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[1-3]. 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)[4-9]. One of the previous reports demonstrated migration of a rod into the brain through the skull without any neurological

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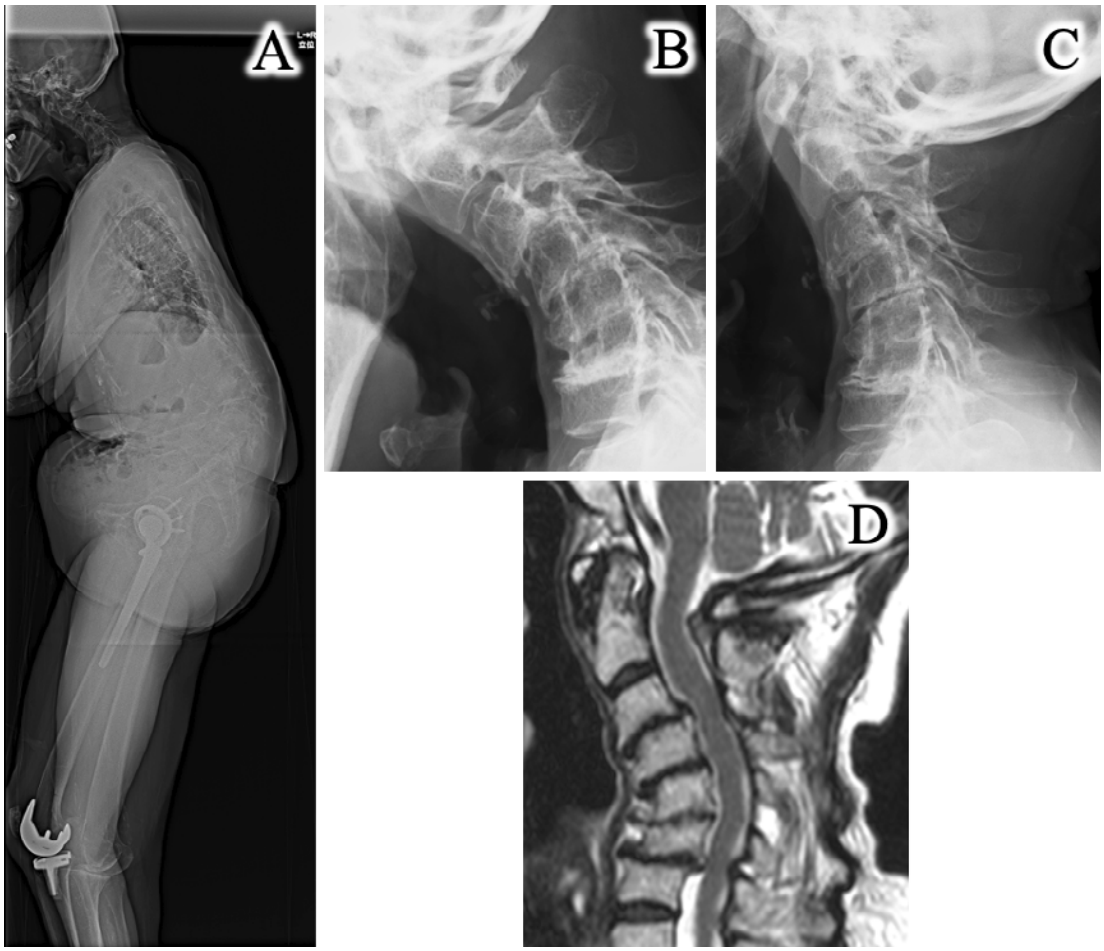

Figure 1. Images before the first operation. (A) An X-ray of the global sagittal alignment of the whole body in standing position. (B) (C) X-rays (lateral view) of cervical flexion and extension. The X-rays show atlanto-axial subluxation, vertical subluxation, subaxial subluxation, and kyphotic alignment. (D) T2-weighted MRI.

