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Case Report

Rapidly Destructive Arthrosis of Bilateral Humeral Heads Caused by Subchondral Insufficiency Fracture

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The bilateral shoulder pain of an 81-year-old Japanese woman due to falls persisted despite celecoxib treatment, and plain X-rays later showed bilateral collapsed humeral heads. After ruling out osteoarthritis, infectious arthritis, crystal-induced arthritis, neuropathic arthropathy, and osteonecrosis, we diagnosed bilateral shoulder joint rapidly destructive arthrosis (RDA). Lumbar bone mineral density showed very low T-score (−4.1). Primary osteoporosis was observed. Histology of biopsied humeral head indicated the features of fracture healing process: callus formation and osteoclasts without empty lacunae. Her history thus included an insufficiency fracture due to severe osteoporosis. Bilateral humeral head replacement was performed; her shoulder joint function improved. This case is extremely rare in that RDA was caused by simultaneous bilateral shoulder joint collapse within a very short time, with minimal or low mechanical stress and severe osteoporosis.

Key words: rapidly destructive arthrosis, differential diagnosis, humeral head replacement, osteoporosis, shoulder joint

Rapidly destructive arthrosis (RDA) has been reported to occur in the hip joint [1]. Rapid destruction of the hip joint is defined as destruction of >2 mm or 50% joint space narrowing within 1 year. RDA in the shoulder joint is rare and is characterized by a rapid collapse of the humeral head, with no evidence of other non-septic articular arthropathies [2]. In 1982, Lequesne *et al.* described the cases of six elderly female patients with similar clinical and radiological pictures including osteolysis of 24-100% of the articular surface within a 6-month period, rotator cuff disintegration, and they defined these disorders as "rapid destructive arthritis of the shoulder" [3]. Nguyen provided a report of patients with RDA of the shoulder and reviewed the relevant literature [4]; RDA of the shoulder affected mainly elderly patients and was female-dominant with a rapid development of joint destruction in the presence of a large noninflammatory effusion (often blood-stained) containing large amounts of hydroxyapatite crystals.

RDA is observed mainly in the hip joints; other joints involved are the shoulder, knee, ankle, wrist, and elbow [5]. A case series of bilateral shoulder RDA was reported, but shoulder RDA remains very rare [6]. Here, we report a case of RDA in bilateral humeral heads combined with a subchondral insufficiency fracture most likely caused by severe primary osteoporosis. This case is notable in that we performed a detailed differential diagnosis using several imaging techniques, *i.e.*, plain radiography, computed tomography (CT), and magnetic resonance imaging (MRI) and laboratory

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examinations, *i.e.*, evaluations of inflammatory and autoantibody markers for rheumatic diseases and bone metabolic markers. We also ascertained the detailed pathology of RDA.

Case Presentation

An 81-year-old Japanese female developed bilateral shoulder pain and a limited range of motion in both shoulder joints. She had no history of alcohol intake, smoking, methylprednisolone treatment, hypertension, diabetes mellitus, rheumatoid arthritis, or decompression. She had never been diagnosed with osteoporosis and had not undergone treatment for osteoporosis. She had directly injured her left shoulder when she fell (defined as day 0). Thirteen days later, in another incident, she lost her balance but supported her body with her right arm to prevent a fall. She initially presented with left shoulder pain followed by right shoulder pain. The bilateral shoulder pain persisted despite the administration of celecoxib (Celecox®, Astellas, Tokyo, Japan) during the initial appointment. At day 41, she visited our clinic. No redness or local heat was noted. No radiography had been performed on the bilateral shoulders prior to this visit. The passive ranges of motion in the shoulders were as follows: right: 70°/40°, left: 60°/40° during flexion/extension and right: 50°/20°, left: 70/20° during abduction/adduction.

