



Synovial osteochondromatosis of the shoulder in an elderly rheumatoid arthritis patient

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Abstract

Synovial osteochondromatosis (SC) of the shoulder is a rare condition characterized by synovial membrane proliferation and metaplasia, with development of multiple osteocartilaginous nodules: the osteochondromas. Any joint may be affected. The knee is the most frequent site but the shoulder is rarely reported in the literature. It can be divided into primary and secondary forms, which are not clinically or pathologically different. Conventional radiography may be negative in the first stages of the disease, but magnetic resonance imaging and computed tomography are often crucial for early diagnosis, the treatment remains surgical. We presented a 62-year-old woman, a case of seropositive rheumatoid arthritis with secondary synovial chondromatosis of the shoulder. Diagnosis is established with the help of Computed tomography and histo-pathological examination.

Keywords: shoulder, synovial osteochondromatosis, rheumatoid arthritis

Introduction

Synovial chondromatosis is a rare tumor-like lesion of multiple cartilaginous nodules, which may occur intra-articularly (articular synovial membrane) or rarely, extra-articularly (bursa or tendon sheath synovium) ^[1]. It is classified as either primary or secondary; primary synovial osteochondromatosis is a benign metaplasia of the synovial membrane, which manifests as multiple cartilaginous or osteocartilaginous nodules within a joint, bursa and tendon sheath. It typically results in pain and stiffness of the affected joint, and is thought to arise as a result of synovial membrane proliferation and metaplasia ^[2]. In contrast, secondary synovial chondromatosis results from the growth of separated particles from the articular cartilage or osteophytes in joint diseases, such as osteochondritis dissecans, osteoarthritis, and osteochondral fractures ^[3].

The knee is involved most frequently, followed by the hip, elbow, shoulder, ankle, and wrist. However, smaller joint involvement, including that of the spine, foot, and hand, has been reported ^[4, 5]. Historically, synovial osteochondromatosis of the shoulder is characterized as a rare monoarticular benign arthropathy of uncertain etiology, typically involving a single large joint in a young adult. In this article, we presented a 62-year-old woman, a case of seropositive rheumatoid arthritis (RA) with secondary synovial chondromatosis of the shoulder. Diagnosis is established with the help of imaging modalities and histo-pathological examination. This case is reported for its rarity.

2. Case presentation

A 62-year-old woman, retired teacher, was operated in 2016 for right supraspinatus rupture. The patient had a history of 10 years deforming erosive rheumatoid arthritis, affecting large, medium and small joints, treated by corticoid and methotrexate with

significant evolution. There was no history of trauma or any other inflammatory or infectious disease. At the time of her hospitalization to evaluate her rheumatoid arthritis, the disease was in remission; nonetheless, she reported a history of left shoulder chronic pain. Physical examination showed no obvious deformity, there was some limitation of the range of motion of the left shoulder, which was painful beyond 110 degrees abduction; resisted active motion was also painful, shoulder impingement signs were positive. Distal neurovascular status was intact, the right shoulder and other joints were normal on examination; the rest of the examination was within normal limits. Plain radiographs of left shoulder revealed several geodes making an impression on the humeral head (Fig.1).



Fig 1: Anteroposterior radiography of the left shoulder in a patient with RA shows several geodes making an impression on the humeral head

In this stage, radiographic characteristics were not typical for osteochondromatosis, several differential diagnoses were discussed, hydatid cyst located in the subacromial–subdeltoid bursa, bursal osteochondromatosis of the shoulder, synovial sarcoma, pigmented villonodular synovitis, multiple rice body formations, which are seen in the septic or rheumatoid affections,

whose diagnosis is histological. To confirm the diagnosis of osteochondromatosis of the shoulder, a computed tomography was performed; magnetic resonance imaging (MRI) was not available; it showed many rounded bodies, around the glenohumeral joint and into the subacromial bursa with high density and signs of degenerative osteoarthritis (Fig.2).

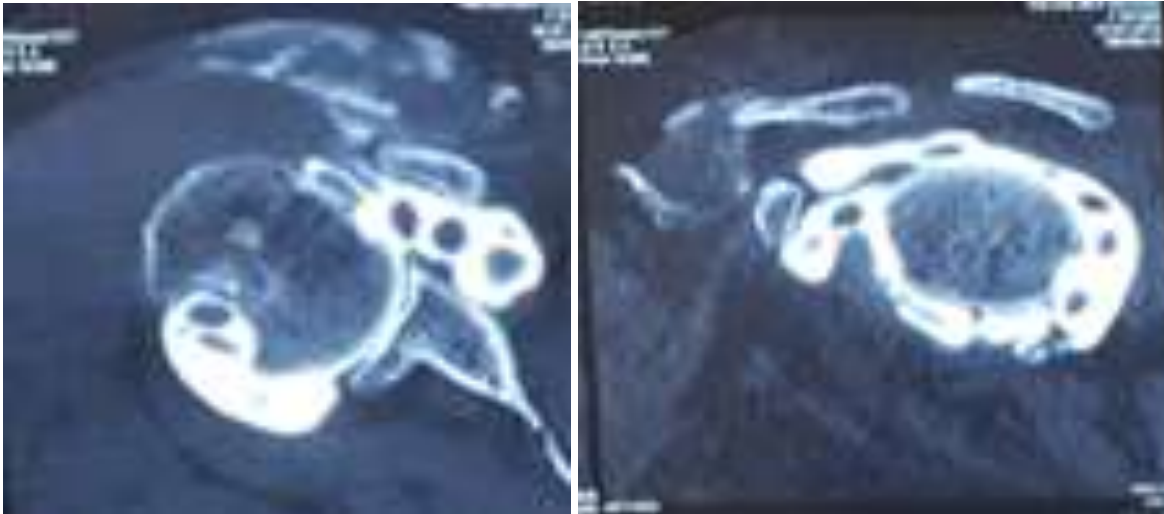


Fig 2: Computed tomography of the left shoulder showing several rounded bodies of subacromial bursa and the glenohumeral joint

