

Hemiarthroplasty for pigmented villonodular synovitis of the shoulder: a report of two cases

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ABSTRACT

Pigmented villonodular synovitis is a reactive condition characterised by exuberant proliferation of synovial villi and nodules. It may be localised or diffuse and can cause severe articular damage. This report is of 2 patients with pigmented villonodular synovitis of the shoulder causing extensive arthritic changes. Both patients underwent shoulder hemiarthroplasty and total synovectomy and achieved satisfactory painless range of motion, with no signs of local recurrence or loosening of the prosthesis after 4 to 5 years of follow-up.

Key words: *arthroplasty, replacement; shoulder; synovitis, pigmented villonodular*

INTRODUCTION

Pigmented villonodular synovitis (PVNS) is a reactive condition, not a true neoplasm, resulting in proliferative formation of villi and nodules in the synovium. Despite being a benign process, PVNS

can cause severe articular damage.^{1,2} Less than 2% of PVNS affects the shoulder.³ Several cases of PVNS have been discovered incidentally during surgery for joint instability or rotator cuff repair.³⁻⁷ The condition may present as osteoarthritis with cystic erosions, usually on the humeral side. When the joint is already severely destroyed, synovectomy may have to be combined with arthroplasty. This report is of 2 patients with diffuse PVNS of the shoulder causing extensive arthritic changes that were treated with shoulder hemiarthroplasty and total synovectomy.

CASE REPORTS

Patient 1

In May 2004, a 63-year-old woman presented with a 6-month history of deep, constant pain in her left, non-dominant, shoulder. She had no history of trauma. Clinical examination revealed diffuse swelling of the shoulder, and limited and painful range of motion. Blood tests (including erythrocyte sedimentation rate) were normal. Plain radiographs showed severe erosion of the humeral head, extended calcification of the deltoid, and swelling of the soft tissues (Fig.

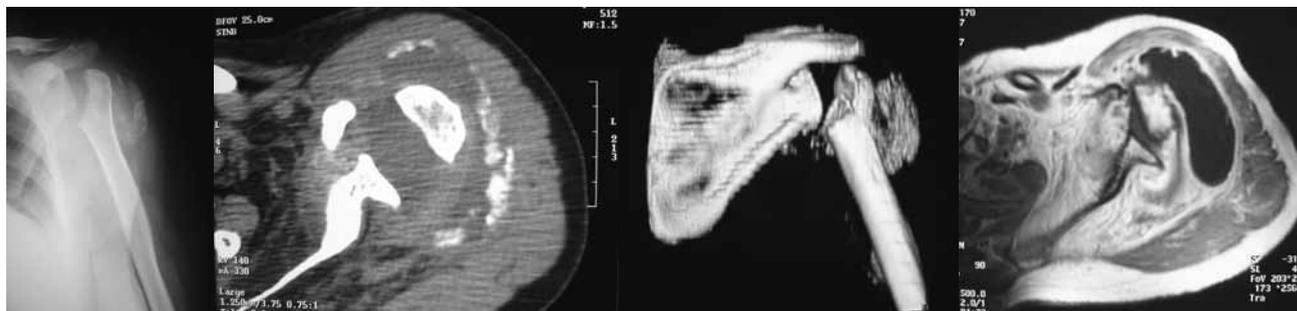


Figure 1 Radiological findings of the left shoulder showing severe erosion of the humeral head, extended calcification of the deltoid, and swelling of the soft tissues.

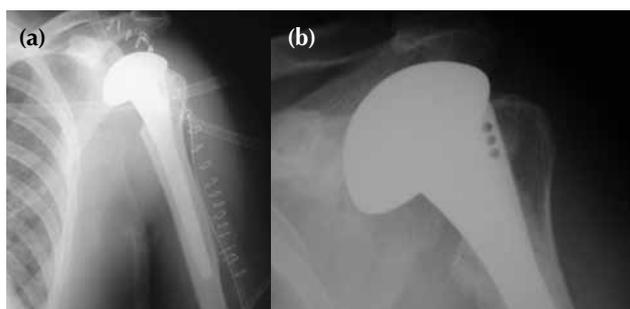


Figure 2 Patient 1: radiographs of the left shoulder taken (a) immediately after cemented hemiarthroplasty and synovectomy and (b) at the 5-year follow-up.