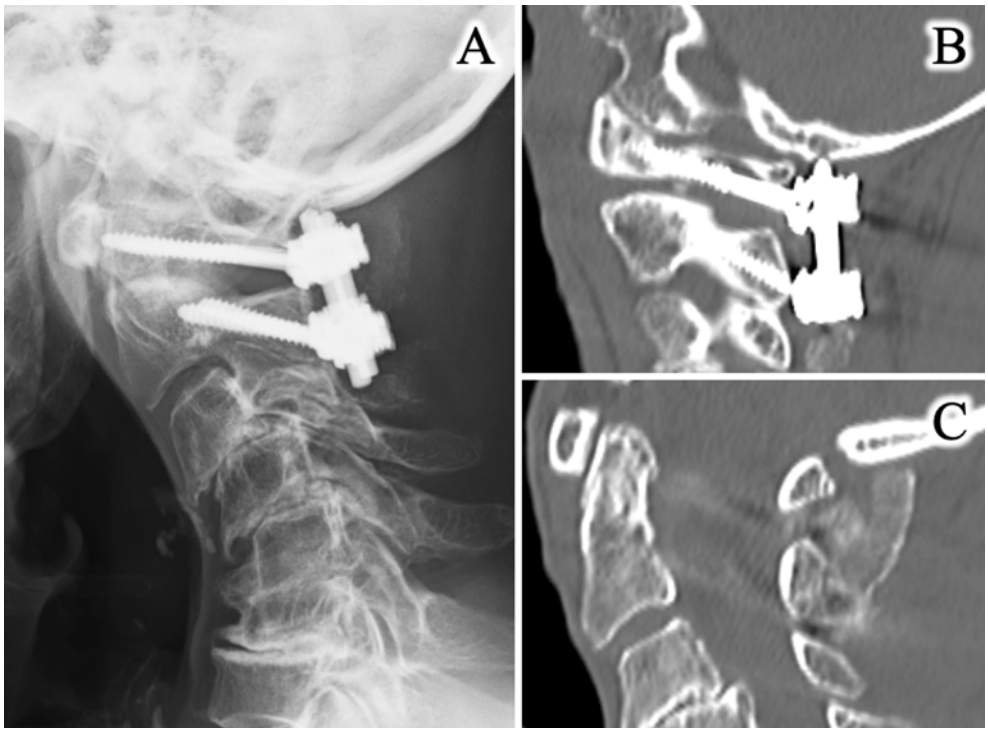

Figure 2. Images after the first operation. (A) (B) X-ray and CT after atlantoaxial fusion using a C1 lateral mass screw and C2 pedicle screw. (C) CT showing bone graft on laminas of C1–C2.

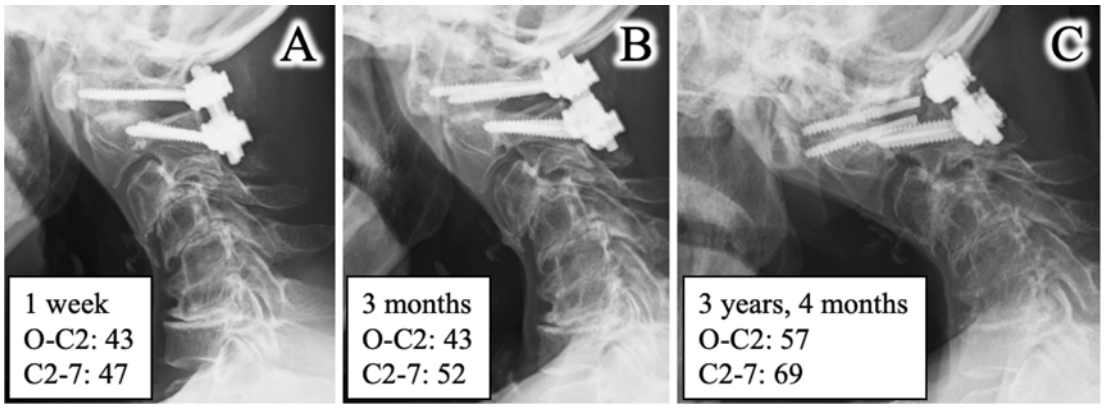

Figure 3. Postoperative X-rays showing the progression of subaxial kyphosis and an increase in the O–C2 angle. C1 screw breakage was recognized at 3 years and 4 months after the first operation.
O–C2, occipitoaxial angle; C2–C7, C2–C7 angle

