

Progression of Hip Joint Deformity during the Management of Pelvic Insufficiency Fracture in a Patient with Rheumatoid Arthritis: A Case Report

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Abstract

Introduction: An insufficiency fracture is a type of non-traumatic fracture caused by bone fragility that most commonly occurs in the pelvis and lower extremities. Although sacral insufficiency fracture is not uncommon, its diagnosis is often delayed or missed. We here report the case of SIF with rheumatoid arthritis that occurred during the pre-operative waiting time for total hip arthroplasty.

Case presentation: A 57-year-old woman with rheumatoid arthritis presented with sacral insufficiency fracture that occurred during the pre-operative waiting time for total hip arthroplasty. During the four months after the onset of the sacral insufficiency fracture, her pubis, ischium and acetabulum fractured and deformity of her left hip joint gradually progressed. Total hip arthroplasty was performed using bone grafting onto the acetabulum from the autogenous femoral head. The findings upon clinical examination and the patient's assessment of function were excellent at the time of the most recent follow-up.

Conclusion: When total hip arthroplasty is performed in patients with severe osteoporosis or insufficiency fracture of the acetabulum, it is essential to avoid additional pelvic fractures at the time of setting the acetabular cup component. Moreover, as a total hip arthroplasty might itself be a predisposing causative factor of pelvic insufficiency fracture, recurrence of insufficiency fractures should be monitored in those patients with the history. Aggressive intervention for osteoporosis might be necessary to prevent insufficiency fracture development.

Keywords: Rheumatoid arthritis; Insufficiency fracture; Total hip arthroplasty

Introduction

An Insufficiency Fracture (IF) is a type of non-traumatic fracture caused by bone fragility [1] that most commonly occurs in the pelvis and lower extremities [2]. Although Sacral Insufficiency Fracture (SIF) is not uncommon, its diagnosis is often delayed or missed [3]. Moreover, SIF patients can suffer complications involving IF of the pubis, ischium [4-6].

We recently encountered a case of SIF with Rheumatoid Arthritis (RA) that occurred during the pre-operative waiting time for Total Hip Arthroplasty (THA). Hip joint deformity of the affected side had acutely worsened and we recognized that it was necessary to pay attention to its progression as well as protrusion of acetabuli after the SIF onset. The patient gave informed consent for data concerning her case to be submitted for publication.

Case Report

The patient was a 57-year-old woman who developed RA at the age of 27. She underwent a left elbow synovectomy at 49 years of age, a right Total Knee Arthroplasty (TKA) at 52, a left TKA at 53, and a right Total Hip Arthroplasty (THA) at 54. She was receiving

medication of 7.5 mg prednisolone daily, 4 mg methotrexate weekly and 35 mg alendronate weekly.

At the age of 55, she complained of left hip joint pain. Radiographs of the left hip joint were classified as grade III according to the Larsen classification system of RA [7] (Figure 1A).

A left THA was subsequently scheduled but, during the pre-operative waiting time, severe lumbosacral pain and subcutaneous fluctuation of the sacral region (approximately 6 cm in diameter) occurred without trauma. There were no neurological insufficiencies. Radiographs showed no evident fracture line on the pelvis (Figure 1A) or lumbar spine (Figure 2).

Computed tomography (CT) and 3D-CT scanning disclosed longitudinal fracture lines in the bilateral sacral alae, but there were no signs of osteolysis or space-occupying lesions (Figure 3).

Magnetic resonance imaging (MRI) revealed a low intensity area on T1-weighted images and a mixture of high intensity and low intensity areas on T2-weighted images consistent with fractures. Bone Mineral Density (BMD) and the T-score using dual-energy X-ray absorptiometry (DEXA) in the lumbar spines were 0.627 g/cm² and -3.8, respectively. Serum laboratory findings are shown in Table 1.

The severe lumbosacral pain greatly improved with conservative treatment using analgesics and subcutaneous fluctuation of the sacral region disappeared two months later. Radiographs revealed progression of the left femoral head deformity, and fracture of the

bilateral pubic and ischial rami and the medial wall of the left acetabulum (Figure 1B and C). CT scans at four months after the onset of SIF showed no displacement of SIF but the formation of a callus was apparent (Figure 4).

Seven months after the onset of SIF, a left THA was performed by grafting an autogeneous femoral head onto the acetabulum. Post-operatively, a fracture of the left greater femoral trochanter occurred with no other symptoms. One year post-surgery, the allografts appeared to have united radiographically with no radiolucent lines visible between the graft and the acetabular cup (Figure 2D). The patient experienced complete relief of lumbosacral and hip pain and can walk with a T-cane. She now receives a recombinant human form of parathyroid hormone (teriparatide, 600 µg per day) to medicate against osteoporosis. BMD in the lumbar spines improved from 0.627 g/cm² to 0.704 g/cm² within a year of using teriparatide.

