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Osteochondromatosis of the subacromial bursa Arthroscopic treatment of a case and review of the literature

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ABSTRACT

We present the diagnostic approach and the arthroscopic treatment of a patient with primary synovial osteochondromatosis of the subacromial bursa without glenohumeral joint involvement. Biopsy of the subacromial bursa confirmed the diagnosis and arthroscopic synovectomy resulted in eradication of the disease.

Key words: *arthroscopy, osteochondromatosis, shoulder.*

INTRODUCTION

Synovial osteochondromatosis (SOC) is a rare benign disorder characterised by formation of multiple cartilaginous foci mainly in the synovium and less frequently in bursa or tendon sheaths which eventually break free and form loose bodies. SOC is usually monoarticular and presents between the 2nd and 7th decade of age, mainly in male patients with male/female ratio of 1.8:1[1]. The most frequent location is the knee followed by the hip, the elbow and the shoulder[2,3,4]. SOC has been reported in unusual regions such as the distal radioulnar joint[5], the ankle[6], the Lisfranc joint[7], the temporomandibular joint[8] and the popliteal bursa[9].

In the shoulder, SOC can be localised in the glenohumeral joint[10,11,12] or the subacromial bursa[13,14] where it can accompany a partial or full tear of the rotator cuff. There have also been described cases of SOC in the acromioclavicular joint[15,16] and the subcoracoid bursa[17]. The pathophysiology of SOC remains unknown, while genetic factors, trauma or chronic synovitis have been implicated.

We present the diagnostic approach and the arthroscopic treatment of a patient with SOC of the shoulder localised in the subacromial bursa.

CASE REPORT

The case involves a 72 year old retired military officer with occasional pain and dysfunction of both shoulders during the past two years. The patient was examined at the outpatient clinic due to pain increase at the left shoulder for the last semester accompanied with progressive restriction of the range of motion. The patient did not report nocturnal pain or pain at rest but complained about pain exaggeration on forward flexion and abduction over the shoulder level. The patient does not have any athletic activities and is not involved in demanding labour work. Physical examination did not reveal muscle atrophy or oedema of the shoulder. Forward flexion was 160°, abduction was 170°, external rotation was 75° and internal rotation allowed the hand to reach the level of T12, while there was no difference between passive and active range of motion. The strength of the deltoid and the infraspinatus was normal, while there was reduction in the strength of the supraspinatus.

The clinical tests for subacromial impingement and supraspinatus tendon tear were positive and tenderness at the acromioclavicular joint was also noticed. The Napoleon sign and the lift-off test for the subscapularis as well as the tests for the tendon of the long head of biceps were negative.

Plain radiographs of the shoulder revealed the presence osteochondromatosis in both shoulders (figure 1). Free bodies were found in the subacromial space superimposed on the humeral head. MRI confirmed the presence of osteochondromatosis as well as tendinopathy of the supraspinatus giving the hint of a possible partial tear (figures 2, 3, 4).

Surgery was performed under general anaesthesia and interscalene block of the brachial plexus with the patient in

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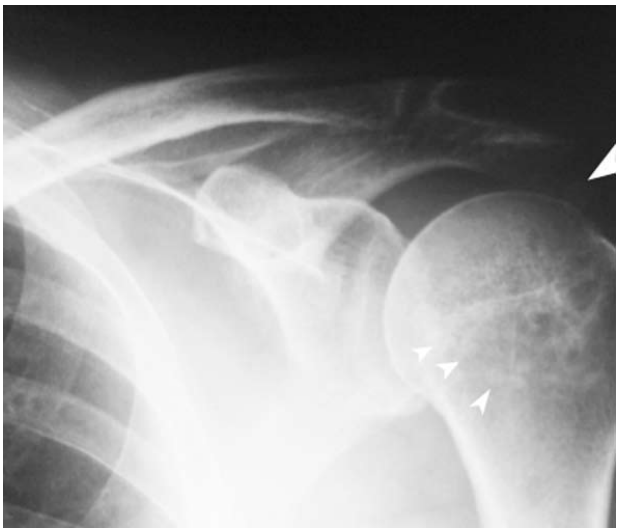

Figure 1. Anteroposterior radiograph of the left shoulder. Loose osteochondral bodies are shown in the subacromial bursa.

