Synovial Chondromatosis of the Shoulder: Open Synovectomy and Insertion of Osteoarticular Allograft With Internal Fixation to Repair Intraoperative Glenohumeral Joint Instability

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Abstract

Synovial chondromatosis is a rare benign disorder characterized by chondroid metaplasia with multinodular proliferation of the synovial lining of a diarthroidal joint, bursa, or tendon sheath. These cartilaginous nodules may become embedded within the proliferating synovium and may extend into the surrounding soft tissues. They also can detach from the synovium, where they can calcify and may present as intra-articular loose bodies. Presence of these nodules leads to joint pain, dysfunction, and ultimately, destruction. Clinically, patients often present with a chronic monoarthropathy.

In this article, we report a case of extensive synovial chondromatosis of the right shoulder and surrounding soft tissues with extensive erosion of the humeral head, discuss combined anterior and posterior surgical excision of the cartilaginous fragments, and describe insertion of an osteoarticular allograft to repair the humeral head defect and secondary anterior glenohumeral joint instability.

Synovial chondromatosis (SC), first described by Reichel[1] in 1900, is a rare benign disorder characterized by chondroid metaplasia with multinodular proliferation of the synovial lining of a diarthroidal joint,[2-13] bursa,[14] or tendon sheath.[15] These cartilaginous nodules may become embedded within the proliferating synovium and may extend into the surrounding soft tissues. They can also detach from the synovium, where they can calcify and may present as intra-articular loose bodies. Presence of these nodules leads to joint pain, dysfunction, and ultimately, destruction.

Clinically, patients often present with a chronic monoarthropathy and endorse a history of years of joint pain and swelling with associated limitation in range of motion (ROM). Large joints are more commonly affected, the knee most often.[16] SC usually manifests in the third to fifth decades of life, but patients as young as 9 years have been affected.[7]

In this article, we report a case of extensive SC of the right shoulder and surrounding soft tissues with extensive erosion of the humeral head, discuss combined anterior and posterior surgical excision of the cartilaginous fragments, and describe insertion of an osteoarticular (OA) allograft to repair the humeral head defect and secondary anterior glenohumeral joint instability. The patient provided written informed consent for print and electronic publication of this case report.

Case Report

A 46-year-old right-hand–dominant machine operator presented with a 6-year history of progressive stiffness and limited ROM in the right shoulder. The man's occupational duties included repetitive lifting of 5- to

Figure 1. Clinical photograph of large mass of right shoulder.

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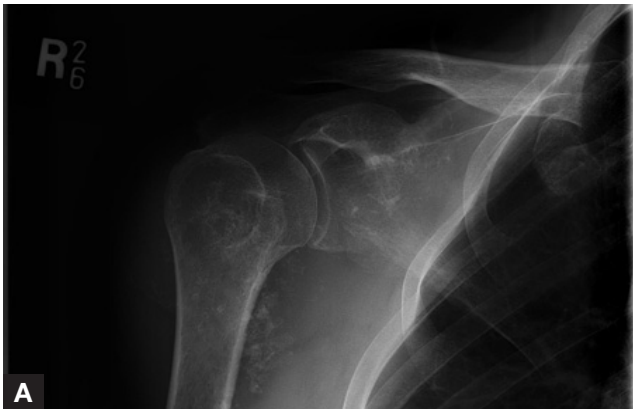
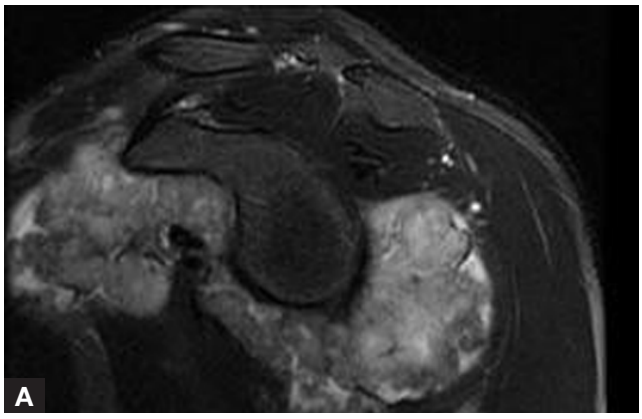


Figure 2. **Preoperative plain anteroposterior (A) and transscapular (B) radiographs show multiple calcific nodules in subcoracoid recess and extending distally along humerus.**

10-lb objects approximately 2000 times per day. Over the preceding 2 years, he had noticed increased swelling, a palpable mass, and the inability to raise the right arm over his head. He had experienced some discomfort during this period, but the decrease in shoulder ROM was his primary problem report. He denied any constitutional symptoms or other joint involvement. Past medical history was significant for hypertension, which was being managed with hydrochlorothiazide and amlodipine besylate.

