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CASE REPORT

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# Acute destruction of the hip joints and rapid resorption of femoral head in patients with rheumatoid arthritis

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**Abstract** We report three rheumatoid arthritis (RA) cases with acute destruction of hip joint and rapid resorption of femoral head. The condition occurred in less than 6 months and closely resembled rapid destructive coxarthrosis. All three patients were postmenopausal women with active RA who had been taking steroids. Two of the patients were taking prednisolone (PSL) of over 20 mg as maximum dose per day, and all patients were resistant to disease-modifying anti-rheumatic drugs (DMARDs). Other than the problems of their hip joints, one had a giant bursitis around the pathological side of the hip joint, another had multiple rheumatoid nodules and skin infarction, and the other suffered from insufficiency fracture of the contralateral femoral subcapital lesion. As a result, all of them had total hip arthroplasty. We recommend taking repetitive radiographs for RA patients with continuing severe hip pain.

**Key words** Femoral head resorption · Rapidly destructive coxarthrosis (RDC) · Rheumatoid arthritis (RA)

## Introduction

Hip joint affects only 5%–15% of all rheumatoid arthritis (RA) cases[1] and its destructive process is gradually progressing. A disappearance of the femoral head is sometimes observed but the destructive changes are usually shown to be slow. However, we had a few rare chances of encountering three RA cases with an acute destruction of the hip joints and a rapid resorption of the femoral head. In this paper, we report the clinical features, pathogenesis, and management of these patients, of whom the hip joint conditions were very similar to rapidly destructive coxarthrosis (RDC).[2]

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## Case reports

Our three patients were all postmenopausal women who were treated at our institute from 1995 to 2005. They had no previous history of major trauma.

### Case 1

The first patient was a 59-year-old woman with a 9-year history of RA who had been treated with daily prednisolone (PSL) of 5 mg per day and methotrexate (MTX) of 5 mg per week. She was 155.0 cm in height and 62.0 kg in weight. She had a total right knee arthroplasty 6 years after the onset. Nine years after onset, she encountered some pain in the left hip joint. Plain radiographic image in November 1999 showed only joint space narrowing (Fig. 1A). Later, her left hip joint pain worsened and plain radiographic image in January 2000 showed an initial collapse of the femoral head (Fig. 1B) but by March 2000, almost the whole femoral head had disappeared (Fig. 1C). The condition deteriorated in less than 6 months. At this time, the patient felt a soft tumor anterior to her left hip joint and she could not walk by herself. Therefore, total hip arthroplasty was scheduled. Laboratory findings in March 2000 on admission showed erythrocyte sedimentation rate (ESR) at 60 mm/h, C-reactive protein (CRP) at 5.2 mg/dl, and RA hemagglutination test (RAHA) of ×40. Also, the range of motion of the left hip joint was 0° in extension, 120° in flexion, 15° in abduction, and 0° in adduction. The length of the spino-malleolar distance (SMD) was 87.0 cm in the right extremity and 84.0 cm in the left. Steinbrocker functional class of this patient was III at this point. Magnetic resonance imaging (MRI) showed a soft tissue tumor anterior to the hip joint (Fig. 2A). A noncemented total hip arthroplasty and a resection of bursitis were performed 4 months after the onset

