

Atlantoaxial Subluxation as an Unusual Presentation of Axial Spondyloarthritis: A Case Report

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Abstract

Background: Axial spondyloarthritis (AxSpA) is a chronic inflammatory rheumatic disease that primarily affects the spine and sacroiliac joints. Involvement of the atlantoaxial joint is rare but potentially life-threatening, as it may progress to atlantoaxial dislocation (AAD) and cervical myelopathy.

Case Presentation: A 41-year-old woman with HLA-B27-positive AxSpA on adalimumab presented with a two-day history of quadriparesis and one day of dysphagia, following an inadvertent missed dose of biologic therapy. Examination revealed restricted cervical spine mobility, reduced motor power with brisk reflexes, and bilateral extensor plantar responses. Inflammatory markers were elevated and hemoglobin was low, consistent with anemia of chronic disease. Magnetic resonance imaging of the cervical spine demonstrated atlantoaxial subluxation with retro-odontoid pannus and spinal cord compression, while radiographs of the pelvis and dynamic cervical views confirmed chronic sacroiliitis and atlantoaxial instability. The patient was referred for urgent neurosurgical stabilization.

Conclusion: Atlantoaxial subluxation, though uncommon, is an important and under-recognized neurological complication of AxSpA. Clinicians should maintain a high index of suspicion for cervical spine involvement in patients with long-standing disease, elevated inflammatory markers, or lapses in biologic therapy, as early recognition and timely surgical intervention can prevent irreversible neurological damage.

Keywords: Axial spondyloarthritis; Atlantoaxial subluxation; Cervical myelopathy; HLA-B27; Sacroiliitis; Biologic therapy

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Introduction

Axial spondyloarthritis (AxSpA) is a chronic inflammatory disease that primarily affects the spine and sacroiliac joints, giving rise to back pain, stiffness, and progressive spinal deformity. The disease spectrum encompasses both radiographic disease, or ankylosing spondylitis (AS), and non-radiographic AxSpA (nrAxSpA), with considerable heterogeneity in clinical presentation (1,2).

Although the spine and sacroiliac joints are the characteristic sites of involvement, the atlantoaxial joint is affected only rarely. When it does occur, atlantoaxial dislocation (AAD) or subluxation can result from chronic inflammation and joint destruction and, if left untreated, may cause substantial morbidity and mortality. Early recognition and prompt management are therefore essential. We report a case of AxSpA that first came to clinical attention because of atlantoaxial subluxation, and we discuss the clinical, radiological, and management aspects of this uncommon presentation.

Case Presentation

A 41-year-old woman with a known history of HLA-B27-positive axial spondyloarthritis, maintained on adalimumab for the past several months, presented to the emergency department with sudden-onset weakness of all four limbs of two days' duration and difficulty swallowing for one day. She denied any recent trauma, fever, or upper respiratory tract infection. Her disease had previously manifested as chronic inflammatory back pain without recent exacerbation; notably, she had missed her most recent dose of adalimumab two days before symptom onset. On presentation, she was alert and hemodynamically stable, with a temperature of 36.8°C and a body mass index of 22.3 kg/m². Examination revealed marked restriction of cervical spine movement in all planes, most pronounced on flexion and rotation, with tenderness over the upper cervical spine. Neurological examination showed reduced motor power in the proximal upper and lower limbs (Medical Research Council grade 3/5) with relatively preserved distal strength (4/5), diminished grip strength bilaterally, and brisk deep tendon reflexes with a positive Hoffmann's

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sign. Plantar responses were extensor bilaterally, and sensory testing revealed mild impairment of joint position and vibration sense in the lower limbs without a discrete sensory level; no clonus or muscle atrophy was noted. Cranial nerve examination, including pupillary responses, extraocular movements, and gag reflex, was unremarkable, and there was no dysarthria or facial asymmetry. Schober's test was reduced and the FABER test was positive bilaterally, indicating sacroiliac joint involvement, though no peripheral synovitis or enthesitis was evident at admission. Laboratory investigations showed elevated inflammatory markers, with a C-reactive protein (CRP) of 7.5 mg/L and an erythrocyte sedimentation rate (ESR) of 52 mm/hr. Hemoglobin was 7.2 g/dL, consistent with anemia of chronic disease, while renal and liver function tests were within normal limits. HLA-B27 was positive; rheumatoid factor, antinuclear antibody, and anti-cyclic citrullinated peptide antibodies were all negative.

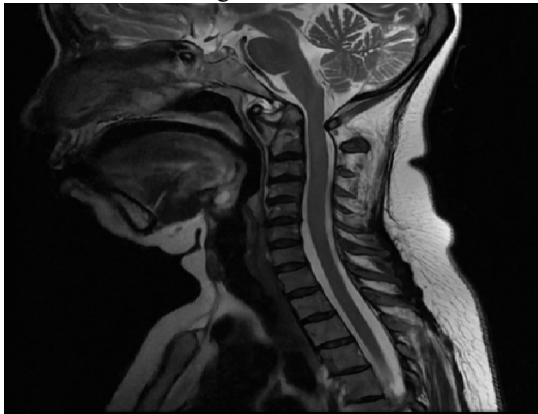


Figure 1. Sagittal MRI of the cervical spine showing atlantoaxial subluxation.