Physical examination revealed no erythema or warmth surrounding the right shoulder. A firm mass was palpable over the anterior aspect of the shoulder near the coracoid and the posterolateral aspect of the right proximal humerus (Figure 1). Neurovascular examination of the upper extremity was normal. Active ROM of the shoulder was quite limited: 70° of forward flexion, 80° of abduction, 10° of external rotation, and internal rotation to the L3 vertebra. Before and after surgery, the patient was evaluated with the Toronto Extremity



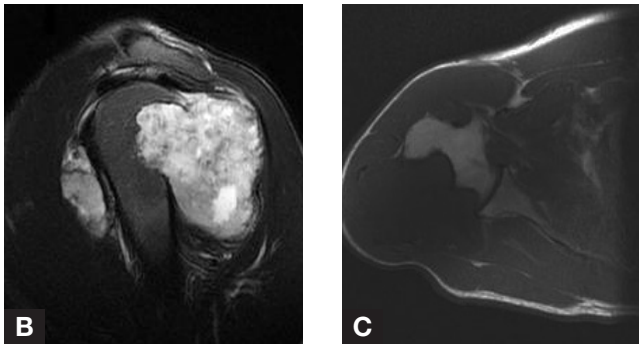


Figure 3. **(A,B) Oblique sagittal T2-weighted (fat saturation; repetition time/echo time, 3700/88.8 ms) magnetic resonance imaging (MRI) shows extensive multiple lobulated masses throughout joint capsule of glenohumeral articulation, around humerus and subcoracoid recess. (C) Axial T1-weighted (repetition time/echo time, 450/9 ms) MRI shows severe erosive changes in humeral head with reduction in size of articular surface.**

Salvage Score (TESS)[17] and the Musculoskeletal Tumor Society–87 (MSTS-87)[18] and MSTS-93[19] rating systems. On initial presentation, his scores were 82.1 (TESS), 21 (MSTS-87), and 57% (MSTS-93).

Plain radiographs showed a well-circumscribed area of osteopenia in the humeral head, with no evidence of periosteal reaction. There was extensive punctuate calcification in the soft tissue surrounding the humerus. The calcification extended distally along the biceps tendon and anterior to the coracoid process (Figure 2). Magnetic resonance imaging (MRI) was used to rule out a malignant lesion, to assess the site of the osteochondral bodies, and to assess intra-articular destructive changes. MRI showed marked glenohumeral joint distension by numerous intra-articular bodies. Large synovial masses were also present in the axillary, anterior capsular, and subcoracoid recesses. An anterior capsular mass (4.5×4.3×4.9 cm) was found (Figure 3). The posteriorly situated bodies had produced severe mechanical erosive changes in the humeral head; with almost half its diameter lost, the head had a scalloped appearance (Figure 3C).

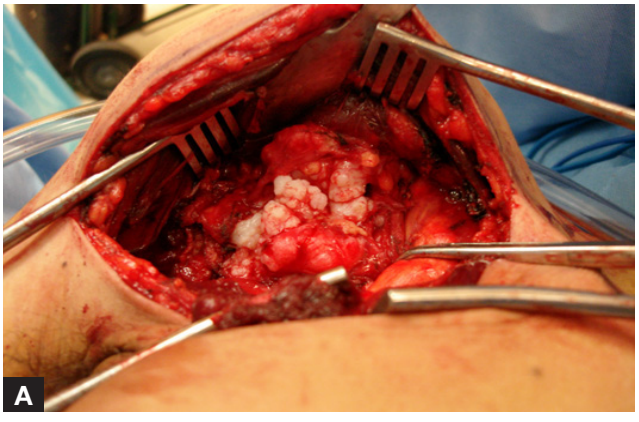
An open biopsy was performed. Pathology showed evidence of multiple focal islands of disorganized

Figure 4. Tissue biopsy shows focal islands of enlarged chondrocytes within disorganized metaplastic synovium.

hyaline cartilage metaplasia in the synovium. Enlarged chondroid lobules with focal dystrophic calcification and ossification were also visible. These findings were highly suggestive of SC (Figure 4).

Given the extent of shoulder involvement, and in line with patient's wishes, conservative management was initially undertaken. Eight months after initial presentation, the patient returned with increasing pain and restriction in ROM. Active ROM was reduced to 45° forward flexion, 30° abduction, 0° external rotation, and internal rotation to the buttocks. Follow-up MRI showed progression of the synovial lesions. Definitive surgical management was offered to the patient.