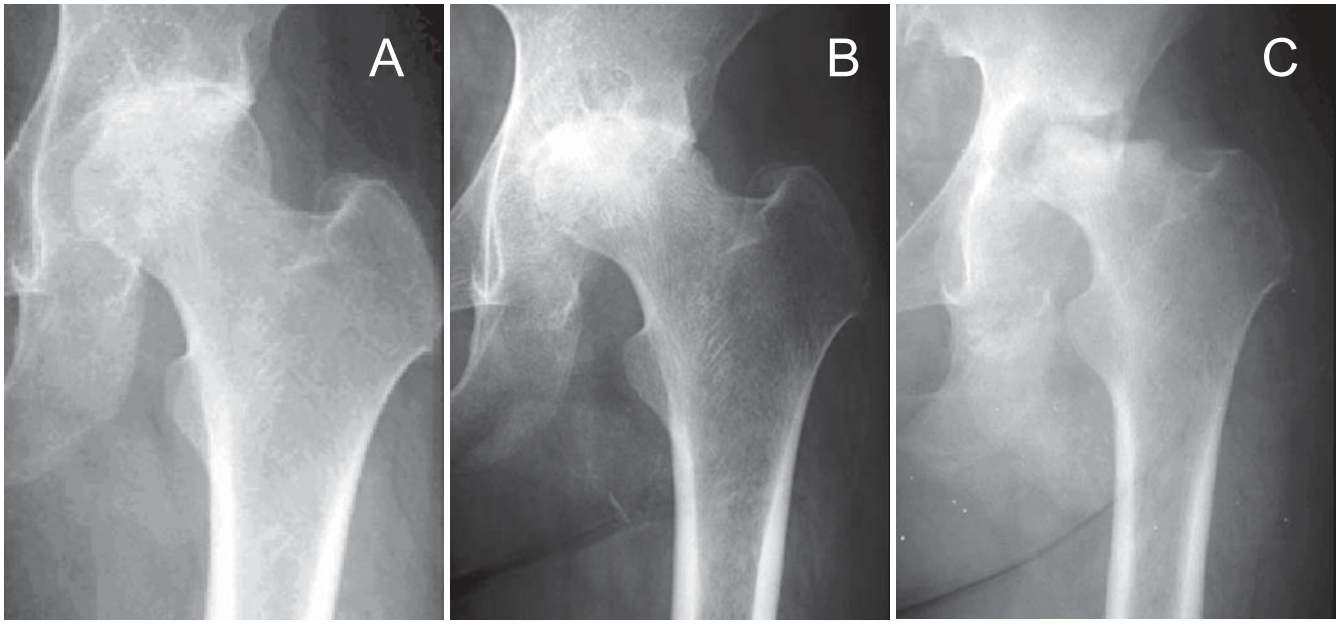


**Fig. 1A–C.** Case 1. **A** Anteroposterior view of the left hip joint of a 59-year-old woman with a 9-year history of rheumatoid arthritis (RA), obtained at the onset of left coxalgia. The radiograph shows space narrowing in the joint and sclerosis of the femoral head. **B** Anteroposterior view of the same patient obtained 2 months later shows the collapse of the femoral head and moderate sclerosis with no osteophytes. **C** Anteroposterior view of the patient obtained again 2 months after the second radiography shows almost total disappearance of the femoral head and osteolysis of the pelvic side

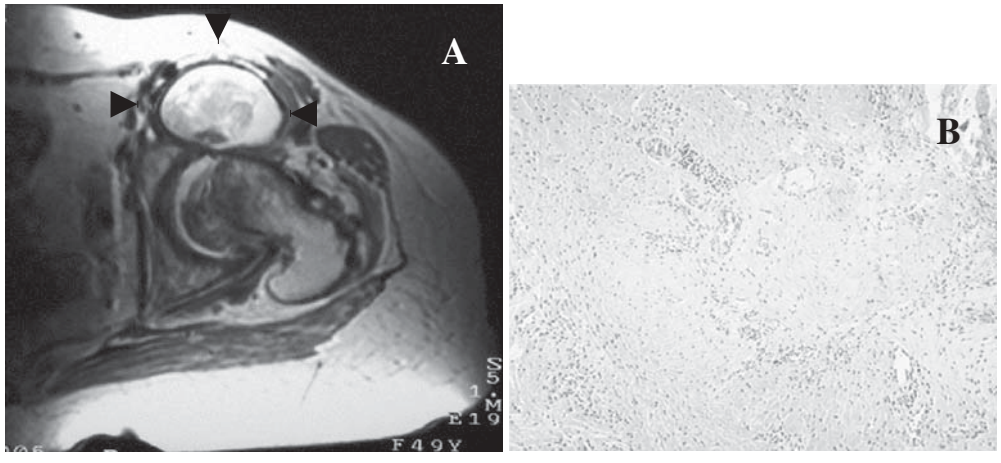


**Fig. 2A,B.** Case 1. **A** Magnetic resonance imaging shows a soft tumor anterior aspect of the patient's left hip joint (*arrowheads*). Density inside the tumor was not the same. **B** In the bursa and hip joint there were many rice bodies, thickening of the synovium, and necrotic bone fragments. The pathological findings show nonspecific inflammatory cells in the synovial tissue. There was no crystal deposition in the synovium (×40, Hematoxylin and Eosin Stain)

of the patient's left hip pain. In the bursa and hip joint there were many rice bodies, thickening of the synovium, and necrotic bone fragments. Biopsy of the peak inflammatory intensity area was obtained and immunohistopathologically analyzed. The pathologic findings showed nonspecific inflammatory cells in the synovial tissue. There was no crystal deposition in the synovium (Fig. 2B). After operation and rehabilitation, she could walk by herself using a walking stick.

