

# Vertical Subluxation Caused by Infection Around the Odontoid Process: A Case Report

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## Learning Point of the Article:

Clinicians should keep infection in mind when treating patients with neck pain.

## Abstract

**Introduction:** There are few reports of cases with vertical subluxation. We report our experience of a vertical subluxation of the atlantoaxial joint caused by infection.

**Case Report:** A 63-year-old man had a 5-month history of neck pain. He was treated for rheumatoid arthritis by a local doctor. At admission to our hospital, he had neck pain and complained of dysphagia and hoarseness. His white blood cell count and C-reactive protein were 7200/mm<sup>3</sup> and 4.86 mg/dL. Cervical X-ray showed atlantoaxial subluxation. Computed tomography showed osteolysis around the odontoid process, including the atlantooccipital and atlantoaxial joints, causing vertical subluxation. Magnetic resonance imaging showed spinal cord and brainstem compression and the presence of fluid. Gadolinium-enhanced magnetic resonance imaging showed enhancement around the odontoid process. We performed occipital-cervical spine fixation and C1 laminectomy. Following surgery, his symptoms completely resolved. Bone fusion was confirmed 6 months after surgery.

**Conclusion:** If subluxation of the upper cervical spine occurs rapidly, infection should be considered.

**Keywords:** Atlantoaxial vertical subluxation, pyogenic spondylitis, atlantoaxial joint, atlantooccipital joint.

## Introduction

Spinal infections are devastating diseases associated with high mortality when not correctly diagnosed. Osteomyelitis of the cervical spine is rare compared with infections of the thoracic and lumbar spine. Malawski and Lukawski reported that cervical spondylitis accounts for 5.8% of all spinal infections, while infections of the upper cervical spine have an even lower incidence (0.7%) [1]. There are several reports of atlantoaxial subluxation (AAS) due to infection around the odontoid process [2, 3, 4]. However, there are few reported cases with vertical subluxation (VS). Patients with VS are at risk of death due to compression of the dentary process on the brainstem [5].

Herein, we report a case of VS due to infection around the odontoid process, resulting in bulbar paralysis.

## Case Report

The patient provided informed consent for publication of these findings. A 63-year-old man with a chief complaint of neck pain was examined at a local hospital. No neurological symptoms were observed. A cervical X-ray showed only degenerative findings (Fig. 1) and no instability, and he was treated conservatively for cervical osteoarthritis. However, his neck pain persisted after 1 month and computed tomography (CT) showed an AAS. Laboratory examinations were negative for

## Author's Photo Gallery



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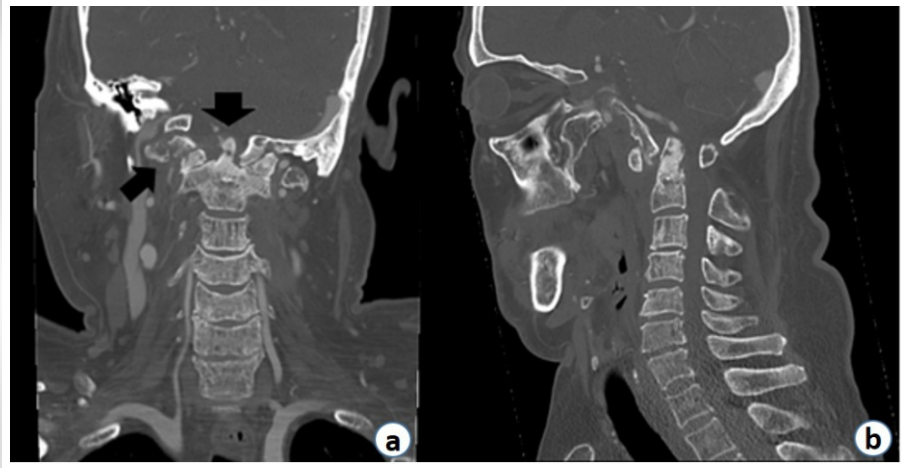
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**Figure 1:** Lateral view of a cervical X-ray acquired at a local hospital. There was no atlantoaxial subluxation or vertical subluxation.



**Figure 2:** Computed tomography (CT) before surgery. (a) Coronal CT showed osteolysis around the odontoid process, including the atlantooccipital and atlantoaxial joints (black arrows). (b) Sagittal CT showed vertical subluxation.

rheumatoid factor and anti-cyclic citrullinated peptide antibodies, although C-reactive protein and matrix metalloproteinase were mildly elevated. A blood culture test was also negative. Due to his family history of rheumatoid arthritis (RA) (his daughter had RA) and the presence of an AAS, he was treated with steroids and methotrexate for seronegative RA. However, his symptoms did not improve. He was referred to our hospital at 4 months after his first visit because of the progression of AAS and the appearance of bulbar paralysis.

