Concomitant Occurrence of the Ossified Posterior Longitudinal Ligament with a Posterior C2 Defect

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Cervical myelopathy secondary to congenital anomalies involving the posterior elements of the axis (C2) is extremely rare.1 We present a case of deeply invaginated unfused C2 laminae into the spinal canal with associated ossified posterior longitudinal ligament (OPLL) at the C2-C3 level presenting with myelopathy symptoms.

A 34-year-old man presented with a 2-year history of neck pain and recent-onset radiating pain in the left upper limb associated with a tingling sensation. Trauma and other medical comorbidities were ruled out. A neurological examination revealed normal power, intact sensation, and brisk reflexes in all four extremities, with bilateral extensor plantar response. A plain radiograph revealed an abnormality in the C2 posterior elements with a normal atlantodens interval. Magnetic resonance imaging (MRI) revealed circumferential cord compression with canal stenosis caused by the disc osteophyte complex anteriorly and the ligamentum flavum posteriorly at the C2-C3 level presenting with myelopathy symptoms.

A plain radiograph revealed an abnormality in the C2 posterior elements with a normal atlantodens interval. Magnetic resonance imaging (MRI) revealed circumferential cord compression with canal stenosis caused by the disc osteophyte complex anteriorly and the ligamentum flavum posteriorly at the C2-C3 level with features of myelomalacia (►Fig. 1A). Dynamic MRI revealed no worsening or additional level of compression. Computed tomographic scan revealed segmental OPLL at C2-C3 (►Fig. 1B), which was initially thought to be a disc osteophyte complex in MRI and unfused bilaterally invaginated lamina of C2 (►Fig. 1C). Pseudoarthrosis of the lamina with remaining C2 posterolaterally was also observed. The C1 posterior arch and C1-C2 joint were both normal. A surgical laminectomy of C2 and C2-C3 instrumented fusion (C2-pars and C3-lateral mass) were performed. Intraoperatively, the C2 arch was underdeveloped, with hypermobile C2 lamina. Following an uneventful course, the patient was discharged with a hard cervical orthosis for 6 weeks. At the 3-year follow-up, there was a significant improvement in clinical symptoms and adequate decompression with solid fusion was noted in the imaging.

Congenital C2 anomalies usually involve the odontoid process, such as the ossiculum terminale, os odontoideum, or dens hypoplasia. The fusion of two halves of the posterior arch is visible by 2 to 3 years of age (failure to fuse results in the bifid arch), and the fusion of the posterior neural arch to the anterior vertebral body occurs by 7 years of age (failure to fuse results in spondylolysis).2 Failure of the above two embryological events results in the floating lamina. In our case, C2 was not associated with a bifid arch but was unfused posterolaterally, indicating failure of fusion of the posterior arch with the rest of the body.

Posterior element defect of C2 with myelopathy is extremely rare, with only a few cases reported in the literature. Passias et al classified congenital C2 defects into four categories: hypoplasia of the posterior arch with invagination, aplasia of the C2 lamina, and hypertrophy of the C2 arch with an associated upper cervical stenosis.3 In most cases, myelopathy is caused by compression from the posterior aspect, either due to hypertrophy or invagination of the hypoplastic lamina/spinous process. Aplasia of the C2 posterior elements causes progressive instability, resulting in C2-C3 anterolisthesis and dynamic compression.4 Circumferential compression was caused in our case by concomitant...
OPLL and invaginated C2 lamina. The etiology of OPLL is poorly understood and spinal instability, on the other hand, may play a role in the development and progression of OPLL. Our patient was a manual laborer who had to carry heavy weights over his head for an extended period. This could have increased the mechanical load on the most mobile anterior element (C2-C3 discs), resulting in the development of OPLL due to a preexisting deficient posterior element. Considering OPLL as a sign of segmental instability, we performed posterior decompression (C2 laminectomy) in conjunction with C2-C3 fusion.

To conclude, in the absence of instability, as in hypoplastic or hypertrophic lamina/spinous process cases, optimal posterior decompression of abnormal structure is sufficient for a favorable outcome. However, in aplastic posterior elements causing C2-C3 anterolisthesis can require anterior decompression with or without fusion, followed by posterior fusion.

Informed Consent
The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed. The authors declare that written and informed consents have been taken from the patient/ guardian for publication.

Conflict of Interest
None declared.

References