Although anomalies of the upper cervical spine are not uncommon and have multiple variations, only 5 cases of cervical myelopathy caused by invagination of an anomalous lamina of the axis have been reported. We describe an extremely rare case of cervical myelopathy caused by invagination of the bilaterally separated lamina of the axis and present a review of the literature.

Case Report

The patient was informed that the authors would describe his case for publication and he provided consent.

History. A previously healthy 68-year-old man reported bilateral hand numbness that began 1 year before admission to Kansai Rosai Hospital in Amagasaki, Japan. During the year before admission, he had also experienced a slowly progressive gait disturbance, clumsiness of his right hand, and right dominant sensory disturbance in all 4 extremities and the trunk. He had no history of neck or head injury.

Examination. Physical examination at admission revealed right dominant hypesthesis of all 4 extremities and the trunk, motor weakness of the right hand, and diffuse hyperreflexia in all extremities. A lateral radiograph of the cervical spine showed a bony abnormality in the spinal canal at the C2–3 level (Fig. 1). No instability was evident on lateral flexion/extension radiographs. Magnetic resonance images taken before surgery showed that the spinal cord was markedly com-

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Myelopathy caused by invagination of anomalous lamina of the axis

pressed at the C2–3 level, on the right side, by a deeply invaginated anomalous lamina of the axis, corresponding to the intramedullary T2 high-intensity area (Fig. 2). Multiplanar reconstruction of the CT scans revealed that the right side of the axis lamina was deeply invaginated into the spinal canal and revealed a bilateral separation of the C-2 lamina (Fig. 3).

Operation. The patient underwent removal of the anomalous invaginated fragment of the separated lamina of the axis. During surgery, the enlarged C-2 spinous process, the bilaterally separated lamina of the axis, and the defect of the posterior atlantoaxial ligament were observed. The anomalous osseous fragment was found to be somewhat mobile and to be free from the C-2 vertebra. After the bilateral fibrous tissue between the bilaterally separated lamina and the adjacent lamina was released and the right side of the adjacent lamina was partially excavated, this anomalous osseous fragment was removed because it was connected to the C-2 vertebra by fibrous tissue only. After this fragment was removed, the dural tube was decompressed and pulsing well.

Postoperative Course. At 1 year after surgery, the patient was doing well. The spastic gait and clumsiness of his right hand had completely resolved; however, the right dominant sensory disturbance in his trunk and extremities partially remained.

Discussion

Five previously published reports of cervical myelopathy caused by invagination of an anomalous lamina of the axis have been identified1–5 (Table 1). We classified these 6 reported cases into 2 groups according to presence or absence of spina bifida occulta of the axis. Three patients (Cases 1, 3, and 5 in Table 1) had spina bifida occulta, and the other 3 (Cases 2, 4, and 6 [the patient reported here]) did not.1–5

Embryologically, at the 10th week of gestation, 4 posterior vertebral arch chondrification centers unite to form 2 ossification centers on both sides of the vertebra.
Each of the 2 ossification centers forms a pedicle, a lateral mass, and half of the lamina (Fig. 4). The 2 ossification centers fuse posteriorly when the child is 2–3 years of age. The pathogenesis of this rare laminar anomaly could be the failure of the 2 chondrification centers on either side to fuse into a single ossification center, resulting in a bilaterally separated lamina. Moreover, failure of the 2 ossification centers to fuse posteriorly, when associated with failure of the 2 chondrification centers on either side to fuse into a single ossification center, could lead to bilateral separation of the lamina with spina bifida occulta,1,4

Of the 6 reported patients, 5 remained asymptomatic until the 4th, 5th, or 6th decade of their life. Sakai et al. explained that this late onset might be the result of developmental adaptation of the spinal cord. Asakawa et al. considered that the late-onset myelopathy might be the result of complications arising during the aging process, which caused progressive narrowing of the cervical spinal canal and spinal cord compression in a congenitally small spinal canal. Interestingly, for the patients in Cases 2 and 6, lateral flexion/extension radiographs of the cervical spine showed slight movement of the spinous process of the axis. This movement could gradually promote progression of the osseous protrusion into the spinal canal, producing late-onset symptomatic spinal cord compression.

All 6 patients underwent surgical decompression, including removal of the anomalous bony fragment, which resulted in favorable outcomes. Therefore, surgical decompression is an effective treatment choice for cervical myelopathy associated with this condition.

**Table 1: Summary of cases with cervical myelopathy caused by invagination of anomalous lamina of the axis**

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs), Sex</th>
<th>Radiographic Findings</th>
<th>Surgical Treatment</th>
<th>Authors &amp; Year</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>54, M</td>
<td>bilat invaginated lamina &amp; spina bifida occulta of axis</td>
<td>removal of anomalous bone</td>
<td>Koyama et al., 1986</td>
</tr>
<tr>
<td>2</td>
<td>42, M</td>
<td>bilat invaginated lamina of axis</td>
<td>removal of anomalous bone</td>
<td>Kawano et al., 1987</td>
</tr>
<tr>
<td>3</td>
<td>46, M</td>
<td>lat invaginated lamina &amp; spina bifida occulta of axis</td>
<td>removal of anomalous bone &amp; C-3 laminectomy</td>
<td>Asakawa et al., 1999</td>
</tr>
<tr>
<td>4</td>
<td>53, M</td>
<td>bilat invaginated lamina of axis</td>
<td>removal of anomalous bone, C-3 laminoplasty, &amp; partial durotomy</td>
<td>Sakai et al., 2004</td>
</tr>
<tr>
<td>5</td>
<td>16, M</td>
<td>lat invaginated lamina, spina bifida occulta of axis, &amp; hypoplasia of posterior arch of atlas</td>
<td>removal of anomalous bone, C-3 laminotomy, &amp; occipitocervical fusion</td>
<td>Jiang et al., 2010</td>
</tr>
<tr>
<td>6</td>
<td>68, M</td>
<td>lat invaginated lamina of axis</td>
<td>removal of anomalous bone</td>
<td>present study</td>
</tr>
</tbody>
</table>

Fig. 4. Illustration of embryological development of the vertebra. At the 10th week of gestation, 4 posterior vertebral arch chondrification centers (gray) unite to form 2 ossification centers (black) on both sides of the vertebra. Copyright Hironobu Sakaura. Published with permission.

**References**