At admission to our hospital, his body temperature was 36.1°C. He had pain from the occiput to the neck, although his neurological findings were normal (normal muscle strength, normal limb tendon reflexes, and no numbness in the extremities). He also complained of dysphagia, particularly with difficulty swallowing solid food, and had hoarseness. His white blood cell count and C-reactive protein were 7200/mm<sup>3</sup> and 4.86 mg/dL, respectively.

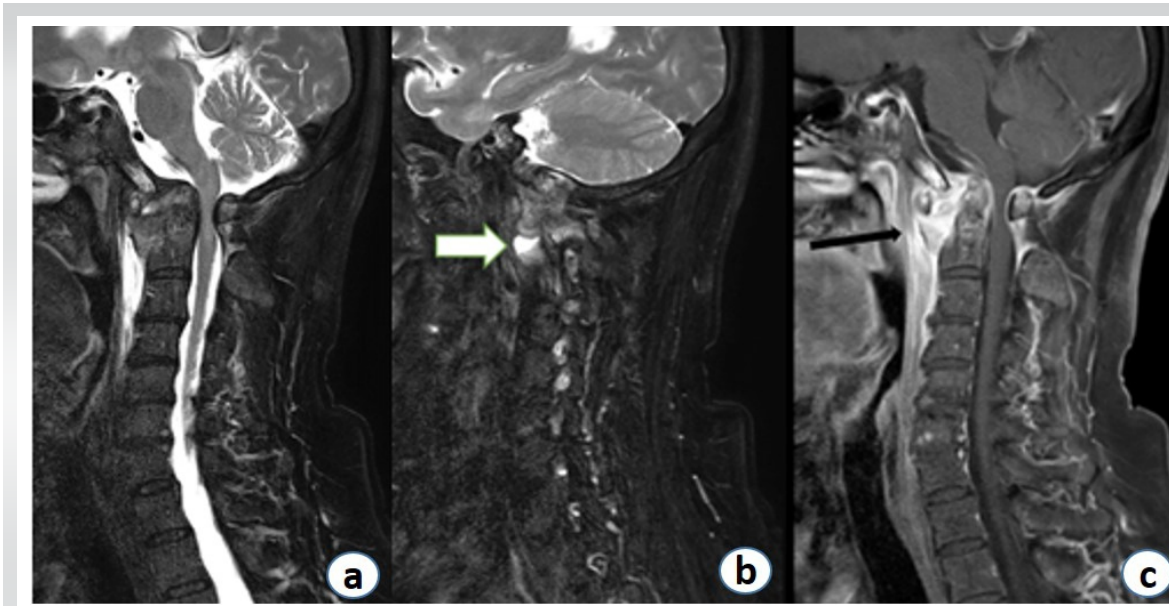
The lateral view of the cervical X-ray showed AAS, while CT showed osteolysis around the odontoid process, including the atlantooccipital and atlantoaxial joints, causing VS (Fig. 2). Magnetic resonance imaging (MRI) showed spinal cord and brainstem compression at the C1/2 level and fluid in the right lateral atlantoaxial joint on a T2-weighted image. Gadolinium-enhanced MRI showed enhancement around the odontoid process (Fig. 3). A blood culture test was performed again and methicillin-sensitive staphylococcus aureus was detected. We concluded that the AAS and VS were caused by infection around the odontoid process and surgical treatment was indicated.

We performed occipital-cervical (O-C4) spine fixation and C1 laminectomy. The patient's head was immobilized with a Mayfield skull clamp and the head was tractioned, retroflexed,

and aligned while viewing X-ray fluorography. During surgery, we confirmed free-running electromyography using motor-evoked potentials and ensured that the spinal cord was not overloaded. We also biopsied the right lateral atlantoaxial joint and performed a culture test, which detected methicillin-sensitive staphylococcus aureus. Thus, the patient was administered intravenous cefazolin (6 g/day) after surgery. His neck pain and dysphagia improved immediately after surgery and he was able to swallow solid food. His hoarseness also improved and his inflammatory response completely resolved. X-ray and CT showed bone union between the O-C4 spine at 6 months after surgery (Fig. 4).

### Discussion

Osteomyelitis of the cervical spine is rare, while infections of the upper cervical spine have an even lower incidence (0.7%) [1]. In 1896, Makins and Abbott first reported two cases of odontoid osteomyelitis [6], while there are several reports of AAS due to infection around the odontoid process [2-4]. A summary of representative case reports is shown in Table 1 [2-4,7-10]. Most patients had AAS; to the best of our knowledge, no reports have described VS. In addition, most of the previously described patients had a shorter time from onset to diagnosis than our patient, suggesting that delayed diagnosis may be a risk factor for progression to VS. Early diagnosis and treatment of VS are important because when a patient develops VS, the neurological prognosis is worse than AAS and there is a risk of death due to compression of the dentary process on the brainstem [5, 11]. In general, AAS is thought to occur in RA, trauma, and congenital diseases, and is particularly common in RA patients. As AAS progresses to VS when the lateral atlantoaxial or atlantooccipital joint is destroyed [12], AAS or



