



Rare case of extra-articular with Intra-articular synovial chondromatosis of right shoulder in 23 years old female

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ABSTRACT:

Introduction: Synovial chondromatosis (SC) is a rare condition in which cartilaginous masses are formed by metaplasia of the synovial membrane with infrequent involvement of shoulder joint and extraarticular presence. It characteristically involves single joint but extra-articular forms in the form of tendon sheath or bursal involvements have been rarely reported. It usually occurs in third-fifth decade and shoulder joint involvement is infrequent. Franklin first reported extra articular synovial chondromatosis in 1977. SC involving shoulder joint is rare but extra articular involvement is very rare.

Case Report: We present a case of Synovial chondromatosis 23 year-old-, right-handed-dominant female had presented with complain of multiple swelling, decreasing range of motion for 5 months and occasionally locking of right shoulder.

Magnetic Resonance Imaging was suggestive of multiple loose bodies in joint capsule with surrounding subacromial bursa and sub deltoid region. Extra articular loose bodies were removed by delto-pectoreal approach followed by shoulder arthroscopic was performed for synovectomy and removal of intra articular loose bodies.

Conclusion: Mono articular synovial chondromatosis with extra articular involvement of shoulder is rare case. We believe the success of arthroscopic surgery in selected patients with synovial chondromatosis. Extra articular lesion should not be neglected and treated aggressively with aim of open excision with through synovectomy. We further emphasize the need to follow these patients to detect recurrence or malignant transformation.

Keywords: Synovial chondromatosis

INTRODUCTION:

Synovial chondromatosis (SC) shoulder joint is a rare condition in which cartilaginous masses are formed by metaplasia of the synovial membrane (1, 2). The synovial chondroid metaplastic focus becomes peduncular then by breaking off, becomes a free fragment within the joint. These free fragments may undergo endochondral ossification or cause erosive damage to the joint surface. (3)

Synovial chondromatosis usually occurs in the age group of 30 yrs to 50 yrs (4) but also has been reported in children as young as 9yrs (5) and 23

years. It usually presents as swelling in or around the joint and restriction of movements and involve single joint. Secondary forms occur in which cartilage is found in the synovial membrane in association with loose bodies and degenerative joint disease.

Synovial chondromatosis is a mono-articular arthropathy rarely seen in diarthrodial joints. Knee joint (70%) is the primary site of involvement followed less commonly by hip, elbow, ankle, temporomandibular, and shoulder joints (respectively (7–9). Primary synovial chondromatosis of the shoulder is a very rare condition (10).

Franklin first reported extra articular synovial chondromatosis in 1977(11) according to him SC involving shoulder joint is rare but extra articular involvement is very rare. Horti (6) has reported SC in subacromial bursa. There are few reports in the literature related to intraarticular and extra-articular localizations (12, 13).

Nowadays with advancement in shoulder surgery, arthroscopic surgery is frequently used in the treatment of shoulder pathologies. Reduced morbidity of arthroscopy in intra-articular, extra-articular, or combined pathologies and its large field visualization capability are its major advantages (14, 15).

Authors present a case of multiple swelling of capsule of right shoulder with large extra articular masses with subacromial and subdeltoid region with concomitant restricted movement of shoulder and its combined open and arthroscopic removal of mass was performed.



Fig. 1: Right shoulder showing swelling and in joint. Loose bodies in joint and periarticular area



Fig. 2: AP view showing multiple calcified fullness

Case Report

23 year-old-, right-handed-dominant female had presented with multiple swelling decreasing range of motion for 5 months and occasionally locking of right shoulder. She had minimal pain on extremes of movements of joint. She had no history of shoulder trauma.

On physical examination, overlying skin was normal. There was no dilated vein, scar, sinus and discoloration of skin. Multiple swellings were present at anterior axillary region and extended over medial surface of arm. Degrees of flexion, abduction, and external rotation were 100°, 70°, and 30°, respectively, and internal rotation was through lumbosacral level. Strength and sensory evaluations were within normal limits and symmetric in both shoulders.

Hemogram, CRP, sedimentation rate, vitamin D, parathyroid hormone, calcium, and phosphate levels were normal. Serologic tests for rheumatoid arthritis, tuberculosis negative.

The pain AP radiograph of right shoulder was normal except multiple calcium flaks without any secondary degenerative changes. Multiple loose bodies in joint capsule with surrounding subacromial bursa and subdeltoid region were diagnosed in magnetic resonance imaging (Figure 3). Under general anaesthesia first in supine position extra articular loose bodies were removed by delto-pectoreal approach followed by arthroscopic examination and arthroscopic removal of intra articular loose bodies was performed.



Fig. 3: Multilobular hyperintense lesion

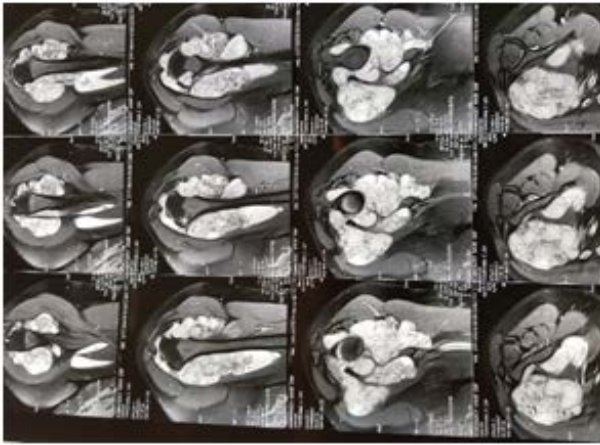


Fig. 4: An Arthroscopic view chondromatosis foci obliterating joint space and extending to adjacent bursa.

