Occipital Erosion as a Late Complication Following Atlantoaxial Fixation: A Case Report

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Several pathologies, including a congenital disease, inflammatory disorder, and trauma, have been reported to cause atlantoaxial instability\(^1\). Such conditions can lead to pain in the occipito-cervical region or cervical myelopathy due to both static and dynamic factors and require atlantoaxial fixation to correct the deformity and provide stability. In recent years, posterior atlantoaxial fixation using C1 lateral mass screws and C2 pedicle screws has become increasingly popular as it provides excellent stability. While several studies have referred to complications following this procedure\(^1\)-\(^3\), few reports have demonstrated occipital erosion induced by the protrusion of spinal instrumentation\(^4\)-\(^8\). In this study, we present a case in which a patient developed occipital erosion following atlantoaxial fixation and review the relevant literature.

A 72-year-old woman with a 17-year history of rheumatoid arthritis presented with persistent neck pain. Her rheumatism was Steinbrocker grade IV, and she was taking 4 mg of methotrexate per week and 500 mg of abatacept every four weeks. Her radiographs demonstrated an atlantodental interval of 5 mm, Ranawat value of 11.2 mm, and subaxial subluxation at C3-C4 with kyphotic deformity. In the dynamic radiographs, the anterior slips of C1 and C3 were 5 and 1 mm, respectively (Fig. 1), and the instability causing the pain seemed to be due to C1-C2. Preoperative angiography showed no evidence of anomalous vertebral artery. She also had osteoporosis with bone mineral density of 0.683 (T score, \(-1.6\)) in the proximal femur measured using dual-energy X-ray absorptiometry. Based on the assessment that her symptom was caused from atlantoaxial instability, posterior atlantoaxial fixation was performed using C1 lateral mass and C2 pedicle screws with bone grafting from iliac bone to C1-C2 lamina (Fig. 2). The postoperative course was uneventful; however, she was lost to follow-up after 5 months.

At three years after the operation, she was referred to our institution again with occipital pain and tinnitus. Physical findings, such as hyperreflexia and muscle weakness of the four extremities, suggested the progression of cervical myelopathy. Radiography and computed tomography of the cervical spine demonstrated bilateral broken C1 screws, the progression of subaxial kyphosis with C3-C4 instability, an increased occipitoaxial angle, and occipital bone erosion induced by the protruding rods (Fig. 3, 4A-C). MRI demonstrated multiple levels of cervical canal stenosis (Fig. 4D). Occipitothoracic fusion surgery was considered, but we decided to perform only C2-C6 decompression and fusion surgery with removal of the rods and C1 screwhead because of her several comorbidities (Fig. 4E). When the rods and screwheads were removed, we confirmed that the sharp-cut side of the rods had been facing the occipital bone. Although the inner cortex of the occipital bone was breached, there was no dural defect or cerebrospinal fluid leakage. Her occipital pain and tinnitus immediately resolved after the revision surgery, and her other neurological symptoms gradually improved.

Posterior atlantoaxial fixation is associated with the potential risk of various specific complications, including massive bleeding, vertebral artery injury, and hypoglossal nerve palsy\(^9\). Erosion in the occipital bone caused by the abutment of spinal instrumentation is a relatively rare complication; nine cases have been previously reported (Table 1)\(^1\)-\(^9\). One of the previous reports demonstrated migration of a rod into the brain through the skull without any neurological
Figure 1. Images before the first operation. (A) An X-ray of the global sagittal alignment of the whole body in standing position. (B) (C) X-rays (lateral view) of cervical flexion and extension. The X-rays show atlantoaxial subluxation, vertical subluxation, subaxial subluxation, and kyphotic alignment. (D) T2-weighted MRI.