**Figure 3:** Magnetic resonance imaging (MRI) before surgery. (a) Sagittal T2-weighted image showed spinal cord and brainstem compression at the C1/2 level. (b) Sagittal T2-weighted image showed fluid in the right lateral atlantoaxial joint (white arrow). (c) Gadolinium-enhanced MRI showed enhancement around the odontoid process (black arrows).

of the upper cervical spine, especially secondary to inflammation of the otolaryngological region, is termed Grisel's syndrome – this condition was first reported by Bell in 1830 and named by Grisel in 1930. The most widely accepted mechanism of Grisel's syndrome is hyperemia

VS is often considered a result of RA. Nevertheless, Zhang et al. reported rates of progression of AAS, VS, and subaxial subluxation of four, six, and three lesions per 100 patients per year in RA patients, while the incidence of new or progressive cervical myelopathy was two cases per 100 patients with known cervical subluxations per year [13]. These findings suggest that AAS and VS are unlikely to progress rapidly in RA patients. Thus, if subluxation of the upper cervical spine occurs rapidly, other causes should be considered.

Non-traumatic AAS associated with inflammatory conditions

following infection or trauma that leads to decalcification of the anterior arch of C1 and laxity of the transverse ligament [14]. In addition, Parke et al. reported that the upper cervical spine may be infected by congestion and effusion from the lymph and veins in the posterior pharynx. In that study, the authors also suggested that this condition is more common in children because the upper spine is more mobile than in adults and they have a hypertrophic status of the parapharyngeal lymphoid tissue, which is located in the area drained by the pharyngovertebral plexus [15].



**Figure 4:** Cervical X-ray and CT showed bone union between the occipital-cervical spine (black arrows) at 6 months after surgery. (a) Lateral view of the cervical X-ray. (b) Coronal view of the cervical CT. (c) Sagittal view of the cervical CT.

In the present case, there were no fever or pharyngeal symptoms before the onset of neck pain. In addition, the patient had no history of trauma and had a family history of RA, which led to a false diagnosis and treatment with steroids – this may have also contributed to the progression of AAS and VS. These subluxations, which

Age/sex	References	Year	Bacteria	Operation	AAS or VS	Time to diagnosis
44/M	Bullock et al. [7]	2010	S. aureus	No	AAS	1 year
60/M	Tsunoda et al. [2]	2011	Unknown	PSF	AAS	1 month
42/M	Barbosa et al. [8]	2013	S. aureus	PSF	AAS	3 months
76/M	Hasegawa et al. [3]	2015	Unknown	No	AAS	2 weeks
53/M	Kobayashi et al. [4]	2015	S. aureus	PSF	AAS	20 days
56/M	Kuyumcu et al. [9]	2017	S. aureus	PSF	AAS	1 month
43/F	Di Russo et al. [10]	2018	P. acnes	PSF	AAS	1 month
63/M	Our case		S. aureus	PSF	VS	4 month

**S. aureus, Staphylococcus aureus, P. acnes, Propionibacterium acnes, PSF: Posterior spine fusion, AAS: Atlantoaxial subluxation, VS: Vertical subluxation**

**Table 1: Summary of representative case reports of upper cervical spine infection.**

are caused by infection around the odontoid process, are very serious conditions because they can lead to paraplegia and death due to cervical cord and brainstem compression [5,7]. Therefore, early diagnosis is critical. However, diagnosis can sometimes be very difficult because these subluxations are caused by numerous factors, including RA, trauma, and congenital diseases, and have no specific clinical signs. In addition, the symptoms are varied and nonspecific, and can present with normal inflammatory markers [9].

In addition to careful physical examinations, imaging assessment is important for the diagnosis of AAS and VS. In the early stages of infection, radiographs are often normal. By contrast, MRI can be used to evaluate both the presence of spinal cord compression and osteomyelitis and soft tissues around the odontoid process. CT is also useful in differentiating crystal-induced arthritis. There is no consensus on the surgical strategy for osteomyelitis of the upper cervical spine. Ueda et al. reported that an accurate diagnosis is the most important step and that antibiotic treatment should be administered if the infection is in the early stages. If the patient is resistant to antibiotic treatment, or if neurological deficits, subluxation, or malalignment occur, then surgical treatment should be

performed [16]. Our patient's treatment was delayed by 5 months because of misdiagnosis. An early and accurate diagnosis of infection around the odontoid process would have likely avoided the requirement for surgical treatment.

### Conclusion

We experienced a case of vertical subluxation of the atlantoaxial joint caused by infection with a 5-month delay in treatment. The neck pain and bulbar paralysis were successfully treated with O–C4 spine fixation. Clinicians should consider the potential for infection when treating patients with neck pain.

### Clinical Message

If we see a patient with long-lasting neck pain, we should also keep in mind infection of the upper cervical spine. In particular, we should be aware that there are cases of rapid progression of subluxation of the atlantoaxial joint.

**Declaration of patient consent:** The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Conflict of interest:** Nil **Source of support:** None



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