The laboratory examination (Table 1) revealed no remarkable findings in the patient's cell blood count but did show an elevated serum alkaline phosphatase (ALP) (538 IU/l) level, suggesting the presence of a fracture. The levels of uric acid (3.7 mg/dl), C-reactive protein (CRP) (0.08 mg/dl), serum MMP-3 (24.9 ng/ml), and anti-cyclic citrullinated peptide (CCP) antibody (<0.5 U/ml) were within the normal limits. However, the patient's rheumatoid factor (RF) was slightly elevated (44.3 IU/ml). Thus, an inflammatory response and serological changes indicating rheumatoid arthritis and similar disorders such as polymyalgia rheumatica and collagen diseases were not detected. In addition, the aspirated synovial fluid from the right shoulder joint was yellow and clear, and crystals such as uric acid and calcium pyrophosphate dihydrate (CPPD) were not detected. A bacterial culture examination was conducted, with negative results.

The assessment of bone turnover markers revealed

Table 1 Laboratory findings

Item	Value	Unit	Item	Value	Unit
WBC	7,410	/μl	TP	7.3	g/dl
Neutrophil	56.6	%	Alb	3.7	g/dl
Lymphocyte	35.6	%	CK	27	U/l
Eosinophil	1.3	%	AST	21	U/l
Basophil	0.1	%	ALT	13	U/l
Monocyte	6.4	%	LDH	182	U/l
RBC	434	x10⁴/μl	ALP	538	U/l
Plt	20.4	x10⁴/μl	γ-GTP	16	U/l
CRP	0.08	mg/dl	BUN	18	mg/dl
ESR	17	mm/hr	Cre	0.49	mg/dl
RF	44.3	IU/ml	UA	3.7	mg/dl
Antinuclear antibody	11.8	x	Ca	9.6	mg/dl
MMP-3	24.9	ng/ml	iP	3.4	mg/dl
Anti-CCP antibody	<0.5	U/ml			
Bone metabolism related markers	Value	Unit			
BAP	20.9	U/l			
TRACP-5b	1,030	mU/dl			
Intact PTH	27	pg/ml			
ucOC	6.4	ng/ml			
Urinary NTx	117.6	nmolBCE/mmolCr			
deoxypyridinoline	18.6	nM/mM Cre			

that the patient's bone-specific alkaline phosphatase (BAP) level (20.9 U/l) was higher than the reference value (8.1-10.5 U/l). The levels of the following bone resorption markers were very elevated: TRACP-5b (1,030 mU/dl), urinary amino-terminal collagen cross-links (NTx) (117.6 nmolBCE/mmolCr), and deoxypyridinoline (18.6 nM/mM Cre). The level of uncarboxylated osteocalcin (ucOC) (6.4 ng/ml) was slightly higher than the reference value (Table 1). These data indicated an upregulation in bone resorption and a vitamin K insufficiency.

The radiographic findings at the first visit showed that the bilateral humeral heads were severely damaged (Fig. 1A, B). CT images of the right shoulder showed that the humeral head had collapsed but the glenoid was intact (Fig. 1C). MRI of the right shoulder revealed synovial fluid effusion, but neither synovitis proliferation nor rotator cuff injury was detected (Fig. 1D, E). No symptoms of arthritis were noted in the other joints, *i.e.*, hands, wrists, and feet. Cervical MRI demonstrated no syringomyelia, and a Treponema pallidum hemagglutination (TPHA) test was negative. We thus ruled out neuropathic arthropathies (Charcot joint) such as tabes dorsalis as a diagnosis.

On the basis of the patient's bilateral rapid humeral head collapse that occurred within 6 weeks from the first traumatic episode, we diagnosed bilateral RDA in her shoulders. We performed a humeral hemiarthroplasty at 3 months after the first traumatic episode for the right shoulder and 5.5 months after the incident for the left shoulder (Fig. 2A, B) as described below. Intraoperative findings showed a collapsed humeral head and granulation tissue both times. In addition, no synovitis was observed in the glenoid fossa (Fig. 2C).