The patient was taken to the operating room with a plan to use both anterior and posterior approaches. A deltopectoral approach was used to resect the anterior and lateral masses. Arthrotomy of the glenohumeral joint allowed further resection of a large intra-articular mass. Extensive synovectomy was then performed. A posterior approach was used to remove the large posterior intra-articular soft-tissue mass, which extended approximately 120° around the humerus. This mass was removed piecemeal, with much attention given to protecting the axillary nerve. Once the extraosseous soft-tissue mass was removed, a curette was used to remove a significant portion of the tumor within the proximal humerus (Figure 5). This left a large humeral head defect that, when rotated, created significant anterior instability.



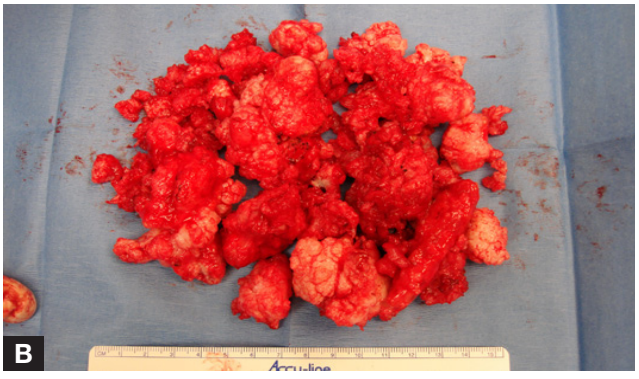


Figure 5. (A) Intraoperative picture of large calcific lesion surrounding humeral head. (B) Sample of gross specimen removed from shoulder.

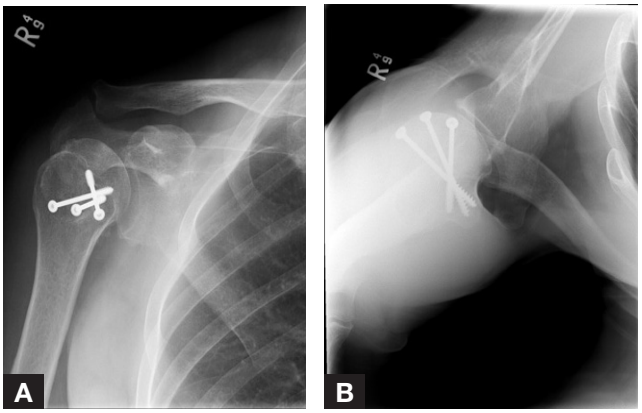


Figure 6. (A) Plain anteroposterior radiograph 3 weeks after extensive calcific nodules were removed. (B) Postoperative plain axillary radiograph shows allograft placement in humeral head with articular margins well aligned.

The OA allograft was cut to fit the defect, and a Kirschner wire was used to provisionally secure the articular margins in proper position. We then inserted 3 partially threaded cancellous screws through the allograft and into the native humeral head, and ensured they were countersunk in the articular cartilage to avoid prominence. With rotation, there was no evidence of screw impingement or anterior shoulder instability.

The patient was instructed to wear a sling and avoid engaging in active shoulder ROM for 3 weeks after surgery. Then he was started on passive ROM and pendulum exercises. Six months after surgery, he returned to work with modified duties. ROM had improved to 110° forward flexion, 90° abduction, 30° external rotation, and internal rotation to T10. Scores on TESS (82.7), MSTS-87 (26), and MSTS-93 (67%) were improved. There were no symptoms of shoulder instability. Plain radiographs showed no signs of nodular foci or loose bodies in the glenohumeral joint. The allograft was healing into the humeral head, and there was no evidence of hardware complications (Figure 6).

One year after surgery, ROM was slightly improved: 120° forward flexion, 100° abduction, 30° external rota-

tion, and internal rotation to T10. There was further improvement in scores on TESS (94.8), MSTS-87 (31), and MSTS-93 (93%). At most recent follow-up, 2.5 years after surgery, ROM and mild weakness were stable. Radiographs at each follow-up showed no evidence of recurrence, arthritic changes, or allograft resorption.

Discussion

Shoulder SC is a rare presentation that has been examined in case reports and small series.[3,5,6,9,11,15] Of the 191 SC cases meta-analyzed by Bloom and Pattinson[2] in 1951, only 10 involved the shoulder. Although the classical radiographic appearance of SC is that of multiple radiopaque calcified or osseous bodies within a joint or bursa, studies have shown these nodules are radiolucent in up to one-third of cases, making the diagnosis difficult.[16] As in our patient's case, the condition may be characterized by periarticular osteopenia.[13] When the patient's history is suggestive of SC but radiographs are normal, MRI can be used to identify noncalcified cartilaginous bodies.[4,16] At presentation, our patient's extensive extra-articular soft-tissue involvement was not visible on radiographs. It is very important to avoid confusing cartilaginous masses with joint effusions or tumors, particularly in patients with no evidence of calcification or ossification. We recommend using histopathologic analysis to definitively rule out a malignant lesion.

