Ossification of the posterior atlantoaxial membrane associated with an os odontoideum: a case report

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ABSTRACT

We report a case of ossification of the posterior atlantoaxial membrane associated with an os odontoideum in a 46-year-old woman. She developed myelopathy following a minor motor vehicle accident. The patient underwent posterior atlantoaxial arthrodesis and resection of the ossified lesion and recovered uneventfully. Long-standing atlantoaxial instability might have played a role in ectopic ossification of the posterior atlantoaxial membrane.

Key words: atlanto-axial joint; ligamentum flavum; ossification, heterotopic; spinal cord compression

INTRODUCTION

The posterior atlantoaxial membrane is a collagenous tissue bridging the posterior arch of C1 and the cranial edge of the C2 lamina and acts as an additional stabiliser of the atlantoaxial complex.1 Ectopic ossification of the membrane is rare and can cause severe myelopathy secondary to spinal cord compression. An os odontoideum is defined as an ossicle with smooth circumferential cortical margins and no osseous continuity with the body of axis.2–4 Patients with an os odontoideum can present with pain or myelopathy, or remain asymptomatic. We report a patient with ossification of the posterior atlantoaxial membrane associated with an os odontoideum.

CASE REPORT

In June 2009, a 46-year-old woman presented with a one-month history of weakness in the upper extremities and gait disturbance and limited cervical motion following a minor motor vehicle accident. Neurological examination revealed weakness (grade 4/5) in her left deltoid, left biceps, left triceps, and bilateral intrinsic muscles. She had decreased sensation in the bilateral upper extremities, below the level of the C5 dermatome on the left and C7
on the right. She also had intact cranial nerves and hyperreflexia on the left upper extremity with a positive Hoffmann sign; knee and ankle reflexes were increased bilaterally with positive Babinski signs. A spastic gait and clumsiness of her hands were evident, but there was no bowel or bladder difficulty.

Lateral radiographs showed an os odontoideum with atlantoaxial instability (Fig. 1). Reconstruction computed tomography demonstrated ossified tissue between the C1 posterior arch and the C2 lamina (Fig. 2). Magnetic resonance imaging demonstrated severe cord compression with intramedullary high signal intensity change (Fig. 3).

The patient underwent atlantoaxial stabilisation with transarticular screws. The dural sac was severely compressed by the ossified ligament even after anatomic reduction of the atlantoaxial joint. The posterior arch of C1 was then undercut for decompression, and the ossified lesion resected. Pathological examination of the specimen showed diffuse ossification in the degenerated ligament with irregular remodelling and hyperplasia of fibrous cartilage.

Immediately after surgery, motor weakness and sensory disturbance improved. At the 6-month follow-up, she had almost regained her full strength, and her gait disturbance had also been relieved. At month 12, she had returned to her job as a sports instructor. At month 18, she remained free of symptoms with solid bone fusion, despite having hyperreflexia in her lower extremities.

DISCUSSION

Ossification of the posterior atlantoaxial membrane is a rare cause of spinal cord compression. Only 5 such cases have been reported.5–9 Most such patients were

Figure 1 Flexion/extension lateral radiographs showing an os odontoideum (arrows) and atlantoaxial instability.

Figure 2 Axial and reconstruction computed tomographic scans showing ossification of the posterior longitudinal ligament (arrows).

Figure 3 T2-weighted magnetic resonance image demonstrating cord compression with intramedullary high signal intensity change.
in their fifth or sixth decade of life and presented with progressive myelopathy (Table). None but our patient showed atlantoaxial instability. Most of the ossified lesions showed laterality, as is often the case with the ossification of the ligamentum flavum in the thoracic spine. Based on axial computed tomography, the condition can be classified as unilateral, bilateral, and tongue type (a flat lesion in the midline, as in our patient). The mainstay of treatment is laminectomy, and outcome is satisfactory for those without atlantoaxial instability.

The pathophysiological mechanism of this condition remains unclear. According to one theory, ossification of the ligamentum flavum often develops in the lower thoracic spine where the spine has more mobility than in the upper or middle thoracic regions. It is postulated that chronic mechanical stress on the ligament may lead to the development of the ossification. Mechanical stress may induce osteogenic differentiation of the ligament cells. In our patient, persistent atlantoaxial instability may have played a role in the ectopic ossification.

### Table

<table>
<thead>
<tr>
<th>Study5–9</th>
<th>Sex/age (years)</th>
<th>Type</th>
<th>C1-2 instability</th>
<th>Coexisting ossification</th>
<th>Surgery</th>
<th>Outcome</th>
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<tbody>
<tr>
<td>Yamaguchi et al5, 1992</td>
<td>M/46</td>
<td>Bilateral</td>
<td>-</td>
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<td>Kimura et al, 1998</td>
<td>F/55</td>
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<td>-</td>
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<tr>
<td>Harimaya et al7, 2003</td>
<td>M/52</td>
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<td>Shoda et al, 2005</td>
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<td>Present case</td>
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<td>Tongue</td>
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<td>No</td>
<td>Laminectomy, atlantoaxial stabilisation</td>
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</tr>
</tbody>
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### REFERENCES