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Case Report

Complete Fusion of Three Lumbar Vertebral Bodies in Ankylosing Spondylitis

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Conflict of Interest
The authors have no financial conflicts of interest.

ABSTRACT

Ankylosing spondylitis is a seronegative arthropathy of unknown etiology affecting mainly young adults. Acute or chronic changes of the spine such as sacroiliitis, spondylodiscitis, osteitis, ossification, and arthropathic lesions characterize the disease. Spinal involvement may accompany ossification of the ligaments, disc, end-plates and apophyseal structures, and seems to be bamboo spine. Here, we report a rare case describing the complete fusion of three lumbar vertebral bodies in ankylosing spondylitis. This rare case is presented alongside a literature review.

Keywords: Ankylosing spondylitis; Vertebral fusion; Lumbar vertebrae

INTRODUCTION

Seronegative spondyloarthritis is a general term for a group of joint conditions that are not associated with rheumatoid factors or rheumatic nodules. Five subgroups of spondyloarthritis are characterized: ankylosing spondylitis, reactive arthritis, psoriatic arthritis, arthritis associated with inflammatory bowel disease, and undifferentiated spondyloarthritis.[5,9]

Among them, ankylosing spondylitis is a progressive inflammatory disease with a predilection for the axial joints. Over time, chronic spinal inflammation may result in fusion of the vertebrae. However, complete fusion of three vertebral bodies and concurrent lack of disc space in ankylosing spondylitis is extremely rare. We describe radiographic features including the magnetic resonance imaging (MRI) results and review the relevant literature.

CASE REPORT

A 37-year-old man with a 5-year history of ankylosing spondylitis was admitted for severe lower back and left leg radiating pain. He was diagnosed with ankylosing spondylitis 5-years ago in another hospital due to history of sacroiliitis, HLA-B27 (+), inflammatory back pain,

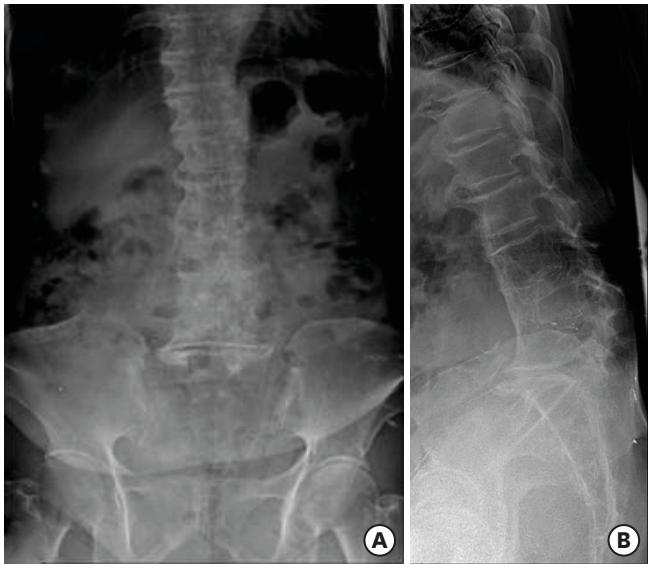

FIGURE 1. Preoperative anterior posterior (A) and lateral (B) radiographs show the complete fusion of three vertebral bodies.

and his father was suffered from ankylosing spondylitis since young age. The pain radiated from his back to his left buttock and posterior thigh. He had long experienced continuous back pain and limited motion for ankylosing spondylitis. He developed the radiating pain in his left leg 6 months prior to his admission, however had received no specific treatment. He denied any history of recent trauma and previous infection, laboratory test results were unremarkable. Lumbar spine radiographs showed complete fusion of the L3, L4 and L5 vertebral bodies and kyphotic change (**FIGURE 1**).

MRI of the lumbar spine was performed to further evaluate symptoms. The T1- and T2-weighted images showed left foraminal stenosis at the L5-S1 level and complete vertebral body fusion. Moreover, the MRI revealed distinctive features of ankylosing spondylitis, such as sclerotic changes at the edges of the vertebral endplates, commonly referred to as the 'Andersson lesion'. MRI revealed spondylitis of anterior vertebral corners, referred to as the 'shiny corner sign' or 'Romaus lesion' (**FIGURE 2**).