Figure 2. Images after the first operation. (A) (B) X-ray and CT after atlantoaxial fusion using a C1 lateral mass screw and C2 pedicle screw. (C) CT showing bone graft on laminas of C1–C2.
Figure 3. Postoperative X-rays showing the progression of subaxial kyphosis and an increase in the O–C2 angle. C1 screw breakage was recognized at 3 years and 4 months after the first operation. O–C2, occipitoaxial angle; C2–C7, C2–C7 angle

Figure 4. (A) (B) X-rays (lateral view) of cervical flexion and extension before the second operation. (C) CT before the second operation showing the erosion of the occipital bone. (D) T2-weighted MRI before the second operation showing C3 subluxation and canal stenosis. (E) X-ray after the second operation.

signs or symptoms. In another case, a penetrating rod caused cerebellar hemorrhage. To prevent such occipital complications, Nakao et al. recommended the avoidance of cranial rod protrusion. Based on the operative finding that the sharp-cut edge of the rod had been facing the occipital bone in our case, we consider that caudally placing the sharp-cut side of the rod would also be useful for preventing this complication.

In the present case, the occipitoaxial angle had increased with progressive subaxial kyphosis after the initial operation. In this case, compensation for subaxial kyphotic alignment to maintain horizontal gaze possibly caused an increased O-C2 angle, which resulted in contact between the rod and the occipital bone. Atlantoaxial fixation reportedly produces subaxial kyphotic sagittal alignment. In addition to the care required to detect such subaxial kyphotic progression, our case highlighted that surgeons should pay attention to the occipital region during postoperative follow-up following...
Table 1. Summary of Previous Reports.

| Authors and year       | Age | Sex | Cause of erosion          | Symptoms                      | Duration until the second surgery | Procedure of the second surgery                  |
|-----------------------|-----|-----|----------------------------|--------------------------------|-----------------------------------|-------------------------------------------------|
| Plant et al., 2010    | 13  | M   | Rod                        | Neck pain                     | 3 years                           | Removal, autologous bone grafting, halo jacket   |
| Oh et al., 2014       | 43  | M   | Rods                       | Occipital pain                | 24 weeks                          | Screw and rod replacement                       |
| Nakao et al., 2014    | 70  | M   | Rods                       | Headache                      | 6 months                          | Rod replacement                                 |
| Arizumi et al., 2015  | 14  | M   | Supralaminar hook          | Occipitalgia                  | 3 years and 3 months              | Removal                                         |
|                       | 61  | M   | Lateral mass screw         | Occipital crepitus            | 1 year and 2 months               | Removal                                         |
|                       | 62  | F   | Supralaminar hook          | Occipital crepitus            | 1 year and 2 months               | Removal                                         |
|                       | 72  | F   | Alras craw hook            | Occipitalgia                  | 8 years                           | Removal                                         |
| Miyaoka et al., 2017  | 81  | F   | Rods                       | Upper neck pain and occipital headaches | 1 month                           | Rod replacement                                 |
| Kobets et al., 2019   | 68  | F   | Rods                       | Headache, neck pain, and vomiting | 4 years                           | Rod cut                                         |
| Our case              | 72  | F   | Broken screw and rods      | Occipital pain and tinnitus   | 3 years and 9 months              | Rod removal, C2–C6 fixation, and decompression   |

Regarding surgical strategy for atlantoaxial fixation, Magerl method as a transarticular screw fixation would be an alternative option. If the occipital bone is close to C1 posterior arch as this case, the available space for the head of C1 lateral mass screws will be small, which may be the limitation for C1 lateral mass screw and C2 pedicle screw fixation.

In conclusion, we experienced a case in which the erosion of the occipital bone occurred after C1 lateral mass screw and C2 pedicle screw fixation. In addition to the cranially protruded rod with sharp-cut edge, an increase in the O-C2 angle as compensation for progressive subaxial kyphosis seemed to be the cause of this complication. Special care should be taken during the follow-up of patients with cervical kyphotic deformity who undergo atlantoaxial fixation.

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Author Contributions: Satoshi Nagatani wrote and prepared the manuscript. All authors participated in the study design. All authors have read, reviewed, and approved the article.

Ethical Approval: This study does not require an approval from IRB because it involves no data analysis or testing of a hypothesis.

Informed Consent: Informed consent was obtained from the study participant.

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