1). Computed tomography demonstrated a tumour-like lesion expanding in the adjacent humeral head, with calcifications mainly in the deltoid muscle. The left ribcage was intact with no erosion or infiltration (Fig. 1). Technetium-99 bone scan showed increased uptake localised in the left shoulder only. Magnetic resonance imaging revealed a large soft-tissue mass expanding from the subdeltoid and subacromial spaces to the glenohumeral joint, exerting pressure on the bony structures (Fig. 1). The mass had also expanded into the axilla, with no signs of brachial plexus involvement. The differential diagnoses included Charcot joint, synovial sarcoma, and an aggressive form of PVNS.

The patient underwent open biopsy through a mini deltopectoral approach. Histology revealed hyperplastic synovial villi containing multinuclear giant cells and foam cells, typical of villonodular synovitis.

Six weeks later, the patient underwent complete excision of the pathologic intra- and extra-articular synovium through a standard deltopectoral approach. Osteotomy of the humeral head enabled removal of the posterior and inferior aspects of the synovium.

As the rotator cuff was intact and the glenoid was completely unaffected, the decision was made to proceed to cemented hemiarthroplasty without glenoid replacement (Fig. 2). Shoulder mobilisation was allowed on day 1 as long as the pain could be tolerated. The postoperative course was uneventful and the patient was discharged 4 days later.

At the 5-year follow-up, there were no signs of local recurrence or loosening of the prosthesis (Fig. 2). The patient had satisfactory painless range of motion (forward flexion, 0°–150°; abduction, 0°–140°; external rotation, 0°–30°; internal rotation, thumb to L2). The Constant score had improved from 35 to 84.

Patient 2

In April 2005, a 60-year-old man presented with a one-year history of pain and limited range of motion in his right, dominant, shoulder. The condition rapidly deteriorated 6 weeks prior to presentation. He had no history of trauma or general illness. Radiographs revealed an eroded humeral head (Fig. 3). Blood tests (including erythrocyte sedimentation rate) were normal.

Stemmed arthroplasty of the right shoulder was planned. During the procedure, extensive destruction of the humeral head was noted, but the rotator cuff was intact and the glenoid was in good condition. Cemented hemiarthroplasty was therefore undertaken (Fig. 3). The synovium was noted to be pathologic with marked proliferation and a villonodular appearance. Meticulous synovial excision was performed. Histology confirmed the diagnosis of PVNS. No complications developed, and the patient was discharged 4 days later.

At the 4-year follow-up, there were no signs of local recurrence or prosthesis loosening (Fig. 3). The patient had pain-free, almost full range of motion (forward flexion, 0°–165°; abduction, 0°–160°; external rotation, 0°–45°; internal rotation, thumb to T10). The

Constant score had improved from 42 to 92.

DISCUSSION

The incidence of PVNS is 1.8 per one million people. The condition affects men and women equally, and is most common in young adults.^{2,4} The mean age at the time of diagnosis is 60.7 (range, 18–84) years for women and 43.5 (range, 5–64) years for men.⁴ The knee joint is the most frequently affected (80%), followed by the hip, ankle, shoulder, and elbow.^{8–10} Other anatomic locations and polyarticular involvement are extremely rare.^{7,11} PVNS of the shoulder typically occurs in older patients as a diffuse form; a combination of intra- and extra-articular lesions is not uncommon.^{4,11,12}

The aetiology of PVNS is unknown and may be attributed to a repetitive inflammatory process.¹³ Fibrohistiocytic proliferation is polyclonal in origin, and polyclonality is consistent with a reactive process.¹⁴ There is a significant association between PVNS and chronic recurring trauma,⁹ but no consensus has been reached regarding causation.⁴ Intra-articular haemorrhage is not a trigger factor.¹⁵ A lipid metabolism disorder and chromosomal deviations (such as trisomy) have also been postulated as causative factors.^{7,14}

