



Osteochondral autograft transplantation for the treatment of steroid-induced osteonecrosis of the humeral head: a case report

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Osteonecrosis is defined as in situ death of a bone segment.³¹ The humeral head is the second most common site of symptomatic osteonecrosis after the femoral head.^{5,6} However, the optimal treatment of symptomatic humeral head osteonecrosis with radiographic collapse is still a matter of controversy. Traditionally, the treatment option is defined according to the stage of disease progression introduced by Cruess.^{4,18,28} The treatment options in the precollapse phase (stages I and II) include conservative treatment and core decompression.^{29,30,34} Prosthetic shoulder replacement options such as humeral head resurfacing,^{42,49} hemiarthroplasty, and shoulder arthroplasty^{25,32,38,40} are generally indicated for the treatment of symptomatic osteonecrosis with radiographic collapse (stages III through V). As for the treatment of young patients, joint-preserving procedures should be considered because young patients under 60 years of age who undergo shoulder arthroplasty are at an increased risk of revision surgery due to glenoid loosening and glenoid arthritis.^{1,7,15} To our knowledge, we report the first case of osteonecrosis

of the humeral head that was treated by osteochondral autograft transplantation (OAT).

Report of the case

A 53-year-old female patient visited our orthopedic clinic with a chief complaint of pain and limited active elevation of the right shoulder. She had a medical history of systemic lupus erythematosus (SLE) and lupus nephritis. She had been treated with steroid pulse therapy, followed by oral prednisolone for 17 years. In addition, she had been diagnosed with osteonecrosis of the medial femoral condyle in her right knee joint and underwent mosaicplasty 10 years before this visit. Three months before her initial visit, she experienced shoulder pain without any history of trauma or injury and visited another clinic. She had been diagnosed with osteoarthritis of the glenohumeral joint. Oral nonsteroidal anti-inflammatory drugs were prescribed for pain relief. However, the pain in her right shoulder persisted even during daily activities. Then, she was referred to our hospital.

On physical examination, active range of motion (ROM) of the right shoulder was 100° in forward flexion, 60° in abduction, 40° in external rotation at the side, and T12 in internal rotation behind the back. All directions of active ROM were restricted owing to shoulder pain. Preoperative functional scores of the American Shoulder and Elbow

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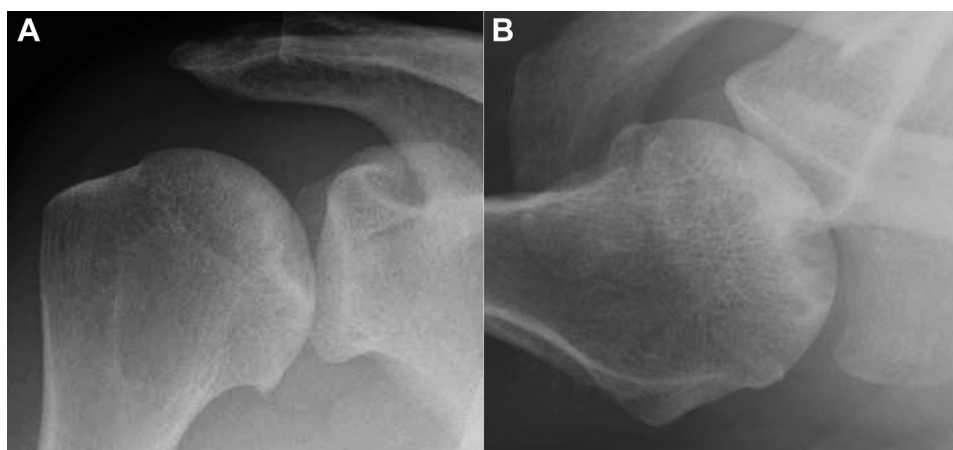


Figure 1 The initial finding of the plain radiograph. A subchondral lucency with a sclerotic rim and mild inferior osteophyte formation were noted in the humeral head. (A) Anterior-posterior view of the right shoulder. (B) Axillary view of the right shoulder.

