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CASE REPORT

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Disturbances of the symphysis pubis in rheumatoid arthritis: report of two cases

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Abstract We present two rheumatoid arthritis (RA) patients suffering from disturbances of the symphysis pubis. Radiography revealed one with pelvic ring disruption with symphysis pubis diastasis, and the other with osteolysis at both pubic rami and disruption of the superior aspect of the symphysis pubis. Both cases had received long-term corticosteroid therapy, including pulse therapy. We recommend reducing the corticosteroid dose to prevent disturbances of the symphysis pubis especially in RA patients on long-term steroid therapy.

Key words Felty syndrome · Osteomyelitis · Pelvic ring disruption · Rheumatoid arthritis · Symphysis pubis diastasis

Introduction

While disturbances of symphysis pubis can be caused by the physiological pelvic ligament relaxation and increased joint mobility that occurs during pregnancy, this natural widening generally decreases following delivery, allowing the symphysis pubis to fully return to its normal state and strength.[1] Although pelvic insufficiency fractures often occur in postmenopausal osteoporotic patients with rheumatoid arthritis (RA),[2,3] there are quite few reports of disturbances of symphysis pubis in RA patients.[4,5]

We present here two RA patients with disruption of the symphysis pubis. One patient had minor injuries associated with a pelvic ring disruption with symphysis pubis diastasis, while the other had symphysis pubis diastasis with pubic osteomyelitis.

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Case 1

The patient was a 69-year-old woman with a 32-year history of RA. Her disease activity was not influenced by a disease-modifying antirheumatic drug (DMARD) regimen that included bucillamine, D-penicillamine, gold, and sulfasalazine. Due to increased disease activity, her DMARD treatment regimen was changed to methotrexate. She had also undergone corticosteroid therapy with prednisolone over the 25 years. Previously, this patient underwent bilateral total knee arthroplasty in 1995. Upon hospitalization in March 1999 because of interstitial pneumonia, methotrexate was discontinued and she received steroid pulse therapy (500 mg/day) for 3 days. After discharge, she was treated with prednisone 10 mg/day and home oxygen inhalation therapy. During a regular check-up, a diagnosis of pelvic insufficiency fracture was made by radiography, and the patient received vitamin D and the bisphosphonate alendronate to prevent steroid-induced osteoporosis (Fig. 1A). The fracture was externally fixed and the pain was reduced following bed rest at home.

When the patient was 71 years old, she accidentally fell on the floor of her home. Thereafter, her chief complaint was sudden onset of hip, groin, and buttock pain and difficulty in ambulation. Radiography revealed pelvic ring disruption with symphysis pubis diastasis (Fig. 1B). The patient's pain was so severe that she could not change position. On admission she was still receiving home oxygen inhalation therapy, which consisted of oxygen treatment to achieve a SaO_2 level >90% if hypoxemia was detected during exercise or sleep. Respiratory functional findings under oxygen inhalation therapy (3 l/min) were as follows: pH 7.395; $PaCO_2$ 42.8 mmHg; PaO_2 87.6 mmHg; HCO_3 25.6 mmol/l. However, the patient was judged to be ineligible for open reduction and internal fixation treatment due to severe interstitial pneumonia. Instead, bed rest, corsets, and pelvic slings were used to achieve pain relief. The pelvic slings were designed to provide reduction and stabilization of open-book pelvic fractures. Unfortunately, the status of her pelvic ring disruption with symphysis pubis diastasis

