy. Seven were male and the age range at presentation was 41–63 years. In a
comparative study of subjects with either OPLL or cervical spondylosis they found a higher prevalence of neurological abnormalities in those with OPLL. Again, no association with DISH was seen. Ono et al reported a large series of 166 Japanese patients with OPLL. They found that the prevalence of cervical myelopathy related to the severity of spinal canal obliteration by the OPLL. In their series radiological features of DISH were commonly present. No inflammatory, endocrine, or metabolic cause could be identified. Posterior decompression was the most effective surgical treatment, and 13 out of 23 patients with neurological signs improved after decompression. Resnick et al reviewed the cervical spine radiographs of 74 North American patients with DISH, and OPLL was detected in half. Spastic quadriplegia did not, however, appear to be a major feature in these cases, neurological signs only developing when significant encroachment of the canal had occurred.

The patient described in this report demonstrates two clinically important features. Firstly, although the OPLL appearances on CT scan were dramatic, the changes on conventional plain cervical spine radiographs were more subtle and were initially overlooked. Secondly, he had a dramatic clinical deterioration at the time of myelography. It would seem prudent to precede or replace conventional myelographic studies by plain or preferably intrathecal contrast enhanced CT imaging of the cervical spine if OPLL is suspected as being the cause of a cervical myelopathy. The clinical index of suspicion of OPLL should be increased if the patient has coexisting DISH.

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