## Case 2

The second patient was a 62-year-old woman with a 9-year history of RA. She was 145.6 cm in height and 50.3 kg in weight. Her chief complaint was pain in the left hip joint and gait disability. She had been treated with disease-modifying antirheumatic drugs (DMARDs) but because her disease activity was high, the drug response was low. Multiple rheumatoid nodules, skin infarction, purpura, and

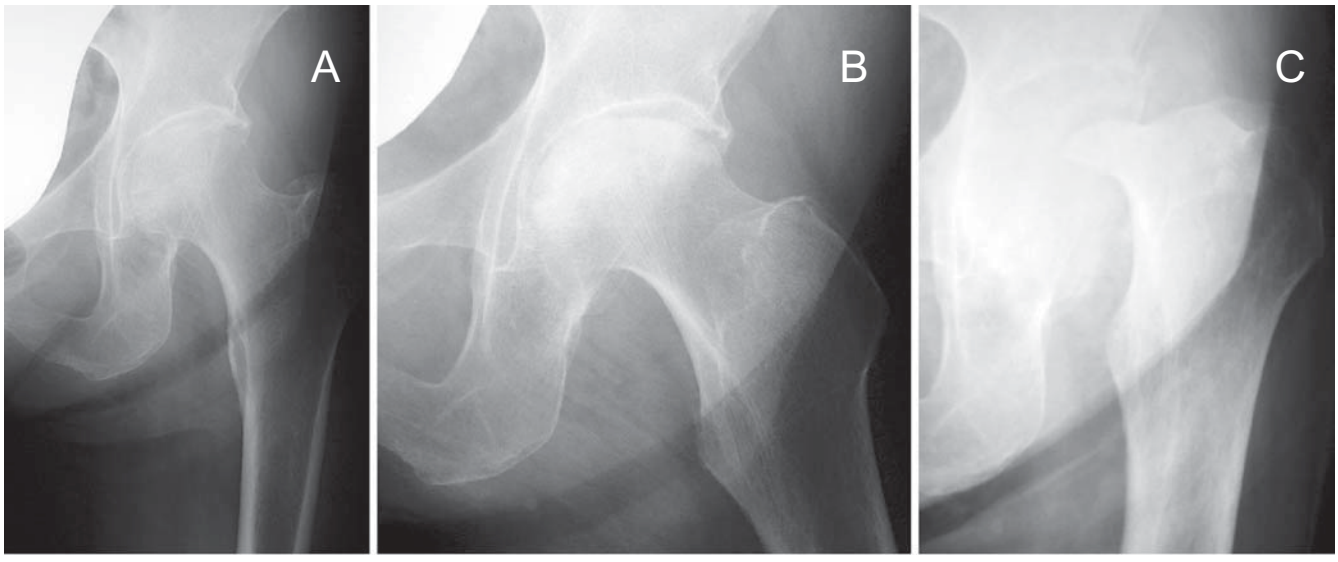


**Fig. 3A–C.** Case 2. **A** Anteroposterior view of the left hip joint of a 62-year-old woman with a 9-year history of RA, obtained at the onset of left coxalgia. The radiograph shows space narrowing in the joint. The patient's chief complaint was pain in the left hip joint, and gait disability. **B** Anteroposterior view obtained 4 months later shows that the space narrowing in the joint has progressed and moderate sclerosis of femoral head was seen. **C** Anteroposterior view obtained again 3 months after the second radiography shows that almost all the femoral head had disappeared. The femoral bone moved proximally and the osteolytic change of pelvic side was observed