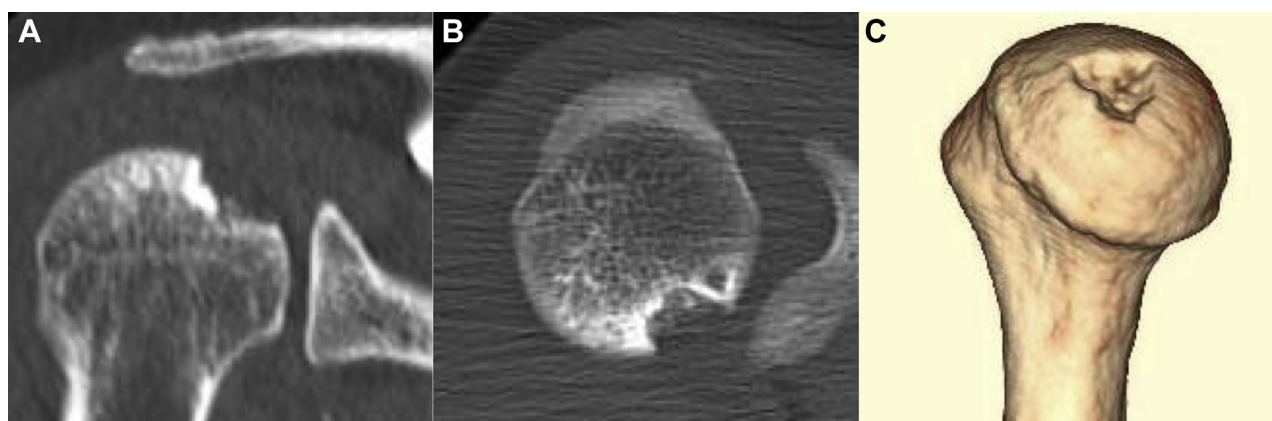


Figure 2 Preoperative computed tomography (CT) images of the right shoulder. A collapsed lesion was noted in the humeral head. (A) Oblique coronal image of the right shoulder. (B) Axial image of the right shoulder. (C) Reconstructed 3-dimensional CT of the right humerus.

Surgeons (a 100-point scoring system) and the Japanese Orthopaedic Association (a 100-point scoring system) were 53.3 and 69.0 points, respectively. The contralateral shoulder and other joints were asymptomatic. The plain radiograph revealed a subchondral lucency with a sclerotic rim and normal articular congruity of the glenoid. In addition, mild inferior osteophyte formation was noted in the humeral head (stage I osteoarthritis, according to Samilson and Prieto⁴⁴) (Fig. 1). The contralateral shoulder showed no abnormal finding. The computed tomography images revealed focal subchondral bone collapse with an articular incongruity of the humeral head (Fig. 2). The size of the affected area was 126 mm² by the measurement using picture archiving and communication systems.

Magnetic resonance imaging (MRI) revealed low signal intensity on both T1- and T2-weighted images (Fig. 3). Therefore, a clinical diagnosis was made of steroid-induced osteonecrosis of the humeral head. According to the Cruess

classification, this case was categorized as stage III (Table I).

To preserve the native joint, OAT was performed. A 10-cm skin incision was made on the anterior aspect of her right shoulder with the patient in the beach-chair position under general anesthesia followed by interscalene block. The shoulder joint was exposed through a standard deltopectoral approach. The proximal two-thirds of the subscapularis tendon and anterior capsule were cut from the lesser tuberosity 1 cm medial to its insertion. We exposed the humeral head by retracting the proximal two-thirds of the subscapularis and anterior capsule medially and rotating the humerus externally. The size of the subchondral bone collapse was 13 mm in length and 13 mm in width (Fig. 4, A). The transplantation procedure was performed with an osteochondral autograft transfer system (Arthrex, Naples, FL, USA). Three recipient sockets were created to provide an adequate press-fit graft fixation. The area along the lateral edge of the trochlea of her left knee joint was chosen