Optimal management for SC of the shoulder is still being debated. Some authors have had success with nonoperative techniques, such as nonsteroidal anti-inflammatory drug use, activity modification, and cryotherapy.[9] Arthroscopy is becoming common for intra-articular loose body removal because of its lower rate of postoperative comorbidity and shorter rehabilitation.[3,5,6,11] In our patient's case, arthroscopic resection was not undertaken, as there was extra-articular disease extending superficially to the subscapularis, and medial to the coracoid, as well as down the biceps tendon. We also believed that distortion of the normal anatomy would make visualization difficult. Whether to perform synovectomy at time of loose body removal is also unclear. Some authors support the idea that synovectomy reduces recurrence,[10,12,20] whereas others do not.[13]

After extensive excision of the tumor, it was apparent that tumor erosion had created a large posterolateral humeral head defect. This defect was anatomically similar to the Hill-Sachs defect commonly found in traumatic anterior shoulder dislocations. A humeral head defect may require operative management if it engages the anterior rim of the glenoid when the arm is brought into 90° of shoulder abduction and 90° of external rotation.[21] In our patient, this abnormality was observed during surgery.

Several surgical procedures have been used to manage large Hill-Sachs defects. Derotational osteotomy of the proximal humeral shaft is designed to direct the posterolateral defect more posterior and allow the undisturbed anterior surface to articulate with the glenoid. Optimally, this procedure prevents engagement of the defect on the anterior glenoid rim. Kronberg and Broström[22] found that patients who underwent this procedure regained normal ROM and function and had maintained shoulder stability for 5 years after surgery. Filling the defect with a transferred infraspinatus tendon also has been described. Purchase and colleagues[23] reported satisfactory results with use of this technique and no complications or loss of rotation. OA humeral head allografts create a mechanically stable joint but do not significantly alter surrounding anatomy. Sekiya and colleagues[24] found that this technique significantly improved joint stability in cadaveric models. Humeral prostheses are considered when defects involve more than 40% of the humeral head. Pritchett and Clark[25] described use of humeral hemiarthroplasty and total shoulder arthroplasty; functional outcomes were good, but postoperative rotational motion was poor.

We ordered the frozen OA humeral head allograft from the Bone Bank, which is on site at the Academic Hospital. Donor–recipient matching of an OA allograft is a 2-step process. First, the previously harvested donor graft undergoes serologic testing for transmissible bacterial and viral pathogens, and is then sterilized using gamma radiation. Second, shape-matching the donor graft to the recipient is based on height, weight, age, and sex. If these demographics are similar, the surgeon compares the donor graft measurements with the recipient's preoperative imaging.

Our choice of surgical reconstruction was based on clinical and operative factors. The patient was a young, active laborer without any degenerative disease in the glenohumeral joint. We believed that an OA allograft would maximize his ability to return to a high level of functioning. After tumor removal created a large posterolateral defect, the humeral head OA allograft optimally restored the anatomical and mechanically stable glenohumeral joint. Furthermore, the allograft would restore bone stock, which could be used should an arthroplasty procedure become necessary in the future.

Conclusion

In the case presented here, involvement of SC in the humeral head and the surrounding joint and soft tissues was extensive. Therefore, we performed anterior and posterior arthrotomies to facilitate adequate resection of the tumor and visualization of neurovascular structures. We believe that arthroscopy may be insufficient when synovial masses and osteochondral loose bodies are extensive. As with our patient, when loose bodies exist deep within the bicipital groove and severe erosive changes have damaged the humeral head, management by arthroscopy alone is not realistic.

Authors' Disclosure Statement

The authors report no actual or potential conflict of interest in relation to this article.