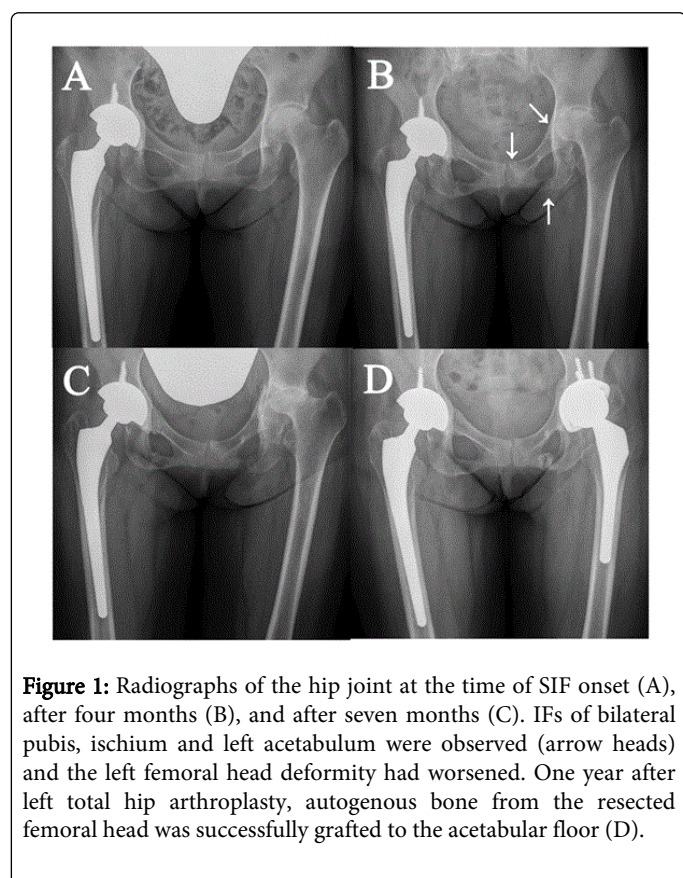


Figure 1: Radiographs of the hip joint at the time of SIF onset (A), after four months (B), and after seven months (C). IFs of bilateral pubis, ischium and left acetabulum were observed (arrow heads) and the left femoral head deformity had worsened. One year after left total hip arthroplasty, autogenous bone from the resected femoral head was successfully grafted to the acetabular floor (D).

Discussion

A characteristic of SIF lies in its difficulty to be diagnosed upon first examination [8], which is attributable to its nonspecific symptoms, unclear fracture lines, overlap with signs of intestinal gas pooling on radiographs [9,10], and a tendency to focus on hip joint or lumbar spine diseases [1]. It is also difficult to distinguish SIF from inflammatory diseases and primary/metastatic bone tumors [11,12]. However, it is important to keep the possibility of SIF in mind in a patient with lumbosacral pain. In the present case, subcutaneous fluctuation of the sacral region caused by hematoma after sacral fracture was helpful in the correct diagnosis of SIF.

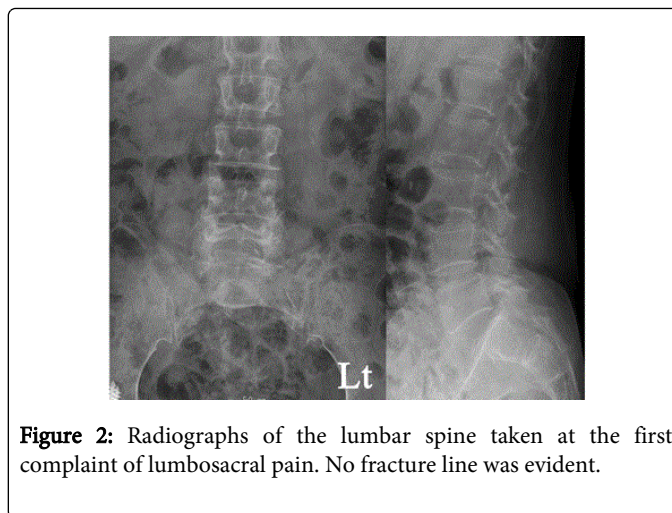


Figure 2: Radiographs of the lumbar spine taken at the first complaint of lumbosacral pain. No fracture line was evident.

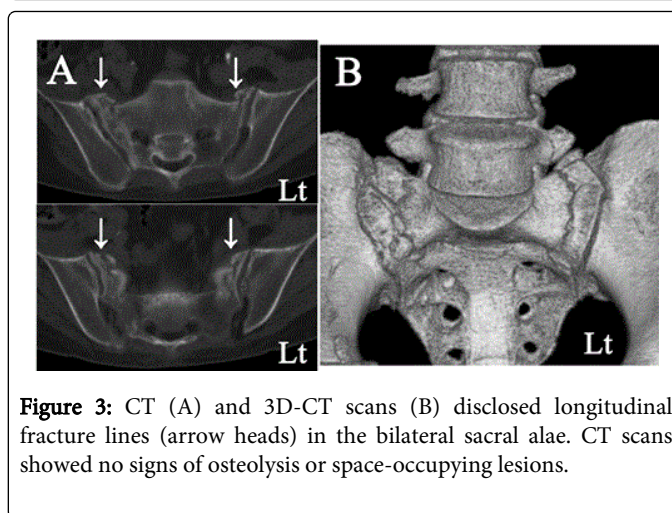


Figure 3: CT (A) and 3D-CT scans (B) disclosed longitudinal fracture lines (arrow heads) in the bilateral sacral alae. CT scans showed no signs of osteolysis or space-occupying lesions.

