Cervical Myelopathy Caused by Invagination of Floating Anomalous C2 and C3 Laminae in the Spinal Canal

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A 70-year-old man with a 3-year history of weakness in his right upper and lower limbs was admitted to our hospital. Physical examination revealed a positive bilateral Hoffmann sign, positive Babinski sign on the right side, and hyperactive deep tendon reflexes in the upper and lower extremities on both sides. He complained of difficulty using the stairs and chopsticks during the year prior to admission.

Cervical X-ray revealed slight scoliosis and vertebral fusion between C2 and C3 laminae (Fig. 1a, b). Computed tomography scans revealed invagination of the bilaterally separated C2 and C3 laminae into the spinal canal (Fig. 1c-f). The separation occurred on both sides of the C2 lamina; however, the separation in the C3 lamina occurred at the right side of the lamina and the center (spina bifida occulta) (Fig. 1f, g). The floating C2 and C3 laminae were fused to each other, as well as the bilateral C2/C3 facet joints (Fig. 1c-e, g). The fused C2/C3 lateral mass was smaller on the right side than on the left side, and C4 lateral mass was larger on the right side than on the left side (Fig. 1c, e, g). The floating invaginated laminae severely compressed the spinal cord from the right side and produced a high-intensity abnormal signal in the spinal cord at the same level in T2-weighted magnetic resonance images (MRI) (Fig. 1h, i). As for the vertebral artery, the anomaly was not clarified because we did not include contrast-enhanced Computed tomography (CT).

The patient underwent posterior decompression using the conventional open approach. The surface of the C2 to C4 laminae was exposed, and the muscle attachments to the C2 spinous process were detached. We found that the floating laminae were unstable without any firm fibrous union between the lateral residual laminae and could be moved manually in both cephalad and caudal directions. Using a high-speed drill, we removed the left-side residual laminae along the left-side gap. The floating lamina was then rotated posteriorly using the left-side gap as a fulcrum as in an open-door laminoplasty. There were no adhesions between the dura mater and floating lamina, and the right-side gap was abraded manually to remove the floating lamina. We did not use spinal monitoring in this surgery. The patient’s postoperative course was uneventful; he reported no axial pain and made a rapid recovery within 3 months (Fig. 2a-d).

C2 anomaly is rare, unlike defects of the posterior atlas. In 1986, Koyama et al. first reported a separated C2 lamina that invaginated into the cervical spinal canal. Only a few reports of a C2 anomaly in isolation have been published. Some C2 anomalies have been reported combined with ossification of the posterior longitudinal ligament, atlas anomaly, or C7 anomaly. However, simultaneous C2 and C3 anomalous separated laminae have not been reported. The pathogenesis of this anomaly has been described in detail. Briefly, there are four chondrification centers in the posterior vertebral arch at week 10 of gestation, and failure in the unification of these centers can cause separated lamina and/or spina bifida occulta.

In case studies, it has been suggested that chronic movement and gradual protrusion of the anomalous bone fragment may contribute to myelopathy. Bilateral gaps between anomalous floating lamina and base lamina might induce stress because the floating lamina attaches to several muscles. Spur formation can occur at the gaps and may cause stenosis of the spinal canal. Here, the patient perceived his symptoms for the first time in his 60s. These C2 anomalies are treated by removing the anomalous spinous
process\textsuperscript{9,10} and undergoing fusion surgery in cases involving instability between vertebrae\textsuperscript{4,5}. These surgical outcomes are usually satisfactory. We performed resection of the invaginated separated C2 and C3 laminae. MRI 1 month postoperatively confirmed sufficient decompression (Fig. 2a, b).

The potential problem of an operation involving resection of the C2 spinous process is impairment of the attachments of several muscles, including the obliquus capitis inferior and rectus capitis muscles, which can cause axial pain or cervical kyphosis. However, Moon et al. suggested that the anomalous lamina does not play a major role in the stability of the cervical spine\textsuperscript{7}. The separated C2 and C3 laminae in our patient were also unstable, which indirectly supports this suggestion. We considered that removing the anomalous bone fragment would be reasonable. Although the short-term clinical outcome was satisfactory for this patient, a careful long-term follow-up is needed.

Conflicts of Interest: The authors declare that there are no relevant conflicts of interest.

Author Contributions: TN, BO, TS, and KM performed the operation, and BO, SF, and SM decided on the treatment strategy. All authors read and approved the final manuscript.

Informed Consent: Written informed consent was ob-
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