



Spontaneous Anterior Atlas Fracture Following C1 Laminectomy without Fusion: A Case Report

Seong-Gon Kim, Myung-Hoon Shin, Jong-Tae Kim, Du-Yong Choi

Department of Neurosurgery, Incheon St Mary's Hospital, College of Medicine, The Catholic University, Incheon, Republic of Korea

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Corresponding author:

Myung-Hoon Shin

Department of Neurosurgery, Incheon St Mary's Hospital, College of Medicine, The Catholic University of Korea, 56 Dongsu-ro, Bupyeong-gu, Incheon 21431, Republic of Korea
Tel: +82- 32-280-5973
Fax: +82-32-280-5991
E-mail: novice97@naver.com

C1 laminectomy without fusion (CLWF) is a widely accepted minimally invasive surgical technique to decompress the spinal cord around the craniovertebral junction. In the case presented herein, a 46-year-old man experienced a fracture of the anterior arch of the atlas that occurred during the postoperative period following CLWF to treat cervical myelopathy associated with a retro-odontoid pseudotumor. The findings demonstrate that disruption of the integrity of the posterior arch of the atlas confers an increased risk of anterior arch fracture. Although CLWF is a good surgical option for treating stable cervical myelopathy associated with a retro-odontoid pseudotumor, posterior decompression and fusion could be beneficial for patients with a relatively high inferior facet angle of C1-2 and subaxial arthrodesis.

Keywords: Atlas fracture; C1 laminectomy; Retro-odontoid pseudotumor

INTRODUCTION

C1 laminectomy without fusion (CLWF) is a widely accepted minimally invasive surgical technique to decompress the spinal cord around the craniovertebral junction. It is performed as a surgical intervention for Arnold-Chiari malformation, intracanal tumor at the level of C1, ossification of the posterior longitudinal ligament developing cranially above the C1 level, and retro-odontoid pseudotumor without instability¹⁾. Choi et al.²⁾ have reported clinical and radiological results of CLWF performed for the treatment of various pathologies of the craniovertebral junction. During

41 months of the follow-up period, the mean visual analog scale and Ranawat scale were significantly improved postoperatively without any resultant instabilities noted. Takemoto et al.¹⁾ have also demonstrated favorable outcomes following CWLF for ten patients with retro-odontoid pseudotumor, defined as a non-neoplastic soft tissue mass. Observing no progression of atlantoaxial instability (AAI) and concomitant regression of retro-odontoid mass, they claimed that CLWF was beneficial, especially for elderly patients given risks of other surgical options using a posterior fusion.

Although the CLWF technique has been reported to have several advantages as listed above, there are still concerns

about iatrogenic disruption of the posterior arch and its associated muscle and ligament structures that are essential supportive components of the craniovertebral junction. Thus, we contented that patients who had undergone CLWF might have an increased load onto the anterior C1 arch and a propensity for associated arch fracture. Here, we present a case of an anterior atlas fracture following C1 laminectomy for treating a retro-odontoid pseudotumor.

CASE REPORT

A 46-year-old man without significant medical history present with clumsiness of the hand and gait disturbance that gradually deteriorated over a month. Four years ago, he underwent an anterior cervical interbody fusion at levels of

C3-4 and C6-7. He reported that previous symptoms such as left-arm radiating pain with paresthesia disappeared post-operatively. A neurological examination conducted at this time revealed diminished hand grip power. The Hoffman sign was positive for bilateral hand. Radiologic examination revealed an increase of atlantodental interval (ADI) of 4 mm on a neutral cervical image (Fig. 1A). However, AAI was not found on dynamic images (Fig. 1B, C). Magnetic resonance (MR) imaging demonstrated spinal cord compression due to a retro-odontoid pseudotumor accompanied by high signal intensity of spinal cord on a T2-weighted image (Fig. 1D, E). Preoperative computed tomography (CT) scan showed no fracture (Fig. 1F). Although the patient was found to have increased ADI, he elected to undergo CLWF considering facts of no sign of AAI and relatively young age. The patient

