Hemiarthroplasty in a Patient With Pigmented Villonodular Synovitis of the Shoulder

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abstract

We report the 2-year follow-up results of a hemiarthroplasty in a 29-year-old patient with pigmented villonodular synovitis (PVNS) with extra-articular extension and severe bony destruction. The patient presented with diffuse pain and swelling of the right shoulder for several years. Radiographs showed severe osteolytic lesions in the humeral head and neck, greater tuberosity, and a small osteolytic lesion in the glenoid neck area. Further examination with magnetic resonance imaging of the shoulder joint showed a large, lobulated soft tissue mass extending from the subdeltoid area to the chest wall invading the adjacent bony structures. The operative findings included a large, multilobulated, yellowish-brown mass extending from the subdeltoid area to the axillary pouch, subacromial space, and thoracic wall. The mass invaded the anatomical neck area of the humeral head and caused a fracture of the humeral head. We observed a complete rupture of the long head of the biceps tendon, supraspinatus tendon, and infraspinatus tendon, but the subscapularis tendon was partially ruptured. We performed the complete excision and cemented hemiarthroplasty. At 2-year follow-up, no signs of local recurrence were present. The patient had no significant pain and achieved a good functional result.

To the best of our knowledge, 1 report with shoulder hemiarthroplasty in PVNS of the shoulder has been published in the English literature. For PVNS of the shoulder with a large extra-articular extension and bony destruction, shoulder arthroplasty with total synovectomy produces the lowest recurrence rates because of better access and excision to the posterior location of the PVNS lesion.

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Pigmented villonodular synovitis (PVNS) rarely affects the shoulder joint. In a study by Schwartz et al., 2 of 99 cases involved the shoulder. Shoulder involvement in PVNS has been reported in approximately 30 cases in the English-language literature. Among these, most cases were treated by arthroscopic or open synovectomy. However, when severe bone and joint destruction is present, arthroplasty with total synovectomy is indicated. Few reports have been published on shoulder arthroplasty as a treatment option for PVNS. This article presents the 2-year follow-up results of hemiarthroplasty in a patient with PVNS with extra-articular extension and severe bony destruction.

**Case Report**

A 29-year-old man presented with pain and diffuse swelling of the right shoulder of several years’ duration. He had a history of minor trauma to the right shoulder approximately 3 weeks prior. Physical examination revealed marked limitation of shoulder range of motion (ROM): 30° of forward flexion, 45° of abduction, 10° of external rotation, and internal rotation to the L5 level. A definite mass was not palpable, but diffuse swelling was seen. Laboratory findings showed normal complete blood count and a mildly elevated erythrocyte sedimentation rate. Simple radiographs showed severe osteolytic lesions in the humeral head and neck, greater tuberosity, and a small osteolytic lesion in the glenoid neck area (Figure 1). Further examination with magnetic resonance imaging (MRI) of the shoulder joint showed a large, lobulated soft tissue mass probably originating from the synovial joint causing destruction of the humeral head. However, no evidence of invasion existed in the brachial artery or nerve. Technetium-99 bone scan showed increased uptake localized in the right shoulder. The differential diagnoses included synovial sarcoma and synovial osteochondromatosis, an aggressive form of PVNS.

The patient underwent open biopsy through a mini-deltoid approach. Histological examination confirmed the diagnosis of PVNS. After 6 weeks, hemiarthroplasty with total excision was performed through the deltopectoral approach. The operative findings included a large, multilobulated, yellowish-brown mass extending from the subdeltoid area to the axillary pouch, subacromial space, and thoracic wall (Figure 3). The mass invaded the anatomical neck area of the humeral head and caused a fracture of the humeral head. The lesser tuberosity, greater tuberosity, and glenoid neck area were also invaded by the mass. The long head of the biceps tendon, supraspinatus tendon, and infraspinatus tendon were completely ruptured by the mass. A partial tear of the subscapularis tendon was observed.

Complete excision of the mass, including removal of the humeral head and synovium, was performed. The subscapularis tendon was repaired but the supraspinatus and infraspinatus tendons were irreparable. The size of glenoid bone defect was small; therefore, cementation was performed at the site of bone defect in the glenoid neck area. Cemented hemiarthroplasty was performed (Figure 4). The permanent histologic findings were compatible with PVNS.