nerve inflammation were observed in 1999. Therefore, PSL of 30 mg per day and cyclophosphamide of 50 mg per day was initiated. In 2001, the PSL was reduced to 9–11 mg per day. She had a total right hip arthroplasty in March 2004. In the nonoperated side of the hip joint, destructive process gradually progressed, but a disappearance of the femoral head was not observed. The patient began to feel some pain in her left hip joint in February 2005. Plain radiographic image showed a narrowing of the joint space (Fig. 3A). Her condition was observed for a while but the pain worsened and could not be relieved with nonsteroidal anti-inflammatory drugs (NSAIDs). In June 2005, the radiographic image showed joint space narrowing and sclerotic change of the femoral head (Fig. 3B). Total hip arthroplasty was scheduled. Laboratory findings on admission in August 2005 showed the following values: ESR 20 mm/h, CRP 0.1 mg/dl, and RAHA ×40, and the range of motion of the left hip joint was 0° in extension, 95° in flexion, 30° in abduction, and 10° in adduction. Steinbrocker functional class of this patient was II before the episode. However, she could not walk by herself any more at this point. The image at admission surprisingly showed that the femoral head had totally disappeared and the femoral bone had moved proximally (Fig. 3C). The length of the SMD was 76.0 cm in the right extremity and 75.0 cm in the left side. A noncemented total hip arthroplasty was performed, which was dated 6 months after the onset of the patient's left hip pain. In the patient's hip joint, some pieces of cartilage fragments and thickening of the capsule were observed at operation. The pathological findings of the synovial tissue showed severe inflammatory change with lymphoplasmacytic infiltration. No crystal deposition in the capsule or synovium was found (Fig. 4). After operation and rehabilitation, she was able to walk by herself with a walking stick again.

## Case 3

The third patient was a 52-year-old woman with an 18-year history of RA. She was 158.2 cm in height and 52.2 kg in weight. She had been treated with a daily PSL dosage of 5 mg per day since she was diagnosed with RA in 1986. Her high disease activity caused resistance to any DMARD. Therefore, a PSL dose of 10 mg was increased to 20 mg per day. Her level of CRP was under control from 0.1 to 1.0 mg/dl. In July 2001, she had a right knee joint arthroplasty. In October of the same year, insufficiency fracture of the right femoral neck was diagnosed (Fig. 5A–D). She had no injury as far as she could remember but she was beginning to feel coxalgia in the right side of her hip. Anteroposterior view of the right hip joint showed an almost normal hip joint at the onset (Fig. 5A). However, we found insufficiency fracture at the subcapital lesion 1 month later (Fig. 5B), collapse of femoral head 4 months later (Fig. 5C), and finally, the femoral head completely disappeared 8 months after the onset of initial pain in the right hip joint (Fig. 5D). We performed a right total hip replacement. In September 2002, the patient again felt pain in her left hip joint without any injury (Fig. 6A), which got much worse with time. The plain radiographic image showed initially a collapse of

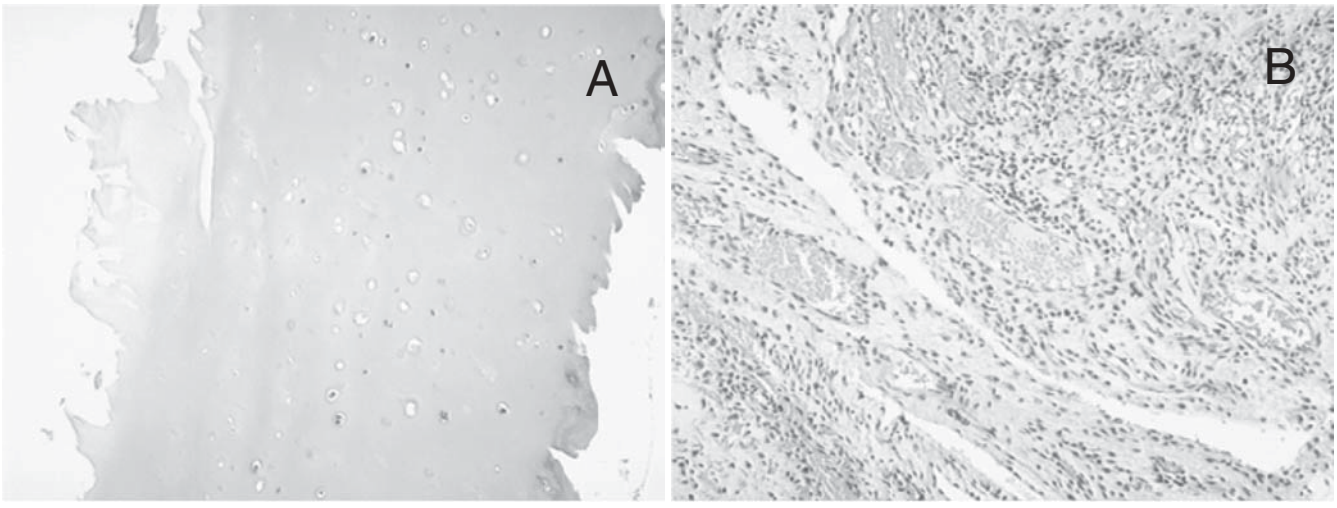


**Fig. 4A,B.** Case 2. **A** In the hip joint, some pieces of cartilage were observed (×40). **B** The pathological findings of the synovial tissue show severe inflammatory change with lymphoplasmacytic infiltration (×40, Hematoxylin and Eosin Stain)