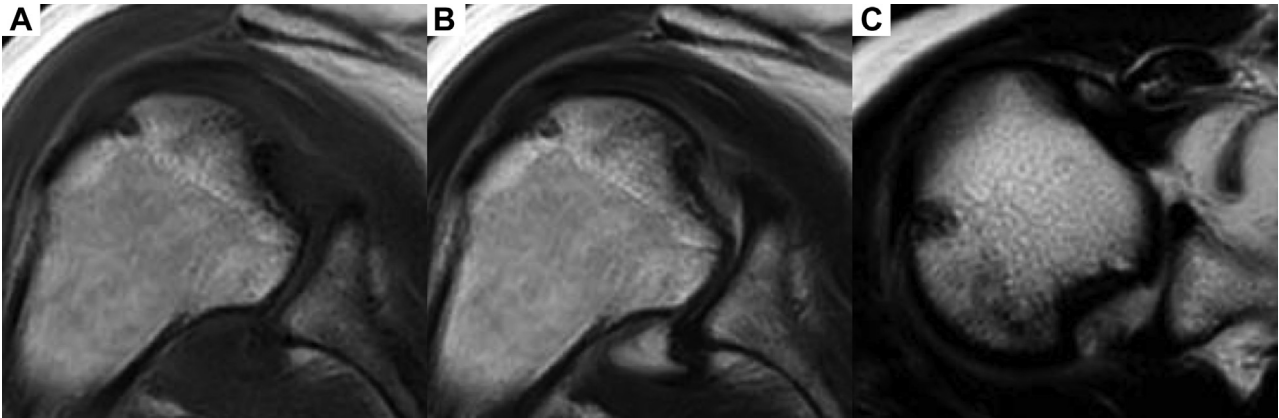


Figure 3 Preoperative magnetic resonance imaging of the right shoulder. (A) Oblique coronal T1-weighted image of the right shoulder. (B) Oblique coronal T2-weighted image of the right shoulder. (C) Axial T2-weighted image of the right shoulder.

Table I Classification system	
Stage	Description
I	No radiographic evidence of shoulder osteonecrosis, but MRI reveals localized bone edema
II	Signs of mottled sclerosis appear, but the curvature of the humeral head remains intact
III	A crescent sign, indicating that subchondral fracture appears. The humeral head may become less spherical as the initial stages of collapse are seen
IV	Advanced subchondral collapse and articular flattening with marked loss of humeral head sphericity
V	Humeral head collapse along with and the extension of degenerative changes in the glenoid cavity

MRI, magnetic resonance imaging.

as a donor site because she had undergone mosaicplasty for the treatment of osteonecrosis of the medial femoral condyle of her right knee joint 10 years before this surgery. We harvested three 6 × 15 mm osteochondral plugs. The harvested osteochondral autografts were then transferred into the recipient site using a press-fit technique (Fig. 4, B). Three bone tunnels were created just lateral to the subscapularis insertion on the lesser tuberosity. Three sutures of No. 2 Fiberwire (Arthrex) were placed through the bone tunnels, and these were sutured to the tenotomized tendon in a modified Mason-Allen configuration. In addition, 3 tendon-to-tendon simple sutures were placed to complete the repair. After the repair of the subscapularis, the surgical wound was closed.

One of the osteochondral plugs collected from the collapsed lesion was used for histopathologic diagnosis, and several empty lacunae were identified (Fig. 5). Thus, an experienced pathologist made a histologic diagnosis of osteonecrosis of the humeral head.