The patient underwent posterior lumbar interbody fusion with screw fixation at the L5-S1 level. There was no L5-S1 facet fusion finding due to infection or ankylosing spondylitis. We just found compressed left L5 nerve root due to ligament and facet hypertrophy.

His pain, particularly in his left leg, improved significantly after surgery. He was able to walk after the second postoperative day and discharged 10 days after surgery. At the final follow-up of 6 months after surgery, he claimed to be pain free but experienced slightly limited back movement (**FIGURE 3**).

This research is about an individual case without the approval of an ethical research. We obtained informed consent from the patient.

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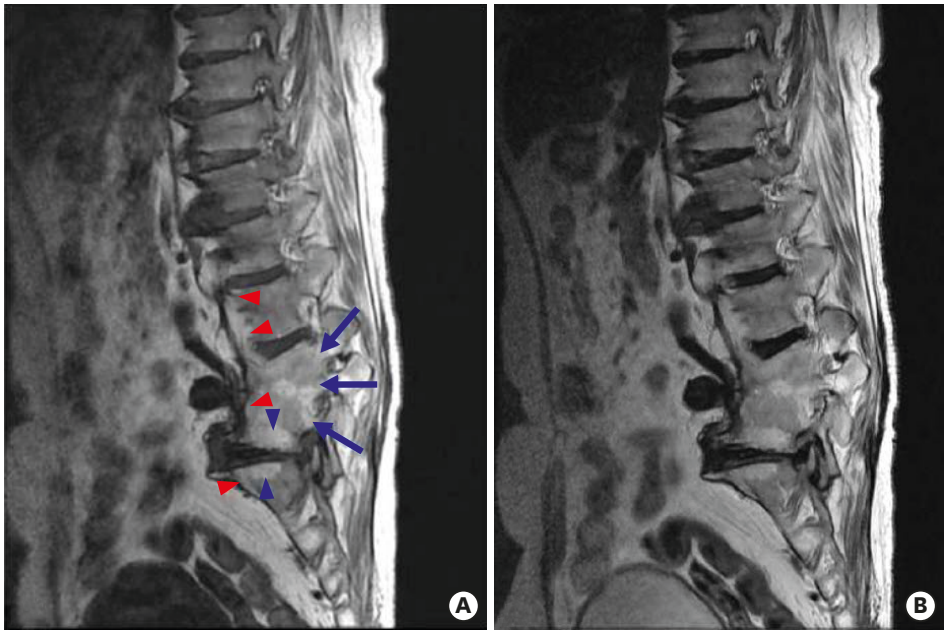

FIGURE 2. T1- and T2-weighted sagittal magnetic resonance images (A & B) reveal severe foraminal stenosis and complete fusion of vertebral bodies. Each of the 3 pedicles is observed (arrows) and the "Andersson lesion" is visible at the antero-inferior portion at L5 and the antero-superior portion at S1 (blue arrowheads). "Shiny corner sign" is visible at anterior vertebral corners as high signal intensity (red arrowheads).

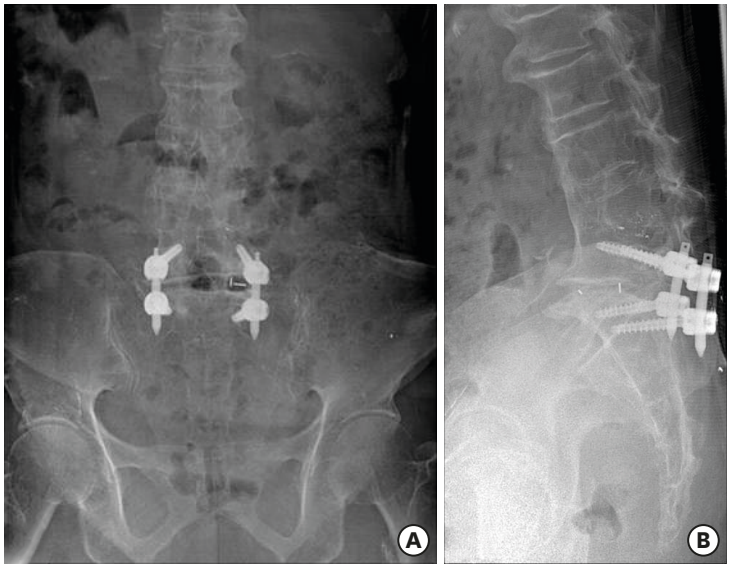

FIGURE 3. Postoperative simple radiographs (A & B) taken 6 months after the posterior lumbar interbody fusion at the L5-S1 level.