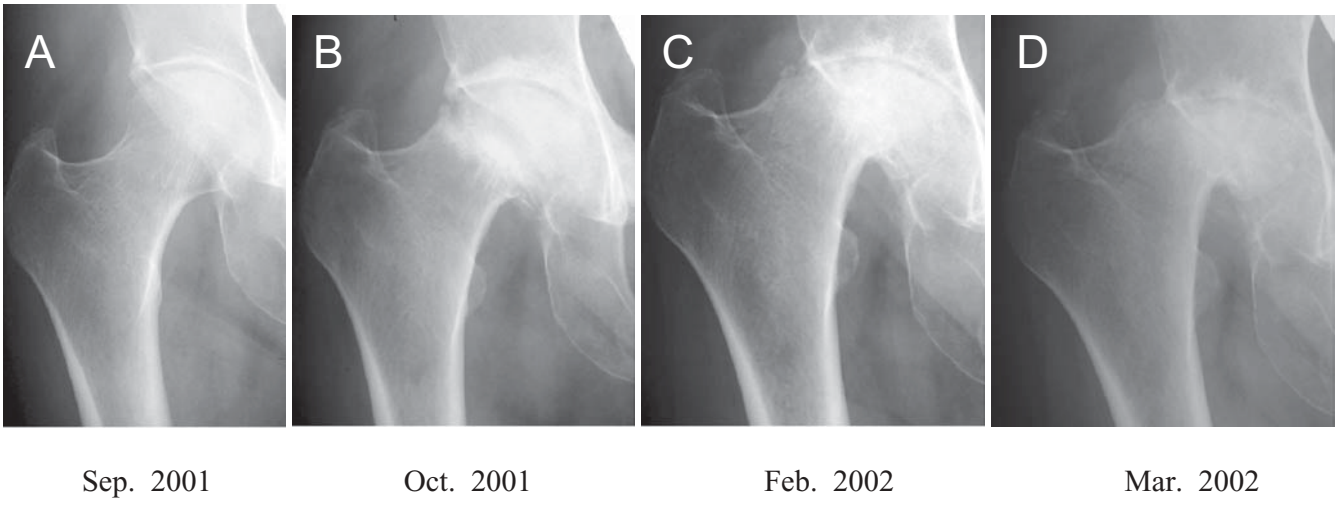


**Fig. 5A–D.** Case 3. **A** Anteroposterior view of the right hip joint of a 52-year-old woman with an 18-year history of RA, obtained at the onset of right coxalgia. The radiograph shows an almost-normal hip joint. **B** Anteroposterior view obtained 1 month later shows insufficiency fracture at the subcapital lesion. **C** Anteroposterior view obtained again 4 months later shows the collapse of femoral head and joint-joint space narrowing. **D** Finally, the femoral head disappeared completely 8 months after the initial pain in the right hip joint

the femoral head in October 2002 (Fig. 6B). The rapid destructive process in the left hip joint was progressing, and almost half of the femoral head had disappeared by January 2003 (Fig. 6C). The patient could not walk by herself and so another total hip arthroplasty was scheduled to the left hip joint. On admission, the functional class according to Steinbrocker was IV. Laboratory findings showed the following values: ESR at 18.8 mm/h, CRP at 0.2 mg/dl, and RAHA of ×320. The range of motion of the left hip joint was 10° in extension, 130° in flexion, 40° in abduction, and 10° in adduction. The length of the SMD was 87.0 cm in the right extremity and 84.0 cm in the left. This patient also underwent a noncemented total hip arthroplasty, and her clinical result has been good. She is able to walk by herself again.

