

Case Report

Sacral Stress Fracture Complicated by L5 Radiculopathy in a Patient with Rheumatoid Arthritis

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A 60-year-old Japanese woman with severe osteoporosis presented with a history of right buttock pain and right lateral lower leg pain in an L5 distribution. She had been treated with methotrexate and methylprednisolone for rheumatoid arthritis (RA) and interstitial pneumonia. Computed tomography demonstrated a sacral stress fracture in the right sacral ala. The right L5 nerve root was compressed by the fracture site. This case is rare in that L5 radiculopathy was complicated by a sacral stress fracture. Clinicians should suspect sacral stress fractures when RA or osteoporosis is present in women who experiences lumbar pain and lumbar radiculopathy.

Key words: lumbar 5 radiculopathy, sacral stress fracture, osteoporosis, rheumatoid arthritis

Sacral stress fractures can occur when normal or low stress occurs in elderly osteoporotic women [1]. In patients with rheumatoid arthritis (RA), a sacral stress fracture is comparably detected [2]. The correct diagnosis is initially delayed because radiographs present as normal, but later show an established fracture based on bone scans and sacral computed tomography (CT) [2]. Low back pain and sciatica are often complicating, and in these cases, lumbar discogenic pain caused by a lumbar disc hernia or lumbar canal stenosis are indicated as the cause of pain. However, an L5 root sign complicated with a sacral fracture is very rare. Here we report the case of a sacral stress fracture complicated by L5 nerve root sign.

Case Presentation

A 60-year-old Japanese woman with a 6-year history

of RA suffered from spontaneous-onset severe right buttock and groin pain and right lateral lower leg pain with paresthesia for a few days, unrelated to trauma. She visited our clinic because she could not sit or walk without support due to pain. Her previous anti-rheumatic therapy included 6 mg/week of methotrexate for 5 years. She had also experienced interstitial pneumonia and was treated with 10 mg/day of methylprednisolone for 6 years. She had been diagnosed with glucocorticoid-induced osteoporosis (her lumbar bone mineral density was 0.507 g/cm², and T-score was -4.55) and was treated with 5 mg/day of alendronate for 2.3 years and with 35 mg/week of alendronate for 1 year. Her physical status showed no laterality in the patella and Achilles tendon reflex tests, and the straight leg raising test was positive (pain was triggered in the right L5 nerve root region at 30° elevation). Kemp sign was negative but we could not confirm Patrick sign because of severe her right hip joint pain. A manual muscle test

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of the lower extremities demonstrated a score of 4 for her right quadriceps femoris and hamstrings, but the score for all other lower extremity muscles were 5.

In plain radiographs, the pelvic anteroposterior view showed that the patient's right hip was protrusio acetabuli (Fig. 1A). The lumbar lateral view showed osteoporotic changes, but did not show vertebral compression fractures or spondylolisthesis (Fig. 1B). The lumbar MRI did not show either lumbar disc herniation or vertebral body compression fracture. The muscle weakness occurred in the innervation region of the L4

nerve root, which was different from the innervation region of the L5 nerve root.

To identify the responsible root for the patient's symptom, we administered a right L5 nerve root block twice. The right L5 nerve root was accurately radiographed with a contrast agent (Fig. 2), and a reproducing pain was detected in her right lateral lower leg. However, the effect of the block persisted for only a few days.

The blood examination at day 12 after admission demonstrated that the patient's serum ALP was suddenly elevated to 906 IU/L (Fig. 3); thus, a malignant

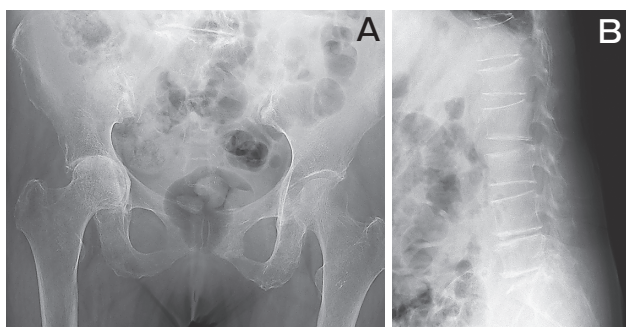


Fig. 1 X-ray findings. **A**, The anterior-posterior view of the pelvis shows right hip destruction (protrusio acetabuli) but no remarkable signs of a right sacral ala fracture; **B**, The lateral view of the lumbar spine shows osteoporotic change and disc space narrowing at the L4/5 and L5/S1 levels. However, neither vertebral compression fractures nor spondylolisthesis was observed.

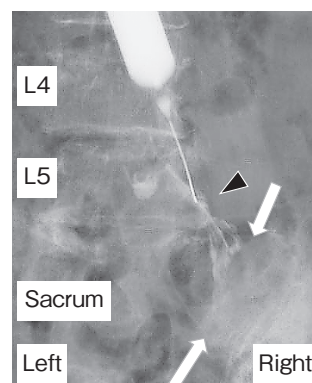


Fig. 2 Right L5 nerve root radiography. The right L5 nerve root (black arrowhead) was detected by contrast agent. The anatomical localization of the right L5 nerve root appears to pass just above the right sacral fracture site (white arrows).