The right shoulder was immobilized using an abduction sling (Block Shoulder Abduction Sling; Nagano Prosthetics & Orthotics Co. Ltd., Osaka, Japan) for 4 weeks after surgery. After the immobilization period, passive- and active-assisted exercises were initiated. Eight weeks

after the surgery, the patient began to perform exercises to strengthen the rotator cuff and the scapula stabilizers. The patient was able to return to her desk work 2 months after surgery. She fully recovered the ability to perform daily activities, and computed tomography images showed graft union at 6 months after surgery (Fig. 6). At 2 years after surgery, the glenohumeral joint space was maintained, although the size of the inferior osteophyte was increased compared with that observed at the preoperative examination by the plain radiograph (Fig. 7). MRI showed complete graft integration and no osteonecrosis of the humeral head 2 years after surgery (Fig. 8). The patient was asymptomatic and active ROM in the right shoulder improved to 170° in forward flexion, 170° in abduction, 70° in external rotation at the side, and T10 in internal rotation behind the back (Fig. 9). The postoperative American Shoulder and Elbow Surgeons and Japanese Orthopaedic Association scores improved to 95.0 points and 100 points, respectively. The Lysholm score of the left knee, from which the osteochondral graft had been harvested, was 95 points 2 years after surgery.

The patient was informed that the data and photographs from the case would be submitted for publication and gave their consent.

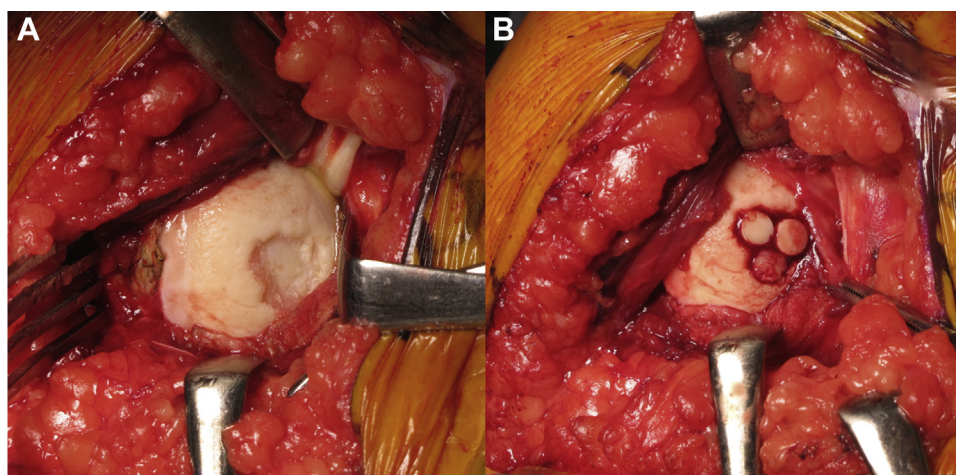


Figure 4 Intraoperative findings of the right shoulder. (A) A collapsed lesion noted in the right humeral head. (B) Three 6-mm-diameter osteochondral autografts transferred into the recipient site using a press-fit technique.