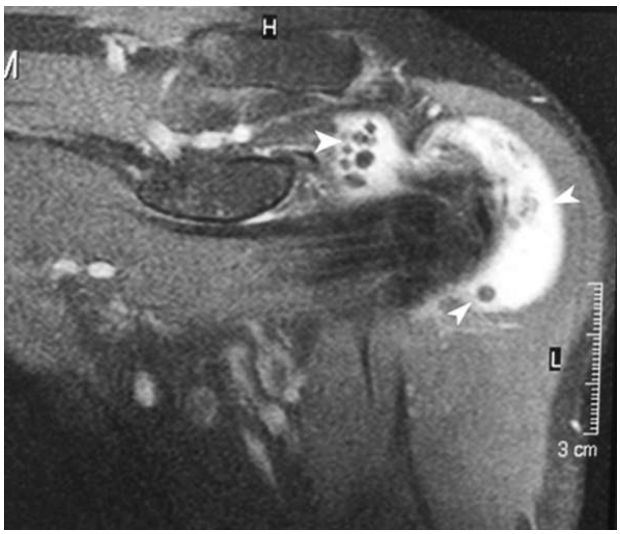

Figure 2. MRI of the left shoulder (T1 sequence, coronal view). Several loose bodies are present in the posterior subacromial space, above the infraspinatus, and at the subdeltoid bursa.

lateral decubitus position. A posterior portal was used for inspection of the glenohumeral joint and the subacromial space and anterior and lateral portals were created for the inspection of the subacromial space. Glenohumeral inspection was performed in accordance to the 15 inspection points as described by S. Snyder and did not reveal the presence of free bodies or hypertrophic synovitis. Upon entering the subacromial space, presence of hypertrophic synovitis was encountered along with multiple osteochondral bodies either free or attached to the bursa of the supraspinatus tendon (figures 5, 6). Extensive synovectomy, acromioplasty and removal of the loose bodies were performed. No complete tear of the supraspinatus was revealed either from the subacromial space or from the joint side. Biopsy of the synovium confirmed the diagnosis of SOC. One year after the operation, our patient has significant improvement of his symptoms and improvement in the range of motion.

DISCUSSION

Osteochondromatosis can be primary or secondary. In secondary osteochondromatosis the presence of free osteochondral bodies is due to osteoarthritis, trauma causing osteochondral fractures or osteochondritis dissecans[4] while in primary SOC the source of the osteochondral loose bodies is the benign synovial neoplasia. The two forms can be distinguished by the history, the accompanying findings and ultimately the histological examination[14]. The exact pathogenetic mechanism of SOC remains unknown. Primary SOC is attributed to metaplasia of the synovium, bursa or tendon sheaths, while multiple microtrauma and chronic synovitis are considered predisposing factors. Recent studies concerning chromosome 6 abnormalities and the effect of bone morphogenetic protein on the synovial mesenchymal cells[10] support the above theory[18,19].

The shoulder is a rare localization for SOC let alone in the subacromial space[12,14]. Milgram described three stages of the disease[10,20]. In Stage I (primary) there is synovial disease without formation of cartilaginous bodies, in Stage II (intermediate) there are cartilaginous bodies both loose and attached to the synovium or bursa, and in Stage III (late) there are solely loose bodies. Cartilaginous bodies undergo several grades of ossification and calcification by the synovial fluid, therefore becoming recognizable on plain radiographs[2]. Loose osteochondral bodies in the subacromial space can cause various disorders such as impingement syndrome[13,14], rotator cuff tears[13,14] and even constrictive capsulitis[21]. The differential diagnosis of primary SOC should include secondary osteochondromatosis[4], autoimmune diseases where loose bodies are formed such as rheumatoid arthritis and seronegative arthritis[22,23,24], tuberculous arthritis[25], pigmented villonodular synovitis[28], osteochondritis dissecans[25], osteochondral fractures[25], and secondary chondrosarcoma[26,27,28]. Valuable assets in the differential diagnosis besides detailed history, physical examination, and plain radiographs are CT scan, MRI[28], biopsy[26,27,28] and serologic examination (CRP, ANA, HLA-compatibility, ESR).

Malignant transformation is considered to be very rare by all authors. To our knowledge, there has been no case of malignant transformation of SOC in the shoulder[14,26,27,28]. However, sudden aggravation of a chronic symptoms, along with pain, oedema, inflammation and inhibition of movement should alarm the physician for the possibility of malignant transformation[26,27,28].