At 2-year follow-up, mild superior migration of the implant due to cuff insufficiency was seen on simple radiography. No signs of local recurrence were present. Active ROM was 70° of forward flexion, 45° of abduction, 30° of external rotation, and internal rotation to the L5 level. The patient was able to do 8 of the 10 activities of daily living normally.
or with slight difficulty. Preoperatively, the visual analog scale (VAS) pain score was 8 points. After 2 years, the postoperative VAS pain score was 1 point.

**Discussion**

Pigmented villonodular synovitis is a nonneoplastic tissue proliferative disorder that originates from the synovium of joints, bursae, or tendon sheaths. Pigmented villonodular synovitis is a rare disorder, with an annual incidence of 1.8 cases per 1 million individuals.4 The knee joint accounts for 80% of cases, followed by the hip, ankle, shoulder, and elbow joints.2,3,6 However, recent study reports have indicated that the knee is the most frequent site, followed by the foot and ankle.7 The hip, hand, and shoulder seem to be the least common sites.7 Although PVNS can occur in all age groups, peak incidence ranges between 30 and 50 years of age and affects men and women equally.2,4,6 However, a recent study also reported that PVNS appears to be significantly more frequent in women than in men.7 Nevertheless, involvement of the shoulder joint is rare.

Pigmented villonodular synovitis with the extra-articular component is a significant diagnostic challenge to clinicians.8 Bony involvement can be mistaken for bone malignancy or soft tissue sarcoma. The differential diagnosis includes soft tissue sarcoma (particularly synovial sarcoma), hemangioma, synovial osteochondromatosis, and rheumatoid arthritis. Synovial sarcoma has no relationship with the synovial tissue. An intra-articular location of synovial sarcoma is rare. Synovial hemangioma is often visualized with poorly defined lobulated margins, less mass effect, and on T2-weighted images, which reflects blood pooling. Synovial osteochondromatosis often has calcified or ossified lesions. The intrasynovial cartilaginous masses have lobulated borders and homogeneous intermediate signal on T2-weighted images.9 Rheumatoid arthritis typically has a more uniform, thinner synovial proliferation, and less hemosiderin deposition. Regardless of these differences, synovial biopsy is essential to confirm the diagnosis.

Most authors recommend local excision in cases of the localized form of PVNS and total synovectomy in cases of the diffuse form of PVNS. For a severely affected joint with bony destruction, synovectomy with arthroplasty is indicated. Total hip arthroplasty is commonly performed compared with that for the other joints. Hip surgery appears to have a high rate of failure in terms of restoration of normal function and often requires total joint replacement.7 Recurrence and secondary osteoarthritis are the most common complications of PVNS of the hip joint.8,9 For total synovectomy and restoration of motion, THA with synovectomy is the treatment of choice in the diffuse form of PVNS. For the shoulder joint, arthroplasty with total synovectomy is rarely performed. To the best of our knowledge, a total of 6 cases published in 5 reports were treated by shoulder arthroplasty with total synovectomy.5 A total of 2 cases published in 1 report were treated by hemiarthroplasty with total synovectomy.5

Recurrence of PVNS is likely due to incomplete synovectomy. Johansson et al2 reported a recurrence rate of 33%, whereas Byers et al9 found a 50% recurrence after total synovectomy in diffuse PVNS. In PVNS of the shoulder, the recurrence rate is low because of its low incidence.4,12,13 The recurrence rate does not increase with bone involvement.13 Complete synovectomy with prosthetic joint replacement produces the lowest recurrence rates14 due to better access to the posterior location of the PVNS lesion. Total shoulder arthroplasty and hemiarthroplasty, in which removal of the humeral head is performed, provide better access to the joint than the more conservative approaches.4,5

Controversy exists regarding whether hemiarthroplasty or total shoulder arthroplasty is superior for the management of glenohumeral osteoarthritis. Despite this controversy, a general consensus exists that the outcome of total shoulder arthroplasty is much better than that of hemiarthroplasty.15-17 If rotator cuff deficiency exists, then total shoulder arthroplasty is contraindicated due to the “rocking horse” phenomenon. In our case, irreparable rotator cuff deficiency and a relatively intact subscapularis tendon were seen intraoperatively. Two prosthesis options were available: hemiarthroplasty and reverse total shoulder arthroplasty. However, because of the young age of the patient, we decided to perform hemiarthroplasty.

For PVNS of the shoulder with a large extra-articular extension and bony destruction, shoulder arthroplasty with total synovectomy is the treatment of choice. Because of better access to the posterior location of the lesion, the recurrence rate after arthroplasty with synovectomy is low. The choice of the prosthesis depends on the clinical situation.

**References**

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