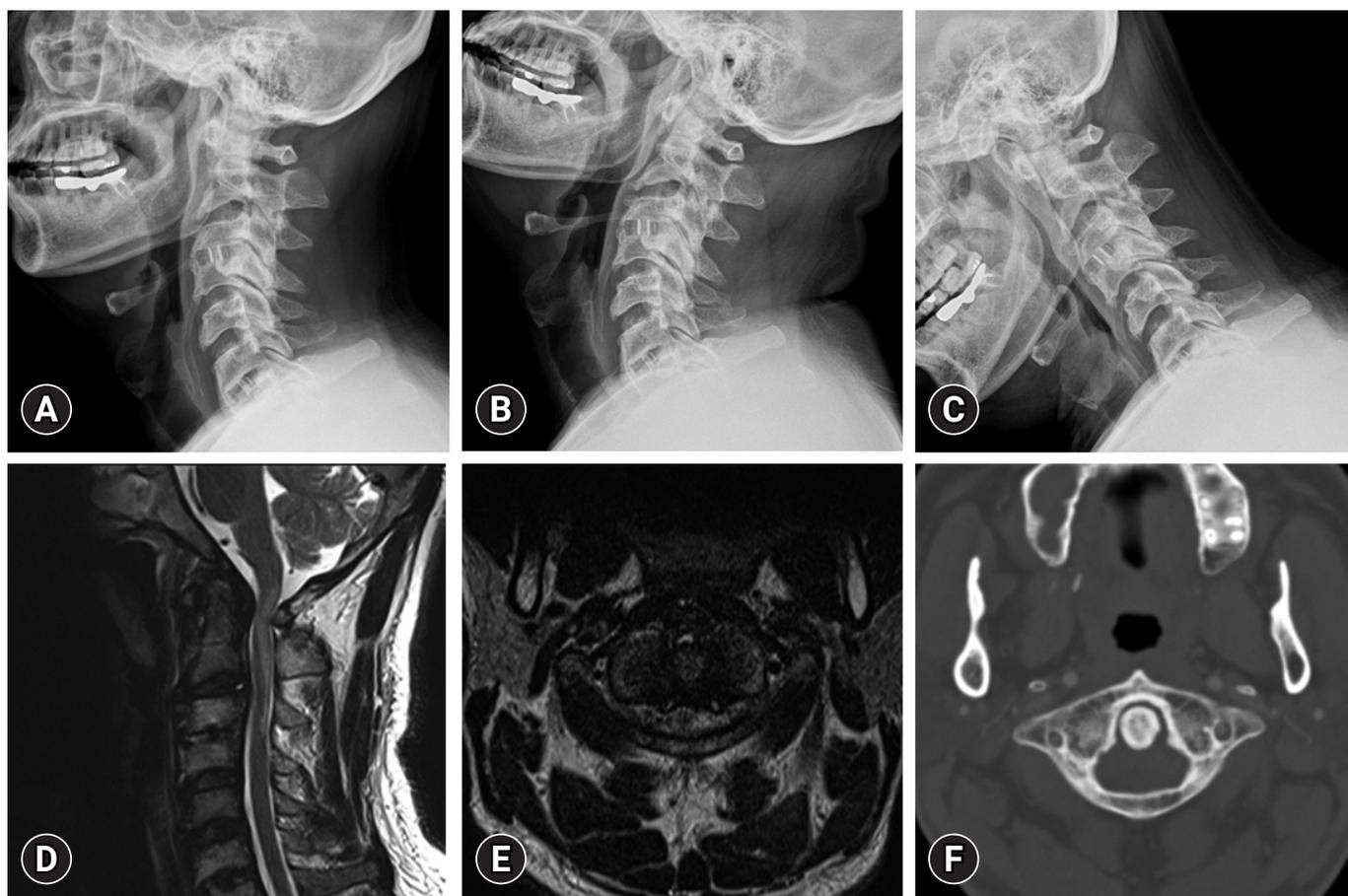


Fig. 1. (A) Preoperative lateral cervical radiograph showing an atlantodental interval of 4 mm and arthrodesis at C3-4 and C6-7. (B, C) Preoperative dynamic views showing no atlantoaxial instability. (D, E) Preoperative T2-weighted magnetic resonance imaging showing spinal cord compression with high signal intensity due to a retro-odontoid pseudotumor. (F) Preoperative computed tomography scans showing no fracture of the anterior arch of the atlas.

underwent an uneventful C1 laminectomy (Fig. 2A). The surgical process was as follows. Following a longitudinal skin incision, the nuchal ligament was bisected in the middle to single out the C2 spinous process. The posterior neck muscles attached to the upper portion of C2 spinous process were partially dissected. The C1 posterior arch was cut out at a distance of approximately 20 mm following dissecting muscles and inserted into the surface (rectus capitis posterior minor). Postoperatively, he experienced noteworthy improvement of neurological disturbances in his upper and lower limbs. On postoperative 10 days, he left the hospital.