Figure 2. Sagittal MRI demonstrating retro-odontoid pannus formation with compression of the cervical spinal cord.

Magnetic resonance imaging of the cervical spine confirmed atlantoaxial subluxation (Figure 1) with retro-odontoid pannus formation (Figure 2) and resultant compression of the cervical spinal cord.



Figure 3. Dynamic cervical spine radiographs, flexion and extension views.

On the flexion view, there was visible widening of the anterior atlanto-dental interval (AADI) between the anterior arch of C1 and the odontoid process of C2, indicating anterior atlantoaxial subluxation, together with disruption of the posterior cervical alignment at the C1–C2 junction. These findings are consistent with atlantoaxial instability, most likely secondary to transverse ligament compromise, as is commonly observed in chronic inflammatory conditions such as AxSpA. The extension view showed partial reduction of the AADI, confirming the dynamic nature of the instability.



Figure 4. Anteroposterior radiograph of the pelvis. The pelvic radiograph demonstrated irregular sacroiliac joint margins, subchondral sclerosis, and joint space narrowing, with sclerosis more prominent on the iliac side — a hallmark of chronic sacroiliitis.

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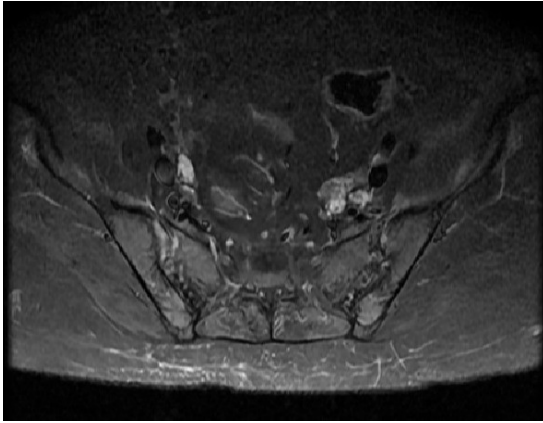


Figure 5. MRI of the pelvis showing bilateral sacroiliitis.

In view of the neurological deficits and imaging evidence of cervical cord compromise, the patient was referred to the neurosurgery team for further management and surgical stabilization.

Discussion

Atlantoaxial dislocation is an uncommon but clinically important complication of axial spondyloarthritis. Although more frequently described in rheumatoid arthritis (3,4), cervical spine involvement — particularly at the C1–C2 level — can also occur in AxSpA, albeit less commonly (2). The atlantoaxial joint comprises a median atlanto-odontoid articulation and bilateral facet joints, the stability of which is maintained principally by the transverse ligament. Inflammatory processes such as enthesitis (5) or pannus formation adjacent to this ligament can weaken its integrity, ultimately resulting in instability and dislocation.

In the present case, quadriparesis and dysphagia were traced to atlantoaxial subluxation confirmed on imaging, findings indicative of cervical spinal cord compression — a recognized but under-appreciated complication of AxSpA. While cervical involvement can remain clinically silent in some patients, symptoms such as restricted neck movement, neck pain, neurological deficits, or bulbar symptoms should prompt detailed evaluation of the cervical spine, and infective or mechanical mimics such as atlanto-odontoid pyogenic arthritis (6) or the crowned dens syndrome (7) should be considered in the differential diagnosis.

Magnetic resonance imaging has emerged as the preferred modality for diagnosing AAD, as it can detect not only osseous malalignment but also soft-tissue changes such as retro-odontoid pannus and spinal cord signal abnormalities, and it remains the primary imaging modality even in atypical age groups such as children (8). Plain radiography with dynamic flexion–extension views can reveal an abnormal atlanto-dental interval, but its sensitivity is lower than

that of MRI. In our patient, MRI demonstrated retro-odontoid pannus with cord compression, supporting a diagnosis of AAD with myelopathy; congenital causes of atlantoaxial instability, such as os odontoideum, remain an important differential in appropriate clinical contexts (9).

Recognized risk factors for AAD in AxSpA include long-standing disease, elevated CRP, peripheral arthritis, and prior or ongoing biologic therapy — particularly following an inadequate response to conventional disease-modifying agents (1). Our patient had several of these features, including long disease duration, peripheral joint involvement, a history of uveitis, and a recent lapse in biologic treatment, which likely contributed to disease flare and consequent cervical instability.

Conservative management may be appropriate in asymptomatic patients or those without evidence of cord compromise, as illustrated by reports of successful non-operative management in select cases (10). However, surgical stabilization — including techniques such as transoral decompression and fusion in irreducible cases (11) — remains the cornerstone of management for symptomatic patients, distinct from purely traumatic presentations of atlantoaxial injury (12). Timely surgical intervention can prevent further neurological deterioration, restore spinal stability, and improve functional outcomes.

Conclusion

This case highlights atlantoaxial subluxation as a rare but serious neurological complication of axial spondyloarthritis, capable of presenting acutely with quadriparesis and dysphagia. A high index of clinical suspicion, supported by prompt MRI evaluation, is essential for early diagnosis, and multidisciplinary management with timely surgical stabilization can prevent irreversible neurological injury and improve outcomes.

Declarations

Patient Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Conflict of Interest: The authors declare no conflict of interest.

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Authors' Contributions: Both authors were involved in patient care, manuscript preparation, and approved the final version for submission.

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