References

1. Reichel PF. Chondromatose der Kniegelenkkapsel. *Arch Klin Chir*. 1900;61:717-724.
2. Bloom R, Pattinson JN. Osteochondromatosis of the hip joint. *J Bone Joint Surg Br.* 1951;33(1):80-84.
3. Buess E, Friedrich B. Synovial chondromatosis of the glenohumeral joint: a rare condition. *Arch Orthop Trauma Surg*. 2001;121(1-2):109-111.
4. Butt SH, Muthukumar T, Casser-Pullicino VN, Maangham DC. Primary synovial osteochondromatosis presenting as constricting capsulitis. *Skeletal Radiol.* 2005;34(11):707-713.
5. Chillemi C, Marinelli M, De Cupis V. Primary synovial chondromatosis of the shoulder: clinical, arthroscopic, and histopathological aspects. *Knee Surg Sports Traumatol Arthrosc*. 2005;13(6):483-488.
6. Fowble VA, Levy HJ. Arthroscopic treatment for synovial chondromatosis of the shoulder. *Arthroscopy*. 2003;19(1):E2.
7. Harish S, Saifuddin A, Cannon SR, Flanagan AM. Synovial chondromatosis of the foot presenting with Lisfranc dislocation. *Skeletal Radiol*. 2005;34(11):736-739.
8. Kim SH, Hong SJ, Park JS, et al. Idiopathic synovial chondromatosis of the hip: radiographic and MR appearances in 15 patients. *Korean J Radiol*. 2002;3(4):254-259.
9. McFarland EG, Neira CA. Synovial chondromatosis of the shoulder associated with osteoarthritis: conservative treatment in two cases and review of the literature. *Am J Orthop*. 2000;29(10):785-787.
10. Ogilvie-Harris DJ, Saleh K. Generalized synovial chondromatosis of the knee: a comparison of removal of the loose bodies alone with arthroscopic synovectomy. *Arthroscopy*. 1994;10(2):166-170.
11. Ranalletta M, Bongiovanni S, Calvo J, Gallucci G, Maignon G. Arthroscopic treatment of synovial chondromatosis of the shoulder: report of three patients. *J Shoulder Elbow Surg*. 2009;18(3):e4-e8.
12. Schoeniger R, Naudie DD, Siebenrock KA, Trousdale RT, Ganz R. Modified complete synovectomy prevents recurrence in synovial chondromatosis of the hip. *Clin Orthop*. 2006;(451):195-200.
13. Shpitzer T, Ganel A, Engelberg S. Surgery for synovial chondromatosis. 26 cases followed up for 6 years. *Acta Orthop Scand*. 1990;61(6):567-569.
14. Horii M, Tamai M, Kido K, Kusuzaki K, Kubo T, Hirasawa Y. Two cases of synovial chondromatosis of the subacromial bursa. *J Shoulder Elbow Surg*. 2001;10(2):186-189.
15. David T, Drez DJ. Synovial chondromatosis of the shoulder and biceps tendon. *Orthopedics*. 2000;23(6):611-613.
16. Crotty JM, Monu JU, Pope TL Jr. Synovial osteochondromatosis. *Radiol Clin North Am*. 1996;34(2):327-342.
17. Davis AM, Bell RS, Badley EM, Yoshida K, Williams JI. Evaluating functional outcome in patients with lower extremity sarcoma. *Clin Orthop*. 1999;(358):90-100.
18. Enneking WF. Modification of the system for functional evaluation of the surgical management of musculoskeletal tumors. In: Enneking WF, ed. *Limb Salvage Musculoskeletal Oncology*. New York, NY: Churchill Livingstone; 1987:626-639.
19. Enneking WF, Dunham W, Gebhardt MC, Malawar M, Pritchard DJ. A system for the functional evaluation of reconstructive procedures after surgical treatment of tumors of the musculoskeletal system. *Clin Orthop*. 1993;(286):241-246.
20. Christensen JH, Poulsen JO. Synovial chondromatosis. *Acta Orthop Scand*. 1975;46(6):919-925.
21. Lynch JR, Clinton JM, Dewing CB, Warme WJ, Matsen FA III. Treatment of osseous defects associated with anterior shoulder instability. *J Shoulder Elbow Surg*. 2009;18(2):317-328.
22. Kronberg M, Broström LA. Rotation osteotomy of the proximal humerus to stabilise the shoulder. Five years' experience. *J Bone Joint Surg Br*. 1995;77(6):924-927.
23. Purchase RJ, Wolf EM, Hobgood ER, Pollock ME, Smalley CC. Hill-Sachs "remplissage": an arthroscopic solution for the engaging Hill-Sachs lesion. *Arthroscopy*. 2008;24(6):723-726.
24. Sekiya JK, Wickwire AC, Stehle JH, Debski RE. Hill-Sachs defects and repair using osteoarticular allograft transplantation: biomechanical analysis using a joint compression model. *Am J Sports Med*. 2009;37(12):2459-2466.
25. Pritchett JW, Clark JM. Prosthetic replacement for chronic unreduced dislocations of the shoulder. *Clin Orthop*. 1987;(216):89-93.

This paper will be judged for the Resident Writer's Award.