## Discussion

We described three cases of acute hip joint destruction and rapid femoral head resorption in RA. Osteoporosis, burrowing and inward protrusion of the acetabulum, and collapse and flattening of the femoral head are found in patients with destructive arthritis, typically in rheumatoid hip joints. The sequential process of femoral head destruction would start with a collapse, flattening, and finally a total disappearance.[3] The usual destructive progress is shown to be gradual. Therefore, in this paper we reported three RA cases with acute destruction of the hip joint and rapid femoral head resorption in less than 6 months. The patterns of their conditions were very similar to that of RDC.

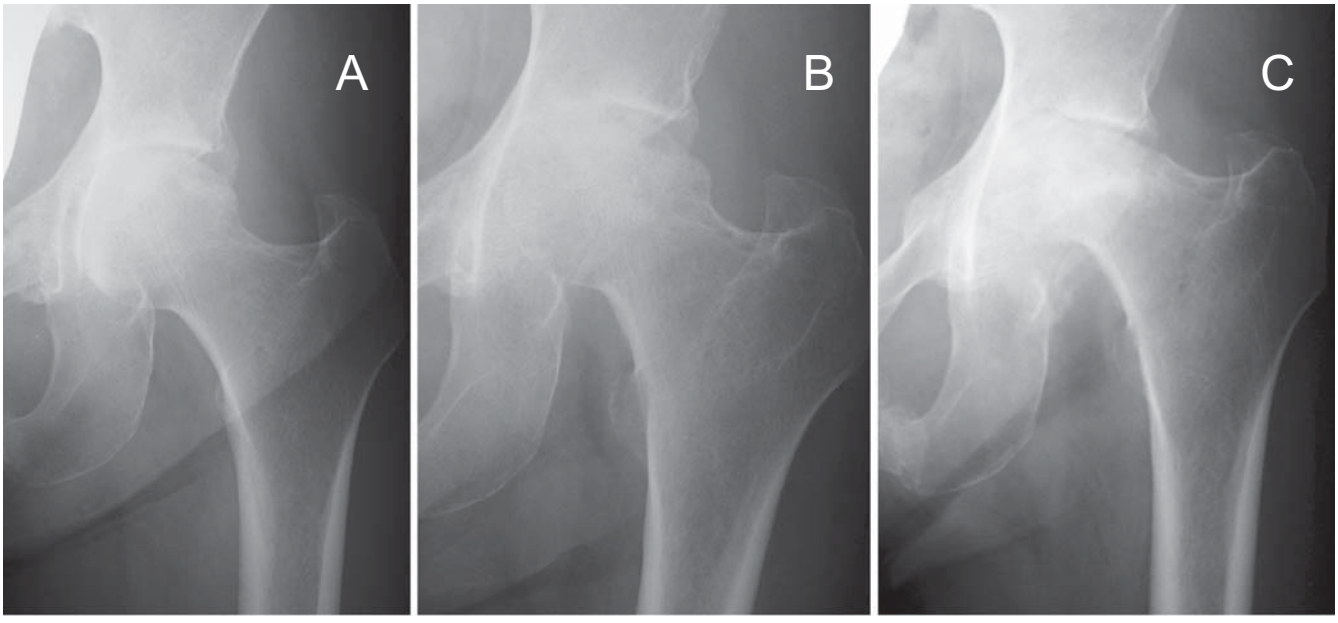


**Fig. 6A–C.** Case 3. **A** Anteroposterior view of the left hip joint of the patient obtained at the onset of left coxalgia. The radiograph shows a slight space narrowing in the joint. **B** Anteroposterior view obtained 2 months later shows that the space narrowing had progressed and moderate sclerosis of femoral head was observed. **C** Anteroposterior view obtained again 2 months after the second radiography shows that almost half of the femoral head had disappeared

**Table 1.** Background of three rheumatoid arthritis (RA) patients with acute destruction of hip joint and rapid resorption of femoral head

| | Case 1 | Case 2 | Case 3 |
|---|---|---|---|
| Age (years) | 59 | 62 | 52 |
| Sex | Female | Female | Female |
| Side of hip joint | Left | Left | Left |
| RA duration (years) | 9 | 9 | 17 |
| Laboratory findings on operation | | | |
| ESR (mm/h) | 60 | 20 | 18.8 |
| CRP (mg/dl) | 5.2 | 0.1 | 0.2 |
| RAHA | ×40 | ×40 | ×320 |
| PSL (maximum dose per day) | 5 mg | 30 mg | 20 mg |
| PSL (total dose) | 16 420 | 59 895 | 58 765 |
| DMARDs (prescribed in past) | Methotrexate<br>Cyclophosphamide | Methotrexate, other DMARDs | Methotrexate, other DMARDs |
| Other total joint arthroplasty | Right TKA | Right THA | Right TKA, right THA |
| Extra-articular disease manifestations | Giant bursitis<br>Skin infarction<br>Purpura<br>Nerve inflammation | Multiple rheumatoid nodules | Insufficiency fracture at the right femoral subcapital lesion |

