

A Rare Case of Multiple Rice Bodies in Glenohumeral Joint, Subscapular Recess and Along Long Head of Biceps

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What to Learn from this Article?

Synovial chondromatosis presenting with multiple rice bodies at a rare site, such as glenohumeral joint, subscapular recess and along the long head of biceps which is < 5% reported till date.

Abstract

Introduction: Synovial chondromatosis is a rare, generally benign condition which affects the synovial membranes and commonly involves the large joints such as the knee, and hip. It is usually mono-articular and more common in males. Synovial chondromatosis is characterized by the presence of multiple cartilaginous nodules in the joint synovium. The definitive diagnosis is achieved after the pathological examination of the synovial tissue. It can be very destructive and can cause severe osteoarthritis, pain and malignant transformation. We present a rare case of primary synovial chondromatosis of the shoulder joint in a 31-year-old male patient

Case presentation: A 31-year-old man presented with pain and restricted movements of left shoulder for past 6 months, which was insidious in onset and gradually progressive. He had no history of trauma, fever, loss of appetite or weight loss. No tenderness or warmth over shoulder joint was observed. Restriction of movements was observed in all directions. No abnormality was detected in central nervous, cardiovascular, respiratory, genitourinary, or gastrointestinal systems. Routine pre-operative investigations (including liver function and renal function tests) were within the normal limit, serological investigation for rheumatoid arthritis was negative. Excision biopsy of loose bodies was performed. Biopsy reported histological features of synovial chondromatosis.

Conclusion : Synovial osteochondromatosis of shoulder joint, subscapular recess and along the long head of biceps is a rare case (less than 5% cases reported till date). Understanding the pathology, recognizing the radiographic and MRI appearance of primary synovial chondromatosis and differentiating it from secondary form, malignancy and other synovial pathologies are important in the diagnosis and clinical management of these patients.

Keywords: Synovial chondromatosis, Loose bodies, Multiple cartilaginous nodule

Introduction

Synovial osteochondromatosis (SOC) is a benign metaplastic proliferative disorder of the synovium [1-5] which affects subintimal fibroblasts in synovial joints [4, 6], tendons, and bursae (involving articular or tendon sheath synovial membranes) in which multiple nodules of cartilage are produced [3, 5, 8]. Most of the nodules subsequently become detached from the synovial membrane and float in the joint [9]. The synovial fluid nourishes the loose bodies which can remain viable and increase in size [2, 3, 10]. It is a rare disease

and rarely affects the shoulder joint (5%); it has a male predominance affecting knee [11], elbow [8], hip [10], and ankle joints (in order of frequency) in the fourth/fifth decade of life [4]. It presents with a dull ache and stiffness of the joint. The intra-articular loose bodies obstruct the joint mobility and the joint may get locked in a particular position.

Three phases of synovial chondromatosis have been described [7]; Phase (1)- Active intrasynovial disease with nodules but no calcifications/intraarticular bodies; Phase (2)- Synovitis with osteochondral nodules in the synovial

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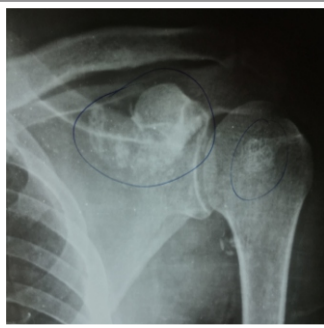


Figure 1: Radiographs at presentation



Figure 2, 3 & 4: MRI images showing loose bodies



Figure 5: Intra-operative picture showing deltopectoral approach.



Figure 6 & 7: Excision biopsy image showing multiple loose bodies.

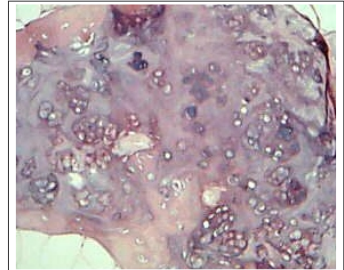


Figure 8: Histological picture showing microscopic appearance of one of the nodules of synovial chondromatosis.

membrane and loose bodies within the joint; Phase (3)- Multiple loose bodies remain but synovitis is quiescent.