Malignant transformation is extremely rare,¹⁶ and should be suspected when the tumour infiltrates both soft-tissue and bone. Malignancy cannot be ruled out on the basis of the frozen section. Nonetheless, a radical surgical procedure is not indicated as routine treatment.⁴ Only one patient with metastasis has been

reported, in whom an ankle lesion metastasised to the lung.¹⁷

The presence of polycystic changes in the humeral head is a possibility in diffuse PVNS of the shoulder.¹³ Increased articular pressure leads to small areas of osteoporosis where the bone cysts develop. These cysts are later invaded by the hypertrophic synovium of the joint through fractures in their walls.⁴ Cysts can also be created by the extension of villonodular tissue into the bone through the chondro-osseous area at the articular margin.¹⁸ Bone can also be invaded by hypertrophic synovium through the vascular foramina along with the epiphyseal vessels.¹⁹ Villonodular tissue then further expands within the bone cysts by pressure atrophy.

Patients usually present with a long history of moderate pain that is intermittent in character.⁴ PVNS usually progresses slowly and lacks systemic symptoms; diagnosis is usually not made until the late stages when the articular damage is extensive, as in the first patient. There is a patient in whom swelling and loss of motion occurred without any pain.²⁰ Joint effusion may be present, particularly in young patients with knee involvement, but this is uncommon and not relevant to other joints.⁸ Aspirated fluid is typically xanthochromatic or serosanguineous, but serosanguineous effusion is usually absent in the shoulder.¹¹

Initial radiographs can be negative, and calcification is not a usual feature of PVNS.⁷ Bone changes can be mistaken for a neoplastic bone or joint lesion such as malignant synovioma. Differential diagnoses include synovial osteochondromatosis, haemophilia, rheumatoid arthritis, gout, and



Figure 3 Patient 2: radiographs of the right shoulder taken (a) preoperatively showing extensive humeral head erosion, (b) immediately after cemented hemiarthroplasty and synovectomy, and (c) at the 4-year follow-up.

tubercular and infectious arthritis.³ Magnetic resonance imaging can reveal synovial proliferation, but the results may be inconclusive in patients with extensive soft-tissue proliferation, as in the first patient.

Total synovectomy is difficult to achieve, and up to 50% of cases recur, especially in diffuse PVNS.²⁰ Arthroscopic synovectomy is the treatment of choice, as it enables more detailed exploration of the subacromial bursa and the glenohumeral joint, and is as effective as open surgery while being less aggressive.³ Systematic adjuvant radiotherapy is advocated for lesions with no or moderate bony involvement.¹⁷

When massive arthritic changes are present, total synovectomy alone may be insufficient and should be combined with shoulder arthroplasty.²⁴ The 2-year results are good for 2 patients with PVNS treated with total shoulder arthroplasty.⁸ The treatment facilitates posterior synovial resection, thus reducing

recurrence,² as osteotomy of the humeral head facilitates access to difficult-to-visualise parts of the synovium.

Recurrence is likely to be due to incomplete synovial excision. Rates vary according to the joint affected and can be as high as 50% in the knee.² In PVNS of the shoulder, recurrence is rare, as is the occurrence^{2,3,17}; recurrence rates do not differ for localised or diffuse types, nor is the bone involved.¹⁷ A study reported no clinical or radiographic signs of recurrence 4 years following total shoulder arthroplasty combined with total synovectomy.² Nonetheless, the mean recurrence interval is 4.5 years.¹⁷

Results of hemiarthroplasty are superior to those of total shoulder arthroplasty in patients with osteoarthritis.²¹ When the glenoid is not affected, shoulder hemiarthroplasty combined with total synovectomy is the treatment of choice for PVNS of the shoulder, as synovectomy is not compromised.

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