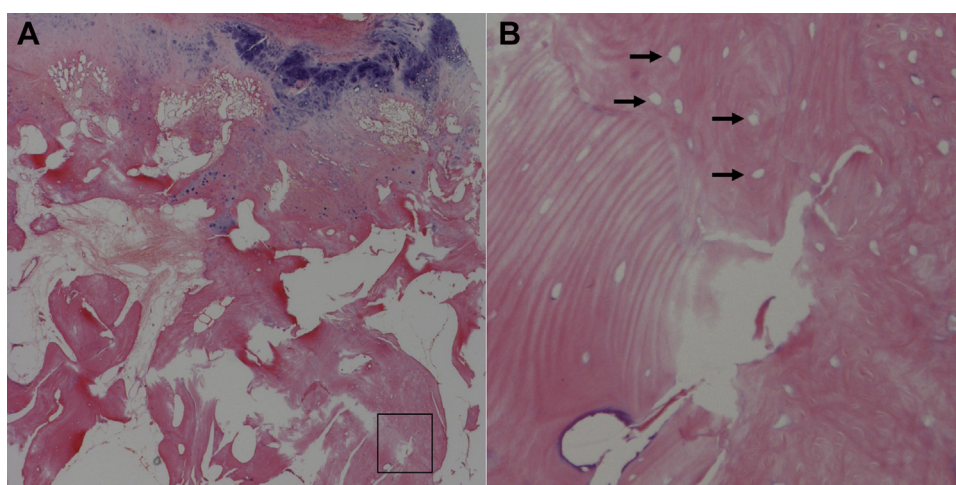


Figure 5 Histologic findings of the collapsed lesion in the right shoulder. Hematoxylin and eosin staining (A, magnification: $\times 20$; B, magnification: $\times 100$). (A) An image of tissue section of an osteochondral plug collected from the collapsed lesion. (B) High-magnification image of the \square in image (A). Several empty lacunae (\rightarrow) were noted.

Discussion

The treatment of advanced stages of osteonecrosis of the humeral head characterized by the collapse of the subchondral bone in young patients remains an unsolved burden in shoulder surgery. Here, we report the first case of osteonecrosis of the humeral head with a focal collapse (stage III) that was treated by OAT. Our patient showed excellent clinical and radiographic outcomes 2 years after surgery.

Osteonecrosis is a potentially painful and disabling bone disease that involves the death of bone tissue.³¹ The etiology of humeral head necrosis can be divided into post-traumatic or nontraumatic. The known etiological factors for nontraumatic osteonecrosis of the humeral head are corticosteroid administration, alcohol abuse, pancreatitis, hemoglobinopathies, diabetes, radiation therapy, and other

systemic diseases such as Cushing's syndrome, rheumatoid arthritis, and SLE.^{12,23,45} Our patient had no history of trauma and injury. However, she had a history of steroid pulse therapy followed by chronic administration of corticosteroids for the treatment of SLE. In addition, the histopathologic section of the collapsed lesion showed the findings of empty lacunae, indicating osteonecrosis. Thus, our patient was diagnosed with steroid-induced osteonecrosis of the humeral head.

Hernigou et al²⁶ investigated the natural history of osteonecrosis of the humeral head related to corticosteroid use by assessing plain radiographs and MR images. They reported that the interval between the initial corticosteroid treatment and the diagnosis of osteonecrosis of the humeral head averaged 15 months (range, 6-24 months). The average age of onset is reported to be between 37 and 46 years.^{26,29,30} The progressive patterns of osteonecrosis of

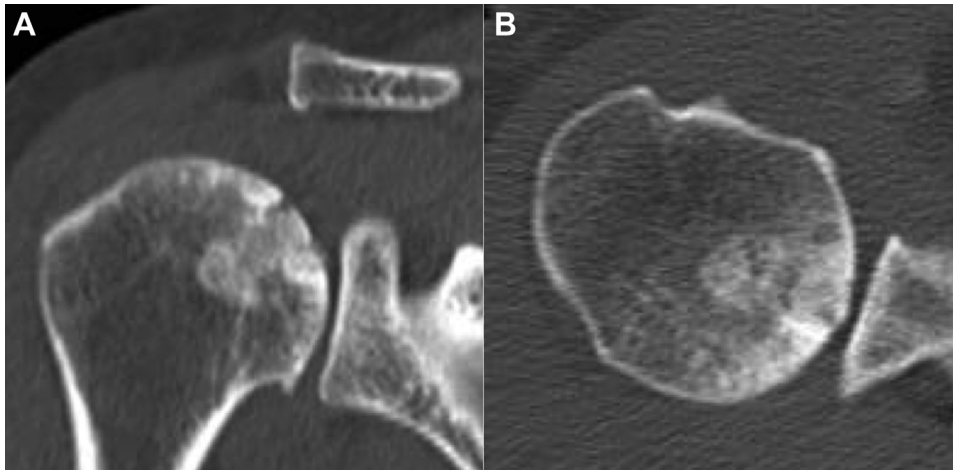


Figure 6 Computed tomography (CT) images 6 months after surgery. CT images showed the graft union. (A) Oblique coronal image of the right shoulder. (B) Axial image of the right shoulder.