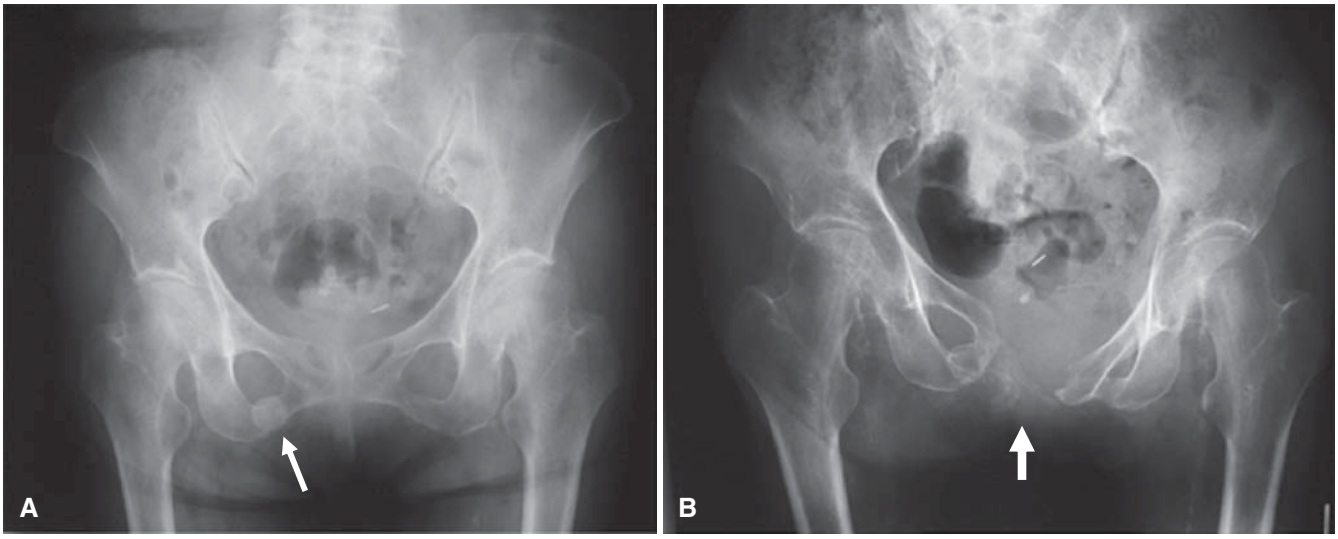

Fig. 1A,B. Case 1. **A** Plain anteroposterior pelvic radiography reveals a pelvic insufficiency fracture that occurred when the patient was 69 years old (*arrow*). **B** When the patient was 71 years old, she accidentally fell on the floor of her home. After the fall, pelvic ring disruption with symphysis pubis diastasis was noted (*arrow*). The pelvic ring disruption remained even after repositioning with pelvic slings and external fixation was attempted

remained unchanged. Three months after the injury, the patient was unable to walk unassisted and could ambulate only with a wheelchair. However, she was able to independently conduct activities of daily living, except for standing and walking.

Case 2

A 77-year-old woman with a 26-year history of seropositive RA had been treated with prednisone over 20 years as well as parenteral gold until October 2002. In December 2002, she was diagnosed with Felty syndrome (FS) based on findings of leukopenia with granulocytopenia and splenomegaly on admission, and she was treated with steroid pulse therapy. In February 2003, the patient suffered from high fever that had been ongoing for a few days. Blood cultures were positive for methicillin-sensitive *Staphylococcus aureus*. Therefore, she was treated with antibiotics (ampicillin/sulbactam) for 10 days, and her general condition improved. During a hospitalization in March 2003, she complained of groin and hip pain in the absence of trauma, and initial radiographs revealed no abnormal findings, so no aggressive therapy was administered and she was observed and treated with complete bed rest. However, she continued to experience pain on hip motion, and follow-up radiography 1 month after initial symptoms showed progressive osteolysis at both pubic rami and disruption of the superior aspect of the symphysis pubis (Fig. 2A). In addition, T1-weighted gadolinium magnetic resonance (MR) images with fat suppression demonstrated two abnormal lesions around the symphysis pubis (Fig. 2B). Moreover, due to the potential risk of infection due to FS-associated granulocytopenia, open biopsy on the pubic rami was performed for differen-