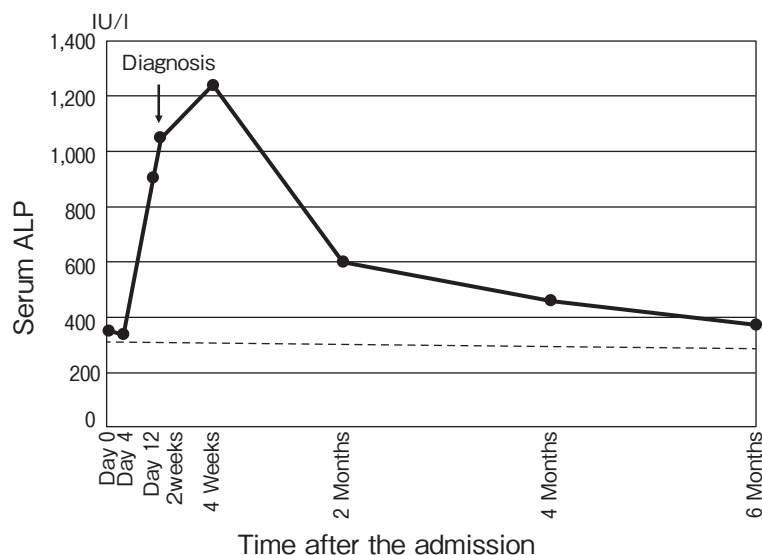


Fig. 3 Transition of the serum alkaline phosphatase (ALP) level. At the onset of the patient's symptoms, her serum ALP (normal range; 106 to 322 IU/L) was 351 IU/L. Four days after onset, it was 338 IU/L, and was remarkably elevated to 906 IU/L at 12 days post-onset. The serum ALP peaked at 1,239 IU/L at 4 weeks post-onset. At 2 months post-onset, the level had decreased to 600 IU/L, and it was finally downregulated to 373 IU/L at 6 months post-onset. Dashed black line: the normal highest value (322 IU/L) of serum ALP.

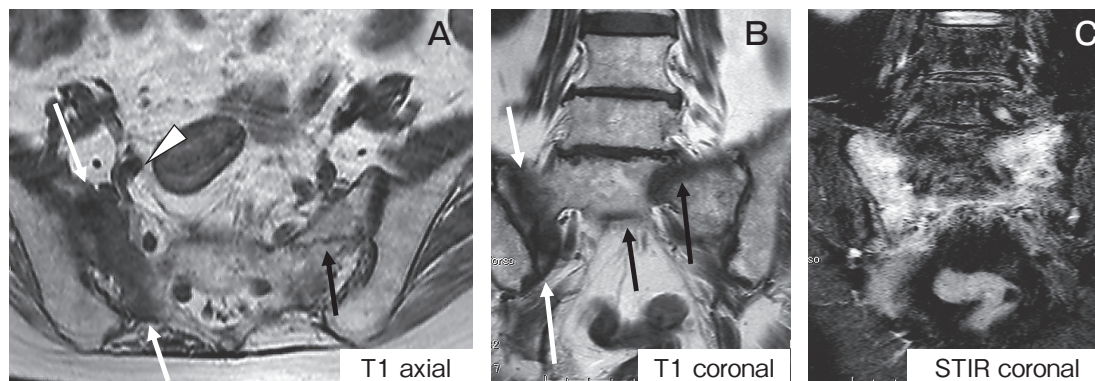


Fig. 4 MRI of the pelvis. **A**, In the T1 axial view, the anterior to posterior fracture line (*white arrows*) in the right ala region of sacrum was detected as T1 low intensity. The right L5 nerve root (*white arrowhead*) was very close to the fracture line. The transverse fracture line (*black arrow*) in the left ala region of sacrum was detected as T1 low intensity; **B**, In the T1 coronal view, the superior to inferior fracture line (*white arrows*) in the right ala of the sacrum was detected as low intensity. The oblique fracture line (*black arrows*) in the left ala and transverse fracture line in the central sacrum (*black arrow*) were detected as low intensity; **C**, In the STIR coronal view, a so-called 'H-shaped sign' was clearly observed in accordance with the area shown in panel B.

primary or metastatic bone tumor was suspected, and pelvic CT and MRI were conducted for a differential diagnosis.

In the pelvic MRI, a low-intensity area (T1 WI) in the right ala of the sacrum was detected (Fig. 4A,B). In addition, high intensities and an H-shaped sign were detected in the STIR coronal view (Fig. 4C). In the pelvic CT, the fracture site protruded into the anterior side of the pelvis and directly compressed the right L5 nerve root (Fig. 5A). Based on these MRI and CT findings, we finally diagnosed a sacral insufficiency fracture with right L5 radiculopathy. We also suspected that the patient's quadriceps muscle weakness was due to the ipsilateral osteoarthritis of the hip joint (Fig. 1A).