He was asked to wear a soft neck collar for a month. Eight months postoperatively, he came back to the outpatient clinic complaining of a sudden onset of intolerable neck pain, which was aggravated on axial neck rotation. He reported no obvious inciting event. Flexion-extension cervical spine radiographs demonstrated no obvious spinal instability. A displaced anterior C1 fracture, which was not clearly detectable on plain X-ray films, was revealed on CT scan (Fig. 2C). The patient was initially treated conservatively for a month with a hard cervical collar and oral narcotic agent. Given his unabated pain and displaced bone frag-

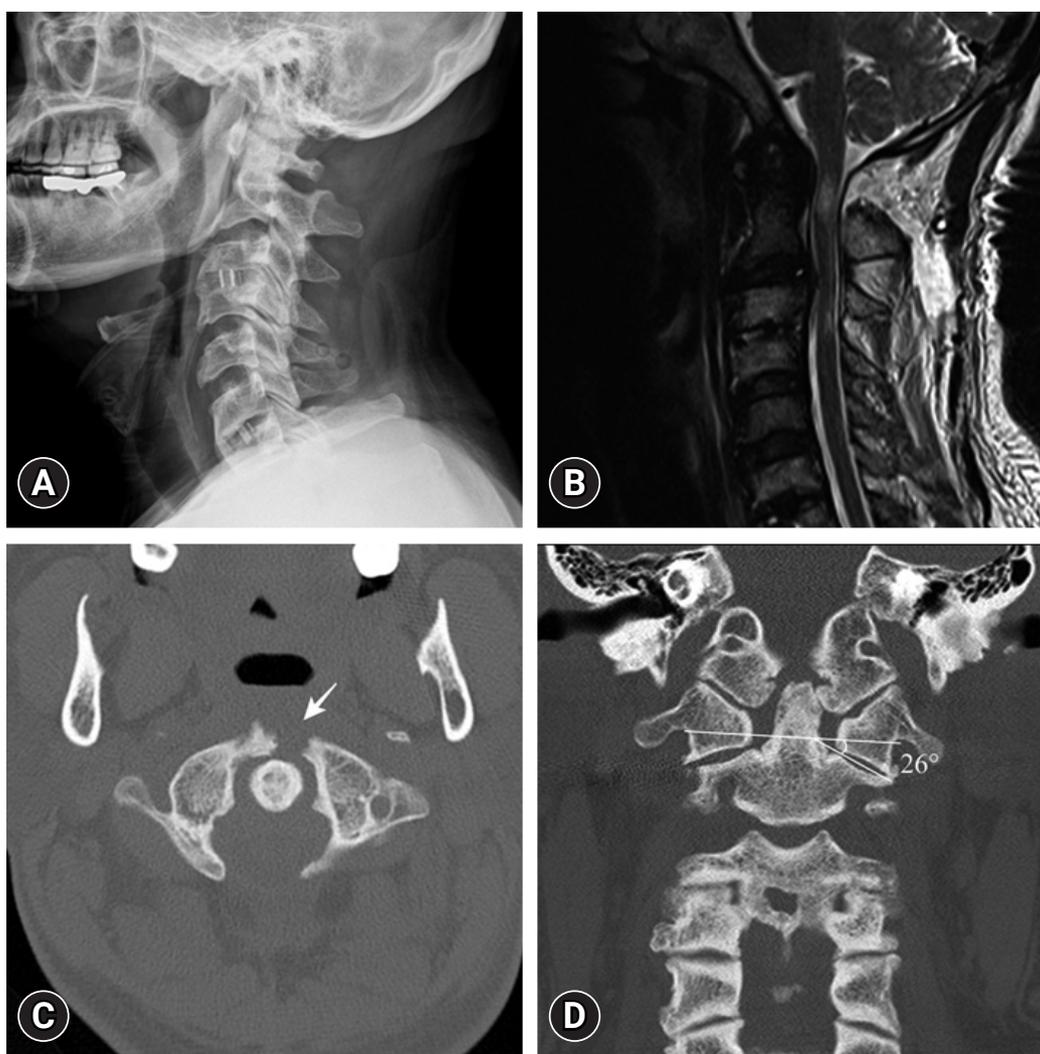


Fig. 2. (A) Lateral cervical radiograph following C1 laminectomy without fusion (CLWF). (B) Postoperative T2-weighted magnetic resonance imaging showing the removed posterior C1 arch and resultant decompressed spinal cord. (C) Computed tomography (CT) scans obtained 8 months after CLWF showing fracture of the anterior arch (white arrow). (D) Coronal view of the CT scan demonstrating a large inferior facet angle of 26°.