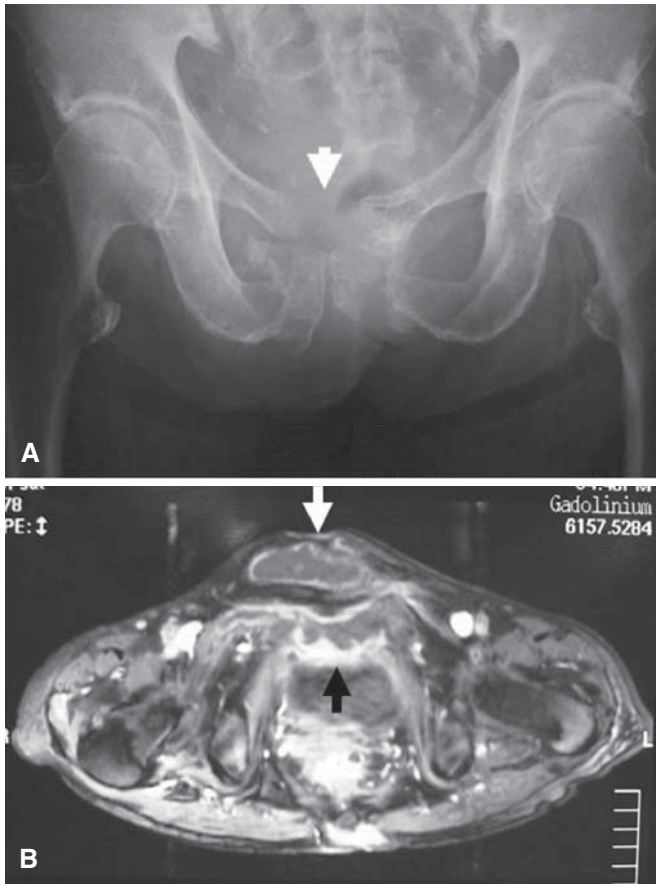

Fig. 2A,B. Case 2. **A** One month after onset of the initial symptom of pain around the buttocks and hip joints, radiography revealed progressive osteolysis at both pubic rami (*arrow*). **B** T1-weighted gadolinium magnetic resonance images with fat suppression demonstrated two abnormal lesions around the symphysis pubis (TR = 716, TE = 20). These lesions are characterized by hypointense central regions surrounded by a hyperintense wall (*arrows*), suggestive of abscesses

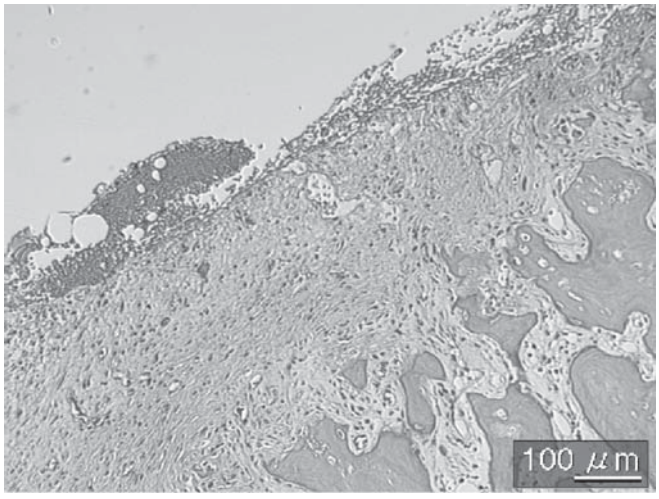

Fig. 3. Case 2. Histological findings of the pubic bone biopsy specimen showed osteomyelitis and were consistent with fracture healing (hematoxylin–eosin)

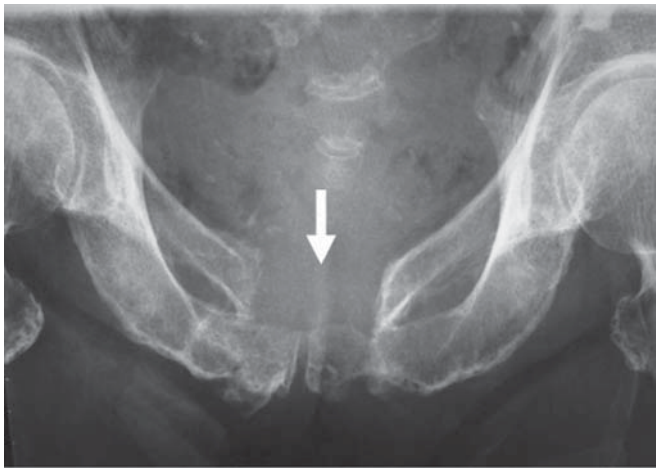

Fig. 4. Case 2. The patient received antibiotics and underwent surgical drainage. After 3 months, radiography showed that symphysis pubis diastasis remained, but inflammation was absent (*arrow*)