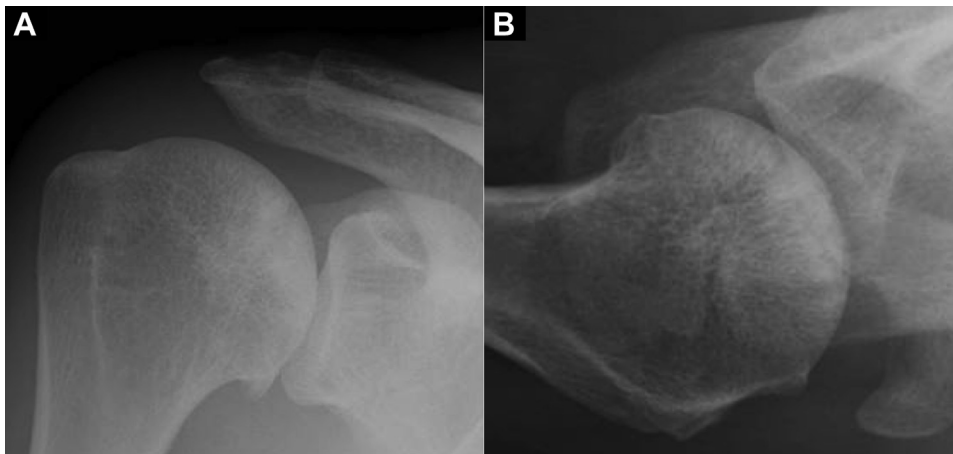


Figure 7 Finding of the plain radiograph 2 years after surgery. The glenohumeral joint space was maintained, although the size of the inferior osteophyte was increased compared with that observed at the preoperative examination. No recurrent osteonecrosis of the humeral head was noted. (A) Anterior-posterior view of the right shoulder. (B) Axillary view of the right shoulder.

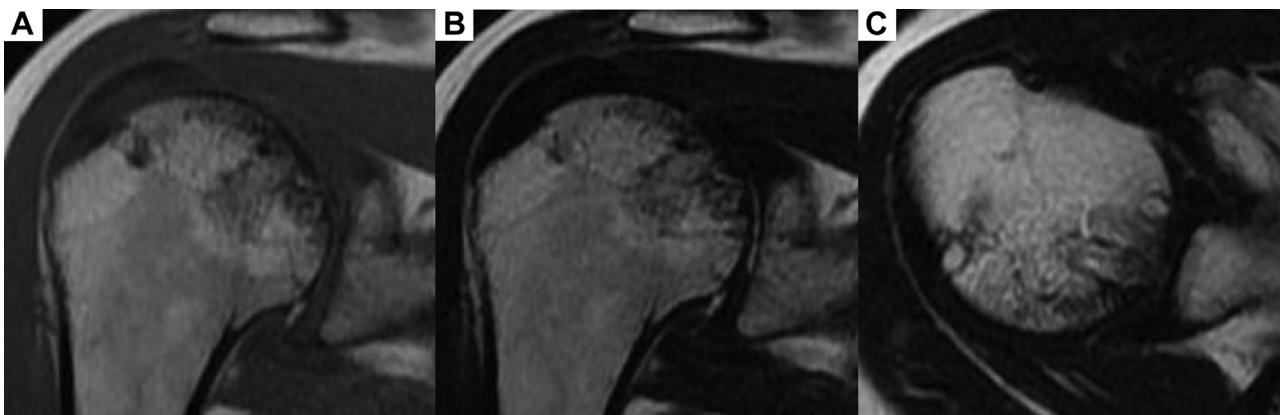


Figure 8 Magnetic resonance imaging 2 years after surgery. Complete graft integration and no recurrence of osteonecrosis were noted. (A) Oblique coronal T1-weighted image of the right shoulder. (B) Oblique coronal T2-weighted image of the right shoulder. (C) Axial T2-weighted image of the right shoulder.