ment, internal surgical fixation was considered appropriate. The patient underwent posterior cervical fusion involving the placement of C1 lateral mass screws and C2 pedicle screws (Fig. 3A). The procedure was performed without any complications. Shortly after the operation, the patient experienced profound relief from his preoperative cervical pain. Moreover, follow-up MR images of the cervical spine revealed a decreased size of retro-odontoid pseudotumor 3 months postoperatively (Fig. 3B).

DISCUSSION

Fractures of the C1 vertebra have been estimated to account for 10% of cervical injuries and 1 to 3 % of all spinal column injuries^{4,5,7,10}. Atlas fracture is mostly caused by falls and motor vehicle accidents. It hardly leads to a neurological compromise largely because of the roomy canal diameter and centripetal spreading of the C1 ring following an axial loading force⁸. In 1927, Jefferson⁶ described details of fractures involving the C1 vertebra. In his initial manuscripts, he attested that structures including the anterior arch, posterior arch, and transverse ligament were paramount in absorbing an axial load and enduring radial shift of the lateral masses.

Unlike typical C1 fractures wherein the mechanism of injury is axial loading, a C1 fracture following C1 laminectomy might not be attributed to this. Interruption of the circular architecture of the atlas can contribute to the concentration of mechanical stress in the anterior arch, resulting in a spontaneous fracture without inciting trauma. Shimizu et al.¹⁰ have encountered three cases of anterior arch fracture following CLWF and reported the result of a biomechanical model, showing that 3.55 times higher stress at maximum is concentrated in the anterior arch after C1 laminectomy. The location in which the superlative stress was detected was a junctional zone or marginally lateral to the anterior tubercle. This stress-concentrated location noticed in the finite element model was consistent with fracture sites that were clinically observed. Regarding the loading condition, much higher stress was observed in extension and lateral bending as compared with other postures. Based on these findings, they assumed that minimal trauma such as coughing might induce fracture, which could be asymptomatic, raising the possibility that occult fractures might be more frequent after CLWF than reported.

However, since all patients with CLWF do not present anterior arch fracture, it has been thought that other possi-