The bone mineral density (BMD) of the patient's lumbar spine (L2-L4) obtained by dual-energy x-ray absorptiometry was 0.561 g/cm^2 (55% of the young adult mean [YAM]) and the T-score was −4.1. Based on these data and the above-described findings, we made the diagnosis of primary osteoporosis (Table 2). Severe osteoporosis was also diagnosed, and we prescribed monthly 75-mg risedronate (Actonel®, Eisai, Tokyo, Japan) with alfacalcidol (One Alpha®, Teijin, Tokyo, Japan) to prevent fragility fractures.

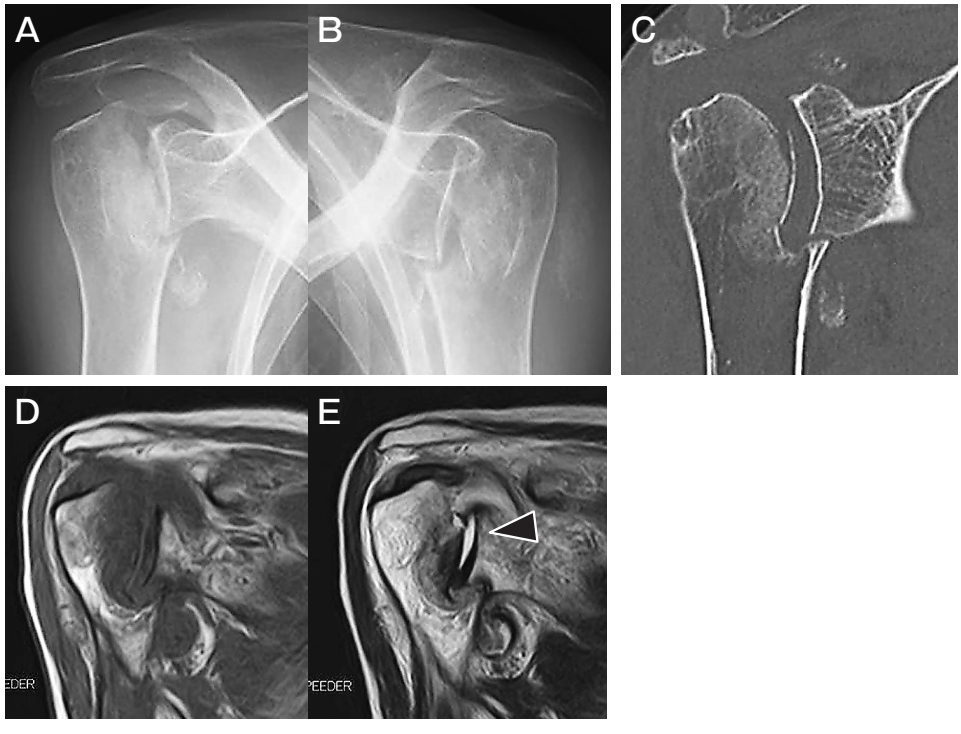

Fig. 1 **A**, **B**: Radiographic findings of the patient's bilateral shoulder joints at the initial visit. The bilateral humeral heads were severely damaged. Plain CT (**C**) and plain MRI (**D**, **E**) of the right shoulder joint. The CT images show an insufficiency subcapital humeral fracture, but the glenoid side was intact. A damaged humeral head is shown in the T1 low-intensity area (D), and the T2 high-intensity area (*black arrowhead*) demonstrated joint effusion in the glenoid joint (**E**).

As noted above, the patient underwent shoulder hemiarthroplasty (humeral head replacement). At the primary surgery, the bony fragments surrounding the granulation tissue were collected for a pathological examination. Hematoxylin and eosin staining from resected humeral head showed osteoid formation and osteoblasts around the newly formed bone (Fig. 3A, B). Empty lacunae were not observed in the osteoid (Fig. 3C, D). Osteoclasts around the newly formed bone and callus formation were also detected, suggesting that the fracture had healed successfully (Fig. 3E, F).