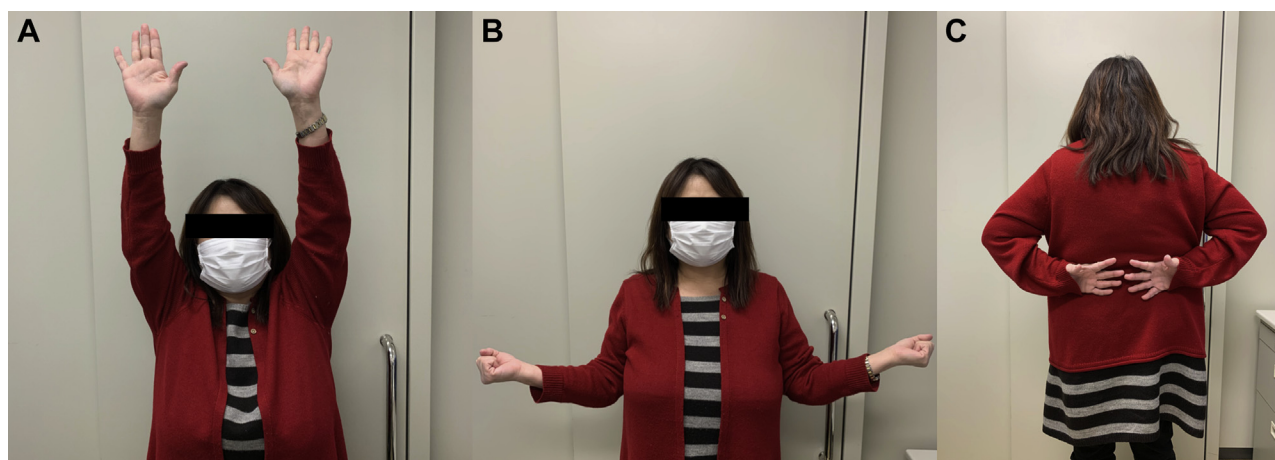


Figure 9 Active range of motion 2 years after surgery. (A) Shoulder elevation. (B) External rotation. (C) Internal rotation.

the humeral head have been described previously. The most widely used staging system for steroid-induced osteonecrosis of the humeral head is the Cruess classification, which is the modification of Ficat's staging system for osteonecrosis of the femoral head.^{4,9} The humeral head collapse was always preceded by pain. The time between diagnosis and collapse averaged 10 years for symptomatic stage I shoulders and 3 years for symptomatic stage II shoulders. The prognosis for shoulders with more advanced stages is less favorable.²⁴ L'Insalata et al²⁹ reported that radiographic stages of III or greater and the presence of radiographic progression were sensitive predictors and each strongly associated with a poor outcome.

The initial treatment of osteonecrosis of the humeral head is conservative, including exercises and the avoidance of overhead shoulder movements and strenuous activities.⁵ Surgical treatments for osteonecrosis of the humeral head include core decompression,^{29,30,34} vascularized bone grafting,^{10,27} humeral head resurfacing,⁴⁹ and shoulder arthroplasty.^{32,40} As for the joint-preserving surgery, a small number of autogenous bone grafting studies have been reported, including the fibular strut bone graft¹⁴ and the vascularized scapular bone graft;²⁷ however, these procedures are complex. Core decompression is a widely used straightforward procedure achieving favorable clinical outcomes identified before collapse.^{29,30,34} Nevertheless, the best success with core decompression is when it is performed in the earlier stages (stages I and II) of this disease before humeral head collapse as with osteonecrosis of the femoral head.^{29,34} For later stages (stages III, IV, and V) after the head collapse, joint replacement such as hemiarthroplasty or total shoulder arthroplasty (TSA) has been widely used. Generally, shoulder arthroplasties are designed to last 10–20 years. However, young patients under 60 years of age have an increased risk of revision at early follow-up.⁷ In addition, younger patients who undergo shoulder

arthroplasty have declining rates of self-reported satisfaction because of higher functional demands and expectations.⁸ Therefore, joint-preserving procedures should be considered for the treatment of young patients even in the later stage of the disease.