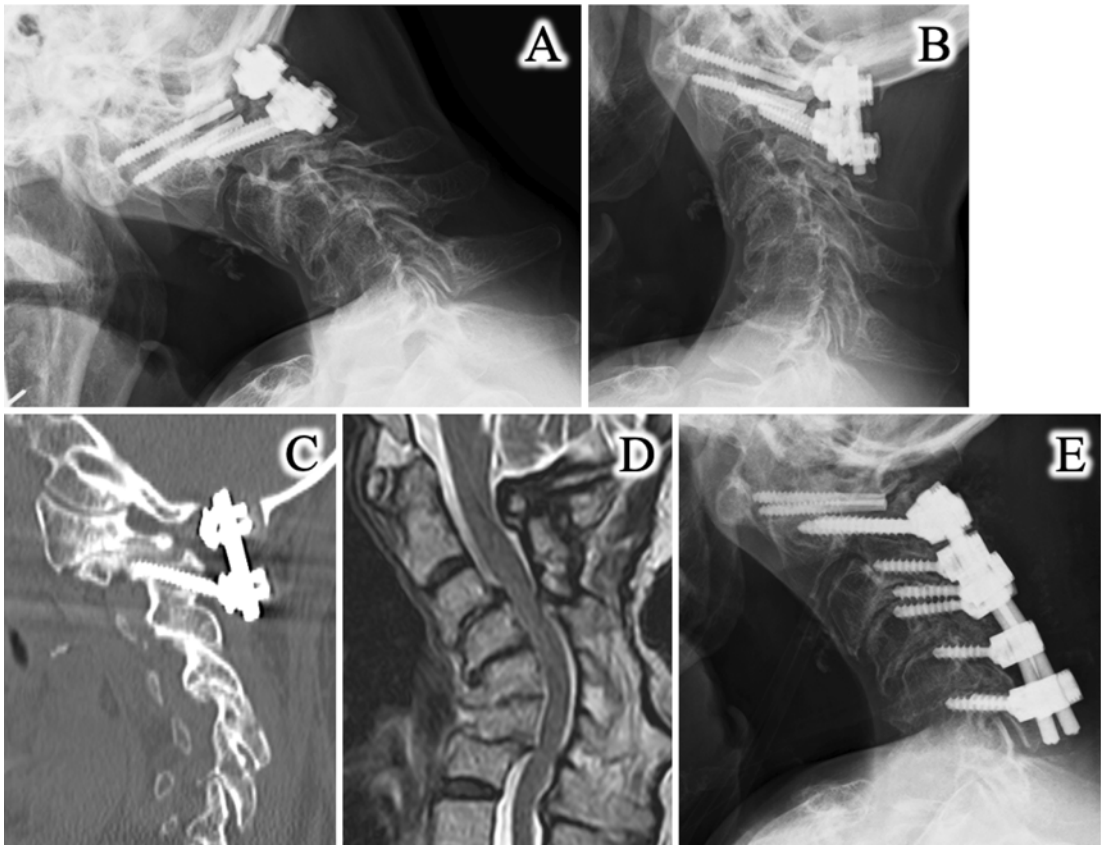

Figure 4. (A) (B) X-rays (lateral view) of cervical flexion and extension before the second operation. (C) CT before the second operation showing the erosion of the occipital bone. (D) T2-weighted MRI before the second operation showing C3 subluxation and canal stenosis. (E) X-ray after the second operation.

signs or symptoms[4]. In another case, a penetrating rod caused cerebellar hemorrhage[8]. To prevent such occipital complications, Nakao et al. recommended the avoidance of cranial rod protrusion[6]. 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[10]. 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

atlantoaxial fixation.

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.

References

1. Yan L, He B, Liu T, et al. A prospective, double-blind, randomized controlled trial of treatment of atlantoaxial instability with C1 posterior arches >4 Mm by comparing C1 pedicle with lateral mass screws fixation. BMC musculoskelet Disord. 2016;17:164.
2. Lee CH, Hong JT, Kang DH, et al. Epidemiology of iatrogenic vertebral artery injury in cervical spine surgery: 21 multicenter studies. World Neurosurg. 2019;126:e1050-4.
3. Hong JT, Lee SW, Son BC, et al. Hypoglossal nerve palsy after posterior screw placement on the C-1 lateral mass. Case report. J Neurosurg Spine. 2006;5(1):83-5.
4. Plant JGA, Ruff SJ. Migration of rod through skull, into brain following C1-C2 instrumental fusion for os odontoideum: a case report. Spine (Phila Pa 1976). 2010;35(3):E90-2.
5. Oh CH, Ji GY, Seo HS, et al. Repeated complication following atlantoaxial fusion: a case report. Korean J Spine. 2014;11(1):7-11.
6. Nakao Y, Shimokawa N, Morisako H, et al. Late complication of surgically treated atlantoaxial instability: occipital bone erosion induced by protruded fixed Titanium rod: a case report. J Chiropr Med. 2014;13(4):278-81.
7. Arizumi F, Moriyama T, Tachibana T, et al. Erosion in the occipital bone caused by the fixation instrument used for posterior atlantoaxial fusion -report of 4 cases-. Springerplus. 2015;4(1):1-5.
8. Miyaoka Y, Takeuchi M, Wakao N, et al. Contusional cerebellar hemorrhage related to placement in the protrusion position after atlantoaxial fusion. World Neurosurg. 2017;101:278-81.
9. Kobets AJ, Nakhla J, Jwland Y, et al. Occipital erosion and pain from excess superior rods as a complication of atlantoaxial fusion. Open Sci Publ J Radiol. 2019;1:1-3.
10. Yoshida G, Kamiya M, Yoshihara H, et al. Subaxial sagittal alignment and adjacent-segment degeneration after atlantoaxial fixation performed using C-1 lateral mass and C-2 pedicle screws or transarticular screws. J Neurosurg Spine. 2010;13(4):443-50.