tial diagnosis. Microbiological culture of the resulting pubic bone biopsy specimen was positive for methicillin-sensitive *Staphylococcus aureus* as identical pathogens, suggesting hematogenous dissemination leading to pubic bone abscess, and histological findings revealed osteomyelitis and were consistent with fracture healing (Fig. 3). Laboratory findings at this time included a white blood cell (WBC) count of 6400/mm^3 and C-reactive protein (CRP) level of 12.4 mg/dl. The patient was first treated with the same antibiotics (ampicillin/sulbactam), which resulted in the temporary disappearance of inflammation. At this time, she was trained to use a wheelchair. However, the infectious inflammation recurred after 2 months. Therefore, she underwent surgical drainage and abscess resection with antibiotics, and returned to bed rest. Following these therapies, her pain decreased, and laboratory findings included an improved WBC count (3900/mm^3) and CRP level (0.66 mg/dl) in August. Radiography still showed osteolysis at both pubic rami and diastasis of the superior aspect of the symphysis pubis (Fig. 4), but no inflammation remained, and she had no complaint of any pain. After treatment, she was able to transfer from the bed to a wheelchair with assistance. The patient was discharged from our hospital with prednisolone 11 mg/day, and moved to another hospital for rehabilitation.

Discussion

In this article we present two RA patients with symphysis pubis disturbances. There are very few reports of disturbances of the symphysis pubis in RA patients.[4,5] Tauber et al. reported one RA patient with disruption of the symphysis pubis and fatigue fractures of the pelvis,[4] and Nicholas et al. reported two RA cases with fractures at or near the pubic symphysis.[5]

Case 1 was diagnosed as pelvic ring disruption with symphysis pubis diastasis. Symphysis pubis diastasis is usually caused by high-energy trauma such as motor vehicle accidents,[6] although a severe case of separation of the symphysis pubis during labor and delivery has also been reported.[7] The pubic symphysis is the midline cartilaginous joint uniting the superior rami of the left and right pubic bones, which are separated by a small gap and are connected by fibrocartilaginous tissue reinforced by ligaments. Soft tissue injuries such as Achilles tendinitis are commonly associated with long-term oral steroid use, particularly in older patients who use these drugs daily to treat systemic conditions.[8] Therefore, we consider that the combination of minor injury and long-term corticosteroid therapy caused the pelvic ring disruption with symphysis pubis diastasis observed in this patient.

Management of symphysis pubis diastasis remains controversial.[9] Conservative methods such as skeletal traction and external fixation generally produce satisfactory functional results.[10] However, it appears that precise reduction of pure sacroiliac lesions, which is difficult to achieve with conservative procedures, is critical for good functional results.[10] Indeed, reduction of the pelvic ring disruption with symphysis pubis diastasis in Case 1 was quite difficult. If this patient had presented in good general and respiratory condition, we would have recommended open reduction.

The patient described in Case 2 was diagnosed as symphysis pubis diastasis with pubic osteomyelitis. The osteomyelitis may have been related to her FS-associated granulocytopenia, which rendered her less resistant to infections. While osteomyelitis of the pubis typically occurs after pelvic surgery or trauma and has been reported in the literature,[11–13] no cases of FS complicated with pubic osteomyelitis have been previously reported. However, because of the potential risk of infection due to FS-associated granulocytopenia and previously positive blood cultures, open biopsy should be immediately performed to assess infection at the onset of symptoms. Moreover, beginning with the first treatment, such a case should undergo surgical drainage and curettage with antibiotics.

Both of the present cases had received long-term corticosteroid therapy, including pulse therapy—one for treatment of interstitial pneumonia, and the other for treatment of FS-associated granulocytopenia. Recently, differential expressions of steroid receptors in various ligaments have been studied.[14] The pubic symphysis is the joint uniting the pubic bones, which are connected by fibrocartilaginous tissue reinforced by ligaments. Therefore, we suggest that chronic long-term corticosteroid therapy may have played a causative role in the pubic symphysis diastasis observed in these cases.

In conclusion, this is a report of symphysis pubis disturbances in RA patients. The etiologies of the disturbances appear to be different for each case, although we believe that both long-term steroid use and additional, unidentified causative factors contributed. Further reports of similar cases must be accumulated to fully elucidate the underlying pathogenesis of symphysis pubis disturbances in RA.

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