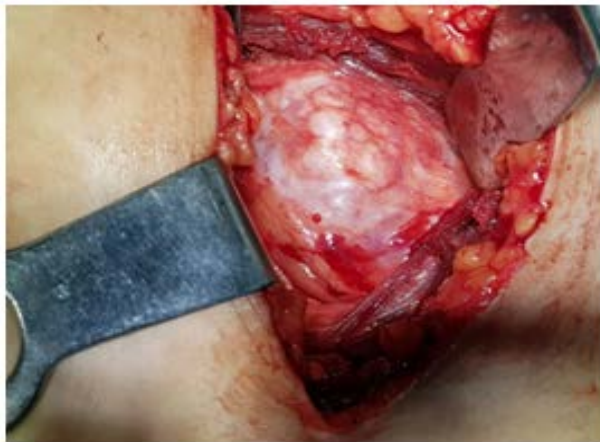


Fig. 5: Intra- Op view



Fig. 6: Loose bodies (size 2mm-10mm)

Multiple well defined approximately 50 osteochondral loose bodies were present with multiple sizes ranging from 10x10 mm size to 3x5 mm size. All loose bodies were removed

arthroscopically with subtotal synovectomy was performed.

Shoulder immobiliser for 3 days in early postoperative period was applied and both passive and active range of motion exercise in the first 3 months was started. Patient started to work again in the second postoperative month. She had gained almost full range of motion at the end of 6 month.

3. Discussion

Synovial chondromatosis (SC), first described by Reichell in 1900(1) , is a rare benign disorder characterized by chondroid metaplasia with multinodular proliferation of the synovial lining of a diarthrodial joint, bursa, or tendon sheath.

Cases of synovial chondromatosis in the shoulder have been rarely reported in literature. Generally the disease is self-limiting. Although the definitive aetiology of synovial chondromatosis is not known, it can be classified as primary or secondary. Secondary reasons include primarily trauma, then osteochondritis dissecans, rheumatoid arthritis and tubercular arthritis. Clinically, symptoms are generally not specific. Restrictions in the joint range of movement occur associated with the mechanical effect of the free fragments and in periods of active use, local pain and swelling may be seen in the shoulder. As the visualisation of lesions which are not calcified would be difficult on direct radiographs, examination with magnetic resonance imaging is recommended in diagnosis (3)

In a study by Lunn et al., it was reported that osteoarthritic changes could be seen in patients with synovial chondromatosis. Therefore, even when the patient has no symptoms, removal of the free fragments was recommended to prevent joint degeneration. In the same study, it was also reported that by damaging the tendon of the biceps long head, free fragments in the bicipital groove could cause tendinitis and pain (3).

The extra-articular form of synovial chondromatosis is particularly rare; it may arise from capsular tissues, tendon sheaths, and bursae that do not communicate with the joint space.(16) It is characterized by a progressively enlarging painful soft-tissue mass and is occasionally misdiagnosed as soft-tissue chondrosarcoma.(16)

Most of the reported cases in the literature have intra-articular involvement and extra-articular disease is an extremely rare entity (1).

In addition, even though the disease is rare, it displays the possibility of malignant transformation. (3).

Both open and closed arthroscopic methods can be used in the treatment of synovial chondromatosis. To reach the joint in the open method, a subscapular tenotomy is necessary and there are reports that associated with this there are disadvantages such as inadequate tendon repair and attachment site (3). This situation extends the healing period and delays rehabilitation. However, the advantages of arthroscopic treatment include good visualisation, low morbidity, rapid healing and early rehabilitation (3).

Arthroscopic or other classic treatments decision is totally dependent on the surgeon's experience. In our case intra articular free fragments are all in anterior, posterior and axillary etc. For clear vision of fragments and effective early postoperative rehabilitation arthroscopic treatment was performed.

Preoperatively with occasional locking of joint, showed rapid recovery and the locking was completely resolved. However, synovectomy is limited in arthroscopic treatment and there are advantages such as potentially access to all regions such as the axillary recess except of biceps sheath (3).

There is no published data comparing the results of the two techniques. There are debates about performing synovectomy concomitant with extirpation of loose bodies. There are some data in the literature consistent with the reduced disease recurrence after synovectomy (10, 17).

In conclusion, Mono articular synovial chondromatosis with extra articular involvement of shoulder is very rare case. The success of arthroscopic surgery has a role in selected patients with synovial chondromatosis. Full visualization capability during surgery, less morbidity, permission of early rehabilitation program, and decreased time of patient recovery are advantageous parameters in selecting

arthroscopic approach especially in the presence of synchronous intra- and extra-articular pathology. Extra articular lesion should not be neglected and treated aggressively with aim of open excision with through synovectomy. Malignant transformation to chondrosarcoma is known complication but fortunately, rarely seen.

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