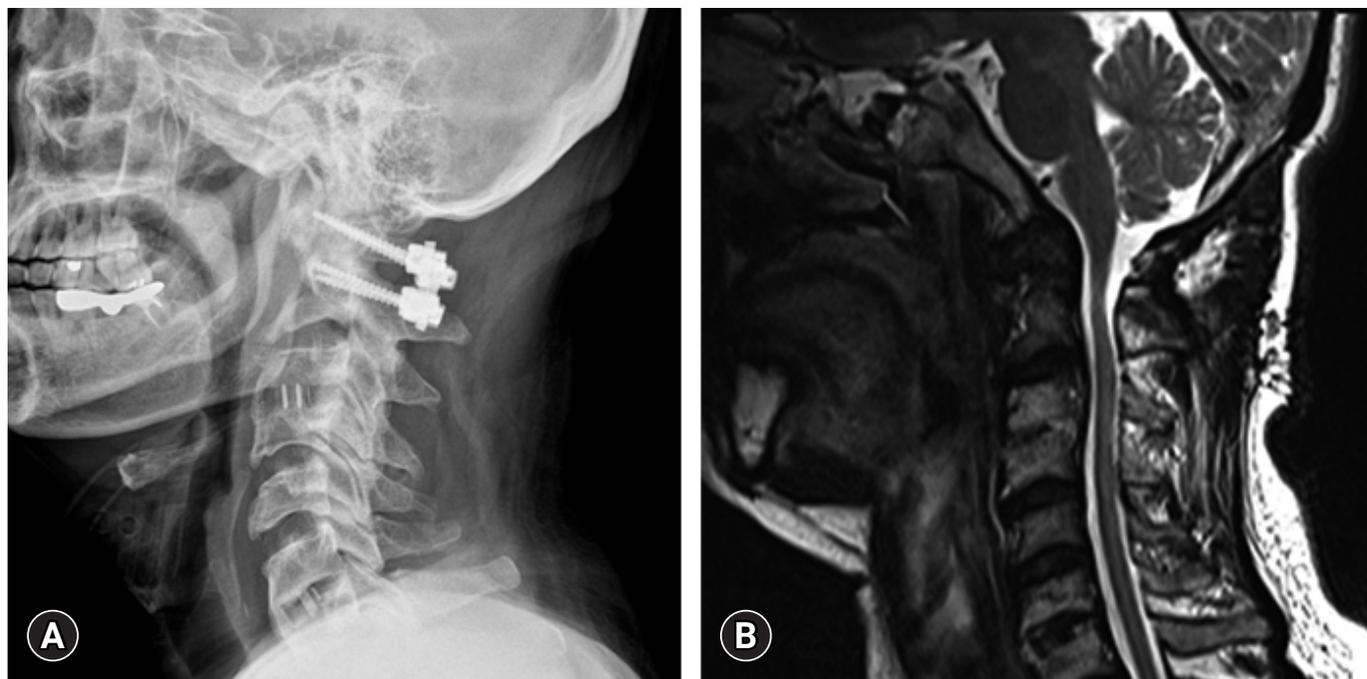


Fig. 3. (A) Lateral cervical radiograph showing C1 lateral mass screws and C2 pedicle screws. (B) Follow-up magnetic resonance imaging of the cervical spine revealing a decreased size of the retro-odontoid pseudotumor 3 months postoperatively.

ble risk factors are likely to contribute to the risk of anterior arch fracture. Shimizu et al.⁹⁾ have analyzed morphological characteristics of the C1-2 facet joint of 10 patients who had developed anterior arch fracture following CLWF after reporting their initial results¹⁰⁾ of the biomechanical model. The incidence of anterior arch fracture was 14.2% (10/70). Multivariate analysis revealed that a large inferior facet angle (IFA; defined as the coronal inclination angle of the C1-2 facet) and subaxial ankylosis were independent risk factors for anterior arch fracture. They regarded the anterior arch fracture associated with posterior defect as a stress fracture led by repetitive low-level stress wherein the greater coronal inclination of the facet could result in a greater quantum of laterally directed force. The ROC curve indicated that a large IFA, especially when IFA was greater than 23°, would draw not only greater flexural force on the anterior arch, but also greater displacement of the inferior articular process of the atlas. IFA of the patient in the current case was 26° (Fig. 2D), consistent with the previous report. Furthermore, the present patient already underwent C3-4 and C6-7 arthrodesis. As another risk factor for anterior arch fracture, subaxial ankylosis could play a role as an adjacent segment disease which might lead to overloading of adjacent structures. Since the C1-2 joint lacks an intervertebral disc, elevated loading might be directly conveyed to the anterior arch through the C1-2 inferior facet under situations wherein the posterior arch is faulty⁹⁾.

Insufficiency of the transverse ligament associated with retro-odontoid pseudotumor could compel the mechanical support mitigating the force, leading to anterior arch fracture¹⁰⁾. The pathogenesis of a retro-odontoid pseudotumor is not well explained. It seems that many authors side with the opinion of Crockard et al.³⁾ that a partial tear of the transverse ligament is followed by fibrocartilaginous metaplasia and a fibrovascular ingrowth. A vicious cycle then occurs, resulting in the production of a gradually expanding mass. Given that the transverse ligament is the most essential structure for stabilization of the atlantoaxial segment, we hypothesize that a weakened transverse ligament might mitigate the support of radial force and lead to severe neck pain in the setting of CLWF.

Optimal surgical treatment for a retro-odontoid pseudotumor has not been established yet. Takemoto et al.¹¹⁾ have reported the beneficial result of CLWF for a retro-odontoid pseudotumor in ten patients. They asserted that CLWF did

not cause severe AAI progression nor serious mass enlargement during an average of 29 months of follow-up. By contrast, some surgeons have used a posterior decompression and fusion technique considering that the reactive mass develops with resultant mechanical stress at the craniovertebral junction. Chikuda et al.¹⁾ have demonstrated that posterior fusion unfailingly yields a pseudotumor regression even in patients without AAI. Smilingly, we also experienced the resolution of the compressing mass after the second posterior fusion surgery. The mass had remained unchangeable following the initial CLWF.

CONCLUSION

CLWF for patients with retro-odontoid pseudotumor may carry the risk of developing postoperative spontaneous anterior atlas fracture. Although CLWF is a good surgical option for the treatment of stable cervical myelopathy associated with retro-odontoid pseudotumor, posterior decompression and fusion could be beneficial for patients with greater IFA of C1-2 and subaxial arthrodesis.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

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