TKA, total knee arthroplasty; THA, total hip arthroplasty; ESR, erythrocyte sedimentation rate; CRP, C-reactive protein; HA, hemagglutination; PSL, prednisolone; DMARDs, disease-modifying antirheumatic drugs

Rapidly destructive coxarthrosis was first reported by Postel and Kerboull[2] but its etiology has not been clarified. Rapidly destructive coxarthrosis is characterized by a rapid joint destruction (within 6–12 months of the onset of symptoms), and a disappearance of the joint space without other forms of rapid destructive arthropathy.[2,4,5] The radiologic changes are dramatic and may mimic neuropathic or septic arthropathy. Since the term RDC was first used, the condition was regarded as a rare syndrome that involved rapid and total deterioration of both the acetabular and femoral aspects of the hip joint.[6] Rapidly destructive coxarthrosis has been defined with different names such as "rapidly destructive coxarthrosis,"[7] "rapidly destructive arthrosis of the hip,"[8,9] "rapidly destructive hip disease,"[6] and "rapidly destructive osteoarthritis of the hip."[10–12] In the present paper we use the term "rapidly destructive coxarthrosis (RDC)," the most popular term. Rapidly destructive coxarthrosis may be caused by osteonecrosis of the femoral head, acetabular dysplasia, pelvic tilt, and RA.[13] Whether RDC is an established entity or should be classified as a

subset of osteoarthrosis or osteonecrosis of the femoral head is unclear.[14] Ishiguro et al. also reported a 48-year-old male hemophiliac who exhibited hip arthropathy that was similar to rapidly destructive arthropathy.[14] We identified our RA patients as RDC by their acute destruction of the hip joint and rapid femoral head resorption. Our three patients shared the following common characteristics: (1) all patients were postmenopausal women, and no male patient was observed; (2) their RA disease activities were high and were resistant to DMARDs; (3) they had been taking steroids.

Bone mineral density of postmenopausal women is usually lower than men. Recently, Yamamoto and Bullough[15] have proved that subchondral insufficiency fracture of the femoral head occurs in elderly women with osteoporosis. They speculated that insufficiency fracture resulting from osteopenia might lead to rapid breakdown of the hip joint. Niimi et al. also reported that subchondral insufficiency fracture of the femoral head seemed to be an important preliminary event in the development of RDC.[16] Previously, we reported that generalized bone loss occurred in postmenopausal women with RA and these patients were characterized by bone resorption that correlated with a high level of inflammation.[17] Therefore, we speculate that highly inflammatory coxarthritis may cause an osteoporotic change in the hip joint, and lead to rapid resorption of femoral head. We think subcapital insufficiency fracture due to osteoporotic change might be a preliminary event in the development of rapidly destructive coxopathy in active RA. However, insufficiency fracture of the right subcapital femoral lesion in fact occurred before the presence of the rapid destructive arthritis in the left hip joint only in case 3. Definite insufficiency fracture of the femoral head was not observed in the other two cases. On the other hand, highly inflammatory destructive coxarthritis in case 1 could have channeled debris and pannus into the iliopsoas bursa, and generated a giant bursitis-like soft tissue tumor.

Long-term steroid therapy may also be responsible for rapid femoral head resorption. The use of steroids and their influence on bone mineral density is widely debated. Most cross-sectional studies agreed that the cumulative dose of PSL is an important determinant of osteopenia in RA.[18,19] This may also explain why femoral head resorption appeared in these cases. The total amounts of PSL taken by these three patients were over 10000 mg, and two of them were taking over 50000 mg.

To conclude, we recommend that orthopedic surgeons be aware of a pathological situation similar to RDC and take repetitive radiographs for RA patients with continual severe hip pain. We believe our report could contribute by adding to the limited literature on the knowledge of RDC.

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