Upper cervical cord compression due to a C-1 posterior arch in a patient with ossification of the posterior longitudinal ligament and a kyphotic cervical spine in the protruded-head position

Case report

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In this paper the authors report the case of a patient with ossification of the posterior longitudinal ligament (OPLL) below the axial vertebra (C-2) at the kyphotic cervical spine, with an atlas vertebra (C-1) posterior arch that compressed the spinal cord with the head in a pathognomonic position, similar to a protruded position. This condition appears to be very rare. The morphological findings between the kyphotic cervical spine and OPLL, the upper occipitocervical junction, and the protruded-head position are discussed. A 40-year-old man presented with severe pain radiating to both legs when he yawned, sneezed, or extended his jaw (a protruded-head position). A kyphotic cervical spine with OPLL below C-2 was observed using CT and radiography, yet sagittal T2-weighted MRI failed to identify abnormal findings in a neutral or extension position, except for a slight cervical canal stenosis. However, in a pathognomonic protruded-head position, sagittal T2-weighted MRI showed a C-1 posterior arch that severely compressed the spinal cord at the upper cervical level. Therefore, the authors believe that the severe pain radiating to both legs was caused by a spinal canal stenosis due to a C-1 posterior arch impingement. The C-1 posterior arch was resected, and after the surgery, the patient indicated that the intolerable pain had disappeared. In conclusion, in patients with OPLL and a kyphotic cervical spine, the authors propose that the pathognomonic protruded position is valuable for estimating disrupted compensatory mechanisms at the upper cervical junction.

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Key Words • C-1 posterior arch • kyphotic cervical spine • ossification of the posterior longitudinal ligament • pathognomonic protruded-head position

Case Report

History and Examination. A 40-year-old man reported experiencing a severe pain radiating to both legs when he yawned or sneezed. He was afraid to do either because this pain gradually became intolerable and appeared easily. He visited another hospital where he was conservatively treated with external stabilization of the neck and medication. One month later the patient’s physician recommended posterior cervical laminoplasty at
C3–6 because his symptoms had not eased. However, the patient hoped to obtain a second opinion from another doctor and was referred to our hospital.

A physical examination identified a severe radiating pain (visual analog scale score of 88) to the legs when he protruded his jaw and very slight paresthesia in both hands. The patient did not have motor weakness or clumsiness in his hands and feet and was without hyperreflexia or hyporeflexia. There were no clinical or laboratory findings of rheumatoid arthritis, and he had no history of head or neck trauma.

The radiographs of the cervical spine showed a kyphotic alignment (C2–7 Cobb angle of −22.5°) in a neutral position (Fig. 1A). The flexion-extension radiographs indicated that the occipitoatlantal (occiput-C1) angles were −4.5° and −2.3° in flexion and extension, respectively, and that the atlantoaxial (C1–2) angles were 34.6° and 46.3° in flexion and extension, respectively (Fig. 1B and C). The patient refused radiography in the protruded-head position because of the beginning of severe pain. Computed tomography revealed that there was a continuous OPLL at C-2 and C-3 and segmental OPLL from C-3 to C-5, with a central-type OPLL at the cervical spine. The maximum space available for the spinal cord was 6.4 mm at the C-2 lesion, and the maximum spinal canal compromise was 42% (Fig. 2) at the site of the same lesion. Sagittal T2-weighted MRI failed to demonstrate an abnormality of the upper cervical lesion in a neutral or extension position (Fig. 3A and B). However, in a pathognomonic head position, such as with a protruded jaw, the sagittal T2-weighted MRI showed that the C-1 posterior arch severely compressed the spinal cord at the upper cervical level (Fig. 3C). We believed that the severe pain radiating to the legs was caused by a spinal canal stenosis due to a hyperlordotic C-1 posterior arch impingement, with OPLL below C-2 at the kyphotic cervical spine.

Operation and Postoperative Course. The patient was placed prone with his neck in flexion, and the C-1 posterior arch was resected. No complications occurred during the perioperative period. Magnetic resonance imaging and CT after surgery indicated that the spinal cord had been decompressed in the protruded position (Fig. 4). The postoperative radiographs of the cervical spine showed an improved kyphotic alignment (C2–7 Cobb angle of −16.3°) in a neutral position (Fig. 5A). The flexion-extension radiographs indicated that the occipitoatlantal (occiput–C1) angles did not deteriorate (−7.7° and 0.5° in flexion and extension, respectively), and that the atlantoaxial (C1–2) angles were 34.4° and 45.6° in flexion and extension, respectively (Fig. 5B and C). After the surgery, the patient reported that the intolerable pain had disappeared (visual analog scale score of 13) without any new neurological deficit for 13 months.

Discussion

We documented an unusual patient with a hyperlordotic C-1 posterior arch compressing the upper cervi-
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Fig. 3. Sagittal T2-weighted MR images (lower row) and corresponding head positions (upper row). A and B: Abnormalities of the upper cervical lesion were not detected on T2-weighted MRI in the neutral (A) or extension (B) positions. C: The spinal cord compression at the upper cervical level caused by the C-1 posterior arch was evident in a protruded-head position, which included the patient extending his jaw.