There is quite an argument in the literature whether simple removal of the loose bodies is sufficient, as SOC is

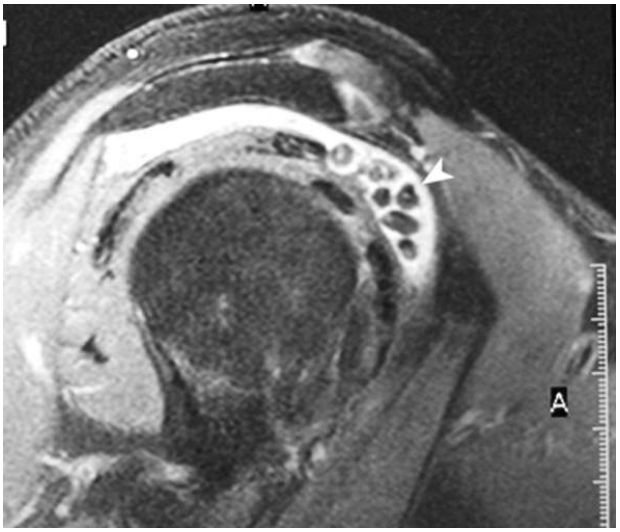

Figure 3. MRI of left shoulder (T1 sequence, parasaggital view). Presence of several loose bodies in the posterior subacromial bursa.

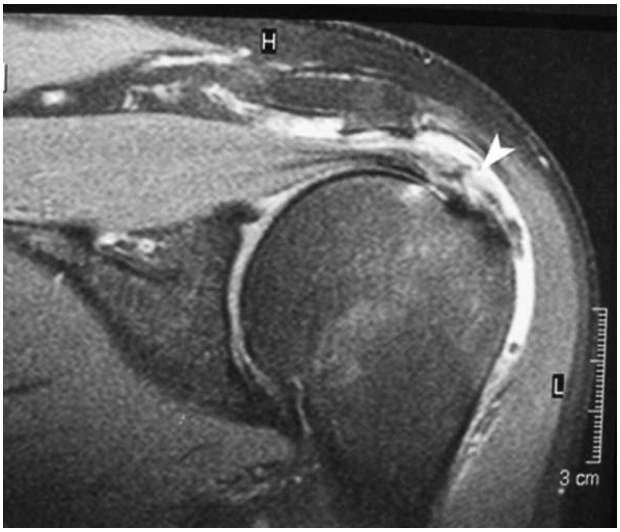

Figure 4. MRI of left shoulder (T2 sequence, coronal view) shows tendinopathy of the supraspinatus along with suspicion of a partial tear on its subacromial side.

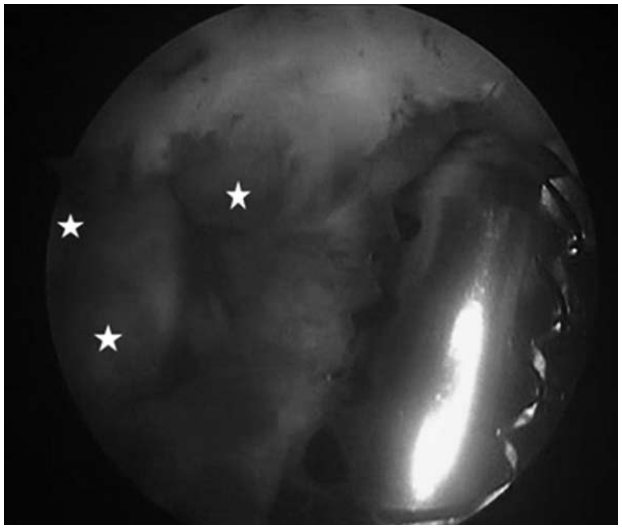

Figure 5. Several of the osteochondral bodies were attached to the supraspinatus bursa and were removed arthroscopically. Synovectomy was necessary to remove all diseased synovium.

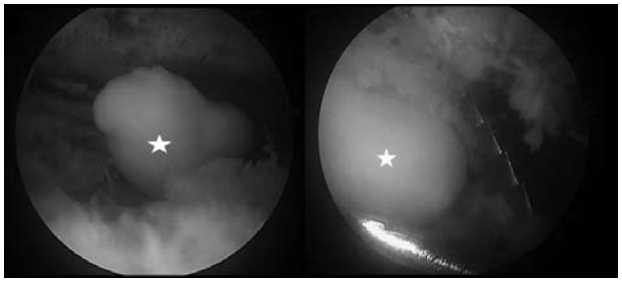

Figure 6. Arthroscopic location and removal of the osteochondral bodies.

considered by many authors to be self-limiting, or additional extensive synovectomy is essential. There is no sufficient evidence to support either side however, it is our belief that synovectomy is expedient as to avoid recurrence and also as a means of reducing intraarticular symptoms such as shoulder pain and third body wear.

Shoulder arthroscopy provides considerable advantages in treating SOC, as it is possible to detect and remove loose osseocartilaginous bodies without arthrotomy or detachment of the deltoid muscle. Perfect visualization of the lesion can be achieved enabling the surgeon to perform extensive synovectomy and rotator cuff repair when necessary.

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