DISCUSSION

Ankylosing spondylitis is a seronegative arthropathy of unknown etiology affecting mainly young adults, with men more severely affected than women. Most patients with ankylosing spondylitis carry the HLA-B27 antigen, and although the quoted prevalence of 0.1–1% seems rather high, it is likely that many cases are mild, manifesting only as pain and stiffness with no other features.[6,7,10]

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Although bilateral symmetric sacroiliitis is a common presenting symptom at both the synovial and ligamentous parts of the joint, many different inflammatory changes may occur in the vertebral column owing to the anatomic complexity of ankylosing spondylitis. Arthritic lesions may affect the vertebrae, the intervertebral discs, and the tendon and ligament attachments. Several vertebral lesions may ultimately lead to chronic abnormalities such as syndesmophytes or ankylosis.

AS is also associated with vertebral osteoporosis.[2,3] Because of sagittal imbalance of the spine and osteoporosis, a pathological fracture can occur with minor trauma or even without any trauma, which is different from a general spine fracture. A pseudoarthrosis usually forms at the fracture site when there is abnormal movement and repeated inflammatory stimuli. Pathological fracture and formation of a pseudoarthrosis progressively increase the kyphotic deformity, with the patient suffering from severe back pain and, in some cases, nerve dysfunction.[11]

Osseous erosion (osteitis) of the vertebral endplates combined with the ossification of ligamentous insertions (entheses) lead to anterior vertebral concavity squaring. Focal destructive changes at the discovertebral junction described by Romanus and Ydén[8] result in reactive sclerosis of the antero-superior and antero-inferior aspects of the vertebral bodies and lead to the “shiny corner sign” observed on plain radiographs or MRI. Spondylodiscitis (inflammatory Anderssen lesions) is characterized by high signal intensity at the cortical plates adjacent to intervertebral disc on MRI.[1] Moreover, MRI investigation can yield an unsuspected diagnosis, not always revealed by plain radiographs.

Discovertebral lesions in ankylosing spondylitis have been classified by Cawley et al.[4] into 3 types. Type I lesions involve the central portion of the discovertebral junction, which is covered by the cartilaginous endplate, and have the same features as Schmorl's nodes. Type II lesions involve the peripheral portion of the discovertebral junction, which is not covered by the cartilaginous end plate. Type III lesions involve both central and peripheral portions of the discovertebral junction, and may be of inflammatory origin in early stages of the disease (also referred to as spondylodiscitis). However, this type of lesion is more often characteristic of advanced disease. Multiple levels of the spine can be affected simultaneously, and changes seen may resemble those of infection. Of the many atypical presentations of ankylosing spondylitis, few reports in the literature present isolated or multiple discovertebral lesions. The “shiny corner sign” can aid diagnosis in patients with previously undiagnosed back pain and narrow the differential diagnosis in patients with spondyloarthropathies.

In this case, signal changes in the antero-inferior aspect of L5 and antero-superior aspect of S1 were specific to the ankylosing spondylitis “shiny corner sign” presented together with bilateral symmetrical sacroiliitis. Efforts should be made to diagnose patients with advanced ankylosing spondylitis correctly, and care should be taken during surgery due to the complicated anatomical variations of this condition.

New bone formation occurs at the corners of the vertebral bodies in ankylosing spondylitis. It is characterized by thin, vertically oriented new bone formations on the peripheries of discs. Bony bridges and new bone formation occur in the intervertebral discs. However, complete fusion of three vertebral bodies and concurrent lack of disc space in ankylosing spondylitis is extremely rare.

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A thorough understanding of associated anatomical changes and inflammatory processes will facilitate diagnosis and treatment. In this case, we understood operative anatomy about fused body level through pedicle counting. And we want to share our experience of such anatomic variant and difficulty.

CONCLUSION

We report a rare case of advanced ankylosing spondylitis involving the complete fusion of three vertebral bodies. Ankylosing spondylitis is a complex disease that is difficult to diagnose. A thorough understanding of associated anatomical changes and inflammatory processes will facilitate diagnosis and treatment.

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