Table 2 Bone mineral density

BMD	Present case	Unit
Bone mineral density (L2–4)	0.561	g/cm^2
% Young Adult Mean	55	%
T-score	−4.1	

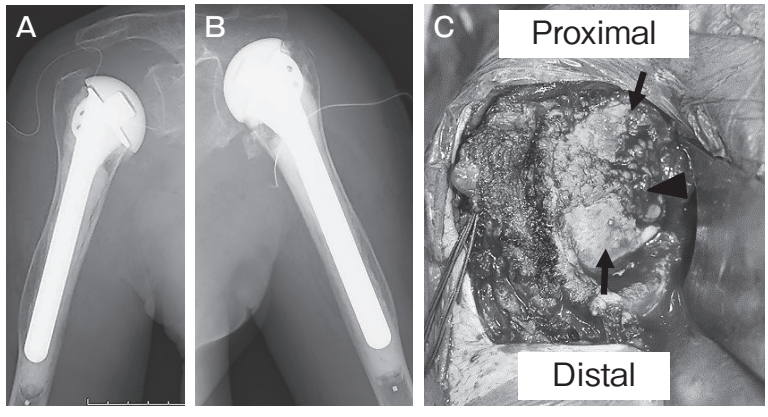

Fig. 2 Postoperative radiographs of the right (A) and left shoulder hemiarthroplasties (B). Each component was inserted with bone cement. C: Intraoperative findings from the right shoulder hemiarthroplasty. The humeral head was diffusely damaged (*black arrowhead*), and the remaining articular surface had degenerated (*black arrows*). No synovitis was detected.

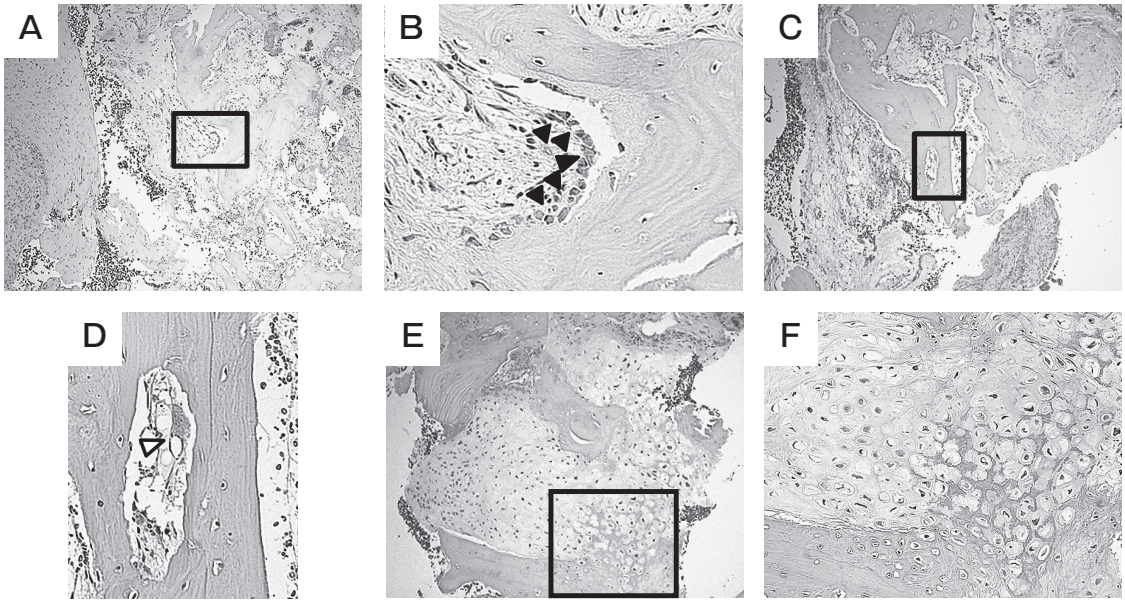

Fig. 3 Histological findings of bony fragments in the right humeral head. All samples were stained with hematoxylin and eosin. B: Enlarged view of the *black rectangle* in panel A. D, F: Enlarged views of the *black rectangles* in panels C and E, respectively. Osteoid tissue was detected (panels A, C). Palisading osteoblasts were detected around the osteoid (*black arrowheads*) (B), and osteoclasts (*white arrowhead*) were also detected (D). Callus formation was diffusely detected (E, F).