Fig. 4. Postoperative sagittal T2-weighted MR image in the protruded-head position (A), and postoperative axial CT scan at C-1 (B).

cal spinal cord, with OPLL below the C-2 position in a kyphotic cervical spine. Although typical MRI failed to demonstrate an abnormality of the upper cervical cord in a neutral or extension position, placing the patient in a protruded-head position induced the severe pain radiating to the lower limbs, and the sagittal T2-weighted MRI showed that the C-1 posterior arch severely compressed the spinal cord at the upper cervical level. Previously, Kawabori et al.8 presented a case in which C-1 posterior tubercle impingement resulted in spinal cord compression in the head extension position, with diffuse idiopathic skeletal hyperostosis. Although that case closely resembled ours, we observed a C-1 posterior arch with OPLL below C-2 in a kyphotic cervical spine that compressed the spinal cord and caused nonspecific myelopathy in a protruded-head position but not during head extension.

The mechanism of upper cervical spinal cord compression by the C-1 posterior arch is very interesting. We suspect that the failure of the compensatory mechanism at the upper cervical level is the principal reason for the patient’s symptoms. Furthermore, we believe that 3 reasons can explain why the compensatory mechanism was disrupted.

First, hyperlordosis at C1–2 occurred to compensate for the cervical kyphosis, and consequently, the compensatory mechanism of the C1–2 junction was unable to withstand the excess hyperlordosis at C1–2. Regarding the C1–2 lesion, Ordway et al. demonstrated that the C1–2 excursion was $6.2^\circ \pm 6.1^\circ$ and $6.2^\circ \pm 5.0^\circ$ in flexion and extension in normal subjects, respectively. In our case, the C1–2 angles were $34.6^\circ$ and $46.3^\circ$ in flexion and extension, respectively. It is likely that the patient became hyperlordotic at C1–2 to compensate for the subaxial cervical kyphotic alignment. Chikuda et al. reported that the ossification of the ligament or ankylosis at the subaxial spine placed excessive stress on the C1–2 complex due to the altered biomechanics of the cervical spine, which resulted in damage to the surrounding tissue or ligaments and instability at the upper cervical lesion. Matsumoto et al. demonstrated that kyphotic stability from C-3 to C-7 resulted in compensatory hyperlordosis at the upper cervical junction. Our case presented with kyphosis...
and OPLL below C-2, which is similar to these findings. Therefore, the kyphotic cervical spine with OPLL may have been one of the mechanical factors increasing hyperlordosis at C1–2.

Second, the range of motion at the occiput–C1 junction disappeared in the flexion (−4.5°) and extension (−2.3°) positions. Therefore, the occiput–C1 junction was unable to compensate for the hyperlordosis at C1–2, whereas the movement of the occiput–C1 joint was able to compensate for the risk of progressive kyphosis in the subaxial cervical spine.16 The occipitoatlantoaxial joint accounts for 45° of flexion and extension.13 However, in our case, the movement almost disappeared, and it was therefore impossible for the compensatory mechanism of the occiput–C1 joint to function.

Lastly, when the patient was placed in a protruded-head position, the additional lordotic force increased on C1–2, and consequently the disrupted compensatory mechanism resulted in spinal cord compression at the upper cervical lesion. Ordway et al. reported that a greater range of motion at C1–2 was observed in the protruded position in comparison with full-length extension positions.12 For all of these reasons, it is easy to propose that the stress placed on C1–2 was too great and that the compensatory mechanism was disrupted when the patient was placed in a protruded-head position, such as jaw projection.

The resection of the C-1 posterior arch was performed without C1–2 fusion. We also considered C1–2 fusion surgery, but did not select it for 2 reasons. First, one of the patient’s hobbies is golf, and he did not want fusion surgery at the upper cervical level because C1–2 fusion induces restriction of neck rotation. Hott et al. demonstrated that C1–2 fusion induced more than 50% rotational restriction in comparison with normal subjects. Second, there was the possibility that C-1 would not be reduced for C-2 in the flexion position because C1–2 would maintain a hyperlordotic angle (34.6°). If C1–2 fusion surgery was performed with the remaining C1–2 hyperlordosis condition, the kyphotic change at the subaxial level would continue. Yoshihito et al. and Yoshida et al. reported that fixation of the atlantoaxial joint in a hyperlordotic position will result in kyphotic sagittal alignment of the subaxial level. Therefore, we performed resection of the C-1 posterior arch with careful follow-up. Although the patient was young (40 years of age), typical myelopathy may appear and progress in the future. A close patient follow-up evaluation is therefore required in this case.
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Conclusions

In conclusion, taken together, our findings suggest that our case is different from those previously reported. In patients with OPLL and a kyphotic cervical spine, it is very important to use the pathognomonic protruded-head position to estimate disrupted compensatory mechanisms at the upper cervical junction.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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