In relation to shoulder joint, primary synovial chondromatosis has been reported in the subacromial bursa, subclavicular region and along the brachial plexus, but all are associated with the involvement of the glenohumeral joint [12-15]. We present this case because it involved subacromial bursa and shoulder joint.

Case Presentation

A 31-year-old man presented with pain and restricted movements of left shoulder since 6 months, which was insidious in onset and gradually progressive. He had no history of trauma, fever, loss of appetite or weight loss. No tenderness or warmth over shoulder joint was observed. There was restriction of movements in all directions. Routine pre-operative investigations (including liver function and renal function tests) were within the normal limit, serological investigation for rheumatoid arthritis was negative. X-ray of left shoulder showed multiple osseous loose bodies in subcoracoid region and shoulder joint overlapping on the bone [Fig. 1]. On MRI, multiple (2 to 3mm) rice bodies were visualized in the glenohumeral joint, subscapular recess and along the long head of biceps. There were large filling defects upto 8 - 12 mm in subscapular recess anterior to subscapularis muscle. Ac joint and rotator cuff appears to be normal [Fig. 2, 3, 4]. The joint was exposed through a linear incision over the deltopectoral groove [Fig. 5]. Excision biopsy of loose bodies was performed [Fig. 6, 7]. Biopsy reported histological features of synovial chondromatosis [Fig. 8]. The wound healed well within 2 weeks and the patient recovered with full range of motion of the affected shoulder. At one year follow-up, the patient does not have any recurrence.

Discussion

Synovial chondromatosis is a rare disease [7] and rarely affects the shoulder joint (5%) [3, 5, 9]. 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. Primary synovial osteochondromatosis is not associated with frank osteoarthritis or synovitis [2] and the source of the

osteochondral loose bodies is the benign synovial neoplasia [14]. Both the synovial membrane and articular cartilage develop from the same mesenchymal tissue, and this tissue arises from embryonic rests [9].

Multiple (even hundreds) cartilaginous nodules are commonly formed. The chondrocytes become pedunculated and encrusted inside the synovium [11]. They are visible on plain X-ray only when the bodies are calcified or ossified [10] but the number is always greater than one would suspect from the film. Air or double contrast arthrography may be necessary to visualize the nonossified chondromatous bodies [11]. However, almost all such bodies were visualized with MRI. Loose bodies must be removed early to halt further damage to articular surfaces [10]. Synovial osteochondromatosis does not resolve spontaneously [7] and complications like degenerative osteoarthritis [10], joint subluxation [10] and bursitis [8] are not uncommon. It is controversial whether to perform complete synovectomy along with excision of all communicating bursae, because recurrences are common [11] irrespective of the extent of the excision. Therefore, many authors favor removal of all loose bodies with subtotal synovectomy [4, 12].

Mukerjee [12] reported SOC of the shoulder joint in a 12-year-old boy. Bues and Friedrich [13] reported a case of a 22-year-old man with SOC of the shoulder joint. Recently, Antonogiannakis et al [14] reported a case of osteochondromatosis of the subacromial bursa in a 72-year-old retired military officer.

Though suspected in some observations, [13] sarcomatous degeneration of the synovial chondromatosis has not been proved.

Conclusion

We conclude that synovial osteochondromatosis of shoulder joint, subscapular recess and along long head of biceps is a rare case (less than 5% cases reported till date). Understanding the pathology, recognizing the radiographic and MRI appearance of primary synovial chondromatosis and differentiating it from secondary form, malignancy and other synovial pathologies is important in diagnosis and clinical management of these patients.

Clinical Message

Clinical and radiological diagnosis of the case is challenging. Removal of loose bodies and synovectomy with histopathological confirmation is of prior importance.

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