The preservation of patients' native joint anatomy has a significant advantage over hemiarthroplasty and TSA. Humeral offset, retroversion, neck-shaft angle, head height, and radius of curvature are patient-specific. Restoring these parameters with an implant can pose a challenge to the surgeon.^{2,41} The changes in biomechanics affect the contact pressure and stress distribution across the glenohumeral joint.^{2,20} The resultant eccentric loading can lead to glenoid loosening after TSA and can increase the rate of glenoid arthritis after hemiarthroplasty. Therefore, it is important to preserve or reconstruct the normal anatomy and biomechanics.

Osteochondral allograft transplantation and allogenic bone grafting are the alternative treatment options of osteochondral defects in the shoulder.^{16,17,33,43} Previous studies demonstrated favorable clinical outcomes after osteochondral allograft or allogenic bone grafting for the biologic resurfacing of osteochondral defects in the humeral head and/or glenoid.^{16,17,33,43} However, the use of allografts has the potential risks of disease transmission or immune graft rejection.^{3,35,47}

OAT is a well-established joint-preserving surgery for focal cartilage lesions or osteochondral lesions of the knee, talus, and humeral head.^{21,22,46} Previous studies reported that OAT had good long-term durability with successful outcomes in patients with articular cartilage defect or osteochondral defect in the knee or ankle joints.^{19,21,39,48} As for the treatment of steroid-induced osteonecrosis, a few reports have described the use of OAT for the necrosis of femoral condyle^{13,36} and femoral head.¹¹ To date, however, there have been no reports about the use of OAT for the osteonecrosis of the humeral head.

The main advantage of OAT is the reconstruction of the mechanically durable articular cartilage and subchondral bone and the removal of the collapsed lesion, including the necrotic bone tissue. OAT is typically used for the treatment of 100–400 mm² focal chondral and osteochondral lesions.²¹ In our case, the size of the collapsed lesion was 126 mm², which was within the indicated size used for the osteochondral lesions; we found the complete graft integration and no recurrent osteonecrosis of the humeral head at 2 years after surgery based on a plain radiograph and MRI examination. Although an increase in the size of humeral osteophyte formation was observed, the patient was asymptomatic and glenohumeral joint space was maintained 2 years after surgery. Furthermore, her right knee joint with osteonecrosis of the medial femoral condyle, which had been treated by OAT (mosaicplasty), demonstrated good clinical outcomes 10 years after surgery. Therefore, we believe that OAT has the potential to preserve the shoulder joint and prevent or delay the prosthetic shoulder replacement. Further observation is necessary to clarify the long-term result.

A disadvantage of OAT is the necessity to expose the knee joint. However, previous studies reported that knee complaints were relatively rare in the group of patients in which grafts were transferred outside the knee joint.^{22,37} Our patient showed excellent clinical score with no complication in the donor site, which was consistent with the previous reports.

Conclusion

We report a case of osteochondral autograft transplantation for steroid-induced osteonecrosis in a patient with SLE. Our patient showed excellent clinical and radiographic outcomes without any sign of recurrence of pain, a radiological progression of osteonecrosis in the shoulder, or donor site pain 2 years after surgery. Osteochondral autograft transplantation may be a viable treatment option for osteonecrosis of the humeral head in young patients with focal collapse.

Disclaimer

The authors, their immediate families, and any research foundations with which they are affiliated have not received any financial payments or other benefits from any commercial entity related to the subject of this article.

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