Parameter	Measurement	Standard range
White blood cells	9800/µl	3000–8500/µl
Red blood cells	458×10 ⁴ /µl	378–499×10 ⁴ /µl
Hemoglobin	13.7 g/dl	10.8–14.9 g/dl
Platelets	27.4×10 ⁴ /µl	15.0–36.1×10 ⁴ /µl
C-reactive protein	1.66 mg/dl	~0.30 mg/dl
Erythrocyte sedimentation rate	7 mm/h	~16 mm/h
Rheumatoid factor	368 U/ml	~10 U/ml
Matrix metalloproteinase-3	117.1 ng/ml	17.3–59.7 ng/ml
Alkaline phosphatase	1036 U/l	115–359 U/l
Calcium	9.5 mg/dl	8.7–10.3 mg/dl

Table 1: Serum laboratory findings at the onset of sacral insufficiency fracture

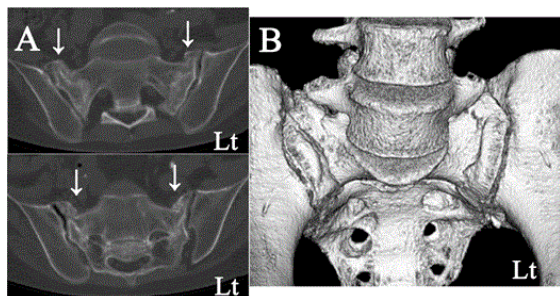


Figure 4: CT (A) and 3D-CT scans (B) four months after the onset of SIF revealed no displacement of SIF but showed the formation of a callus.

Routine radiography of the spine and pelvis is usually inconclusive and offers limited diagnostic sensitivity. Although CT is considered the gold standard modality for diagnosing occult SIF as it can delineate bony details [13], it can also be useful to conduct screening using MRI and bone scintigraphy. Indeed, a previous study reported that bone scintigraphy revealed the 'Honda sign' in 43% of all cases with SIF [14].

Treatment of SIF is usually conservative, for example, percutaneous sacroplasty was previously used as an effective strategy to treat persistent lumbosacral pain [15,16]. In the present case, BMD did not increase sufficiently following alendronate treatment, so teriparatide, a recombinant form of parathyroid hormone, was considered instead as it has been shown to reduce severe back pain after a vertebral fracture [17]. Within a year of teriparatide treatment, the BMD of the current patient increased from 0.627 g/cm² to 0.704 g/cm². Such adequate management of osteoporosis can help prevent subsequent occurrences of IF.

In RA patients, the sacrum and the metatarsal bones are common sites of IF [18]. Therefore the possibility of SIF must be considered in all patients with long-standing RA, especially in women with poorly controlled disease and those undergoing oral glucocorticoid therapy [19,20]. In the present case, IFs of the pubic body, ischium and acetabulum developed after the onset of SIF. It has previously been shown that the likelihood of developing pubic IF increases if stress accumulates near the pubic symphysis from anterior dislocation of the load caused by SIF-associated pelvic ring instability [9,21,22].

Progression of the hip joint deformity was observed in the current patient after the onset of pelvic IFs. We carried out a literature search of this topic and found very few reports of SIF as a complication of progressive RA hip joint deformity [19]. This progression could be attributable to unbalanced stress concentrations on the hip joint from changes in pelvic inclination and lumbar spine curving, as well as RA-induced joint destruction. When THA is performed in patients with severe osteoporosis or IF of the acetabulum, it is essential to avoid additional pelvic fractures at the time of setting the acetabular cup component. Moreover, as THA might itself be a predisposing causative factor of SIF [23], recurrence of pelvic IF should be monitored in those patients with a history of pelvic IF.

In conclusion, aggressive intervention for osteoporosis might be necessary to prevent IF development and it is particularly important to monitor the progression of hip joint deformity after SIF onset in patients with RA. Successful post-operative results, as in the present

study, can be achieved by cautious maneuvers and acetabular bone grafting during THA.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing Interests

The authors declare that they have no competing interests.

Author Contributions

KI, YWN, MN, JM, HG and MK were involved in the conception, design and interpretation. KI and YWN wrote the manuscript. KI, JM and TO collected data, reviewed relevant published reports and provided the images. All authors read and approved the final manuscript.

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