Two years after the hemiarthroplasties, the patient reported that she no longer suffered from bilateral shoulder pain. The passive ranges of motion of the bilateral shoulder joints were as follows: right, 120°/40°; left, 90°/40° during flexion/extension and right, 90°/20°; left, 90°/20° during abduction/adduction.

The patient was informed that data from the case could be submitted for publication, and she gave her consent for the publication.

Discussion

RDA in the shoulder joint is rare and is characterized by a rapid collapse of the humeral head, with no evidence of other non-septic articular arthropathies [2]. Generally, RDA in the shoulder has no specific symptoms or characteristic findings on examination, and thus a differential diagnosis is important [7]. In the present case, the differential diagnoses were infectious arthritis, crystal-induced arthritis such as uric acid and CPPD, rheumatoid arthritis, osteoarthritis, neuropathic arthropathy, and osteonecrosis of humeral head.

In our patient, no local heat or redness and no inflammatory response findings were noted. The bacterial culture results of the synovial fluid were negative. Infectious shoulder arthritis was ruled out. No crystals such as uric acid and CPPD were detected in the synovial fluid. Hence, crystal-induced arthritis was also ruled out. The laboratory analysis revealed anti-CCP antibody negativity, and no bone erosion was detected. Elderly-onset rheumatoid arthritis was also ruled out.

Both of the patient's humeral heads collapsed within approximately 41 days and finally flattened, leading to the diagnosis of RDA. She had bilateral RDA, which is quite rare; to our knowledge, this has been demonstrated in only a few prior cases [6].

In the reported cases, the duration of collapse of the humeral head was 5.6 months on average (range 2-11 months) [6]. Tokuya [8] reported the case of a 77-year-old female with RDA in whom the duration of the collapsed head was approx. 1 month, which is the shortest duration we identified in the literature. The duration of 41 days in our patient's case is the second shortest duration, to our knowledge.

The etiology of RDA remains unknown. The existence of hydroxyapatite crystals in the synovial fluid is a potential cause. Dieppe and Watt [9] indicated that a deposition of crystals is a secondary, opportunistic event in damaged cartilage. In another report [10], hydroxyapatite crystals were found to activate the release of collagenase and neutral protease. However, in our patient, no crystals such as uric acid and CPPD were detected in the synovial fluid. Crystal-induced arthritis was thus ruled out.

RDA has been speculated to be associated with osteoporosis. Our patient was diagnosed with severe osteoporosis, and her histological findings revealed the absence of empty lacunae in the bone matrix, ruling out the possibility of osteonecrosis. Alternatively, the histological findings of fracture repair, *i.e.*, the appearance of osteoclasts (Fig. 3D), callus, and osteoid formation (Fig. 3) were observed. Goshima *et al.* [7] reported the cases of two patients (77- and 74-year-old women) with RDA in the shoulder who also presented with osteoporosis and subchondral insufficiency fractures of the humeral head. In addition, Yoshikawa *et al.* [11] described 2 cases (74- (case 1) and 78-year-old (case 2) females) of ipsilateral RDA with the complication of osteoporosis in case 1. Their findings concur with our present findings; that is, our patient had severe osteoporosis with histological features of fracture healing, *i.e.*, callus formation and the appearance of osteoclasts.

The limitations of the present report are as follows: no radiographic findings were obtained at onset (day 0) because the patient's initial visit was 41 days after the initial traumatic episode, and the medical course of the patient was too short to follow up the natural course of the collapsed humeral heads.

In conclusion, we diagnosed a patient with bilateral RDA in the shoulders after ruling out osteoarthritis, idiopathic osteonecrosis, rheumatoid arthritis, infectious arthritis, crystal-induced arthritis, and neuropathic arthropathy. The patient's illness and pathological findings were key factors in the differential diagnosis. Histological findings from the biopsied humeral head indicated the process of fracture healing; thus, the patient's history indicated an insufficiency fracture due to severe osteoporosis.

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