Conservative therapy, including analgesic and bed rest, were recommended, and gradually both the lumbar pain and right lateral lower leg pain improved. Tension signs diminished, and at 3 weeks after admission the patient was able to maintain a sitting position. At 4 weeks after admission, she could walk with a T-cane, and she was discharged 5 weeks after admission. Oral alendronate (35 mg/week) was continued even after the diagnosis. A pelvic CT exam performed 5 months after the onset revealed that the fracture site had healed and was stabilized. The right L5 nerve root was not compressed by the fracture site (Fig. 5B). The patient reported no further right lateral lower leg pain.

Her serum ALP value was further monitored. After her serum ALP level peaked at 1,239 IU/L at 4 weeks post-onset, it gradually downregulated to 600 IU/mL

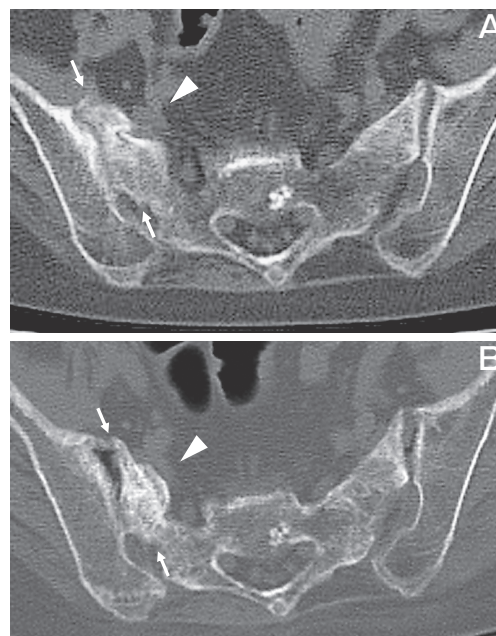


Fig. 5 Pelvic CT findings. **A**, Plain CT findings of the sacral insufficiency fracture at symptom onset. The fracture line was detected in the right sacral ala (*white arrows*) and was displaced. The right L5 nerve root (*white arrowhead*) was localized just anterior to the fracture site; **B**, Five months post-onset. The fracture site had healed, and the right L5 nerve root (*white arrowhead*) was no longer compressed.

at 2 months and was almost normalized at 6 months post-onset (Fig. 3).

This patient was asked whether her case could be submitted for publication, and she gave her consent for

the publication.

Discussion

The risk factors for sacral stress fractures include older age, female gender, the presence of osteoporosis, RA, the use of methylprednisolone, irradiation, and metabolic bone diseases [3]. Our patient's case had nearly all of these risk factors.

Tsiridis *et al.* reported that the incidence of sacral stress fractures in Finnish women >60 years old was 0.095% [4]. The incidence of sacral stress fractures is generally higher in women with RA aged >55 years [5]. Weber reported a rate of 1.8% [5], and they indicated that sacral stress fractures are not rare in postmenopausal women with RA.

The complication rate of a nerve root sign with sacral stress fractures is approximately 2%, and most nerve root signs occur at the S1 or S2 root [6]. The Denis classification of a sacral fracture is well known. Zone 1 is the region of the ala, zone 2 is the region of the sacral foramina, and zone 3 is the region of the central sacral canal [7]. An insufficiency fracture is Zone 1 (sacral ala site) dominant, and a traumatic fracture is Zone 2 or 3 dominant. Our patient was diagnosed with a Denis Zone 1 sacral fracture. Radiculopathy usually occurs in a Zone 2 fracture because the fracture line passes through the sacral lacerated foramen.

There are only a few reports of L5 radiculopathy secondary to a sacral stress fracture [6,8-10]. The prior cases included a mounted police officer (a 26-year-old female), two postpartum sacral stress fractures, and a 70-year-old female after irradiation. In our patient's case, the fracture site itself existed at the right sacral ala (Denis Zone 1) apart from the sacral foramina. However, it directly conflicted with the right L5 nerve root just anterior to the right sacral ala and caused right L5 radiculopathy.

The diagnosis of a sacral insufficiency fracture is difficult using plain radiograph findings, and this results in a delay of the diagnosis. Our patient demonstrated L5 nerve root pain, and she was thus initially diagnosed with a herniated lumbar disc. The finding of her right hip deformity made the diagnosis more difficult. The understanding of a patient's illness and findings from imaging analyses such as CT, MRI, and bone scintigraphy are useful to determine the appropriate diagnosis.

We also observed an elevated serum ALP level during the persistence of our patient's symptoms. The acute upregulation of serum ALP in this case suggested a primary or metastatic bone tumor malignancy. However, the imaging findings eliminated the possibility of a tumor lesion. In previous reports regarding sacral stress fractures, the patient's serum ALP was mildly [3] or prominently [4] increased during the course of the fracture. In our patient, right buttock and right lateral lower leg pain preceded the rapid serum ALP elevation by approximately 10 days. In another report [11], patients' serum ALP was upregulated in the early stage after the incidence of 10 fractures (5 femurs, two tibias, clavicle, radius, and vertebral body) and had recovered to the normal range by the healing stage. Usually, serum ALP upregulates as a reaction of fracture healing and reflects osteoblastic function.

In conclusion, sacral insufficiency fractures should be included in the differential diagnosis when a patient with RA or osteoporosis, with bone insufficiency, experiences sciatic pain.

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