We performed an open surgical exploration to remove the osteochondromes around the glenohumeral joint and into the subacromial bursa (Fig.3), followed by total synovectomy. Post-operative histopathological examination of the loose bodies confirmed the diagnosis of synovial chondromatosis.



Fig 3: Removed osteochondromes of the glenohumeral joint and the subacromial bursa.

3. Discussion

Synovial chondromatosis of the subacromial bursa is rare, with only few cases reported in the literature [6, 7, 8, 9]. This condition is thought to be due to chondroid metaplasia of the synovium, the pathogenesis of the nodules is unknown. Synovial chondromatosis is either primary or secondary, primary synovial chondromatosis occurs idiopathically, secondary synovial chondromatosis appears to be more common, it is a response to an irritation of the joint capsule or bursal lining due to degenerative osteoarthritis, rheumatoid arthritis, osteonecrosis, osteochondritis dissecans, trauma, neuropathic osteoarthropathy and tuberculosis [10]. Synovial chondromatosis is often diagnosed by a thorough physical examination and radiographic examination. The clinical manifestations of SC include swelling, pain, and a palpable mass and limited joint motion [11], all of which are not specific and prevent a precise specific diagnosis. Therefore, early diagnosis requires a high index of suspicion and a detailed examination. The radiographic images depend on the stage of the disease and range from normal findings to the presence of multiple, radiopaque, calcified bodies in the joint cavity [12]. However, plain radiographs of our patient showed several geodes making an impression on the humeral head, calcification has not yet occurred. Milgram proposed three stages of disease process, at stage one, there is active intrasynovial disease but no free fragments, at stage two, there is active intrasynovial proliferation, lesions are seen in transition to free fragments, and finally the third stage is the presence of multiple osteochondral free fragments without any active intrasynovial disease [13]. Plain film radiographs are helpful only in the third phase of the disease, once calcification has occurred. Computed tomography (CT) is the best diagnostic tool for osteochondromatosis, MRI is also useful in identifying and localizing the lesions, and help in distinguishing between other

differential diagnoses^[14]. MRI shows three distinct patterns of synovial chondromatosis, which correlates, with Milgram's phases of SC. Furthermore, advanced imaging modalities including magnetic resonance imaging or CT may show non-specific findings due to comparable intensity of loose bodies with the synovial fluid. Likewise, the clinical presentation of the disease is not special for the diagnosis and may overlap with other conditions including meniscus damage, osteoarthritis, osteochondritis dissecans, discoid meniscus, and synovial cysts. For this reason, the ultimate diagnosis of SC can only be made through direct visualization of free bodies via arthroscopic or open surgery followed by pathological examination of the removed tissues.

The classic treatment for this disease is open arthrotomy, synovectomy and complete removal of the free fragments^[15]. In our case, and when SC is largely diffused, removal of free cartilaginous segments along with the synovium seems inevitable. To provide a curative treatment and prevent future recurrence of the disease, synovectomy is always significant^[16]. The importance of our case presentation was osteochondromatosis concurrent with rheumatoid arthritis (RA), metaplastic transformation of synovial lining cells into chondrocytes has been explained as a potential underlying mechanism^[17]. Nevertheless, the stimulus of this transformation is uncertain, it is not clear if the rheumatoid disease may provoke such a transformative process. Few authors have reported occurrence of SC concurrently with RA while some others have documented SC following arthroplasty surgery^[18, 19]. Overall, synovial chondromatosis in RA is rarely reported^[20, 21].

Conflicts of interest

The authors declare no conflicts of interest.

References

1. Bhadu Danveer. "Synovial chondromatosis involving multiple joints in rheumatoid arthritis: A case report review of the literature." *International journal of rheumatic diseases*. 2017; 20(12):2148-2151.
2. Llauger Jaume. "Nonseptic monoarthritis: imaging features with clinical and histopathologic correlation" *Radiographics*. 2000; 20(1):S263-S278.
3. Milgram James W. "The classification of loose bodies in human joints." *Clinical orthopaedics and related research*. 1997; 124:282-291.
4. Garg Bhavuk. "An unusual cause of shoulder pain: self-assessment questions." *The New Zealand Medical Journal (Online)*. 2007; 120:1260.
5. Sah Alexander P. "Malignant transformation of synovial chondromatosis of the shoulder to chondrosarcoma: a case report." *JBJS*. 2007; 89(6):1321-1328.
6. Huang Tung-Fu, Jiunn-Jer Wu, Teng-Shung Chen. "Bilateral shoulder bursal osteochondromatosis associated with complete rotator cuff tear." *Journal of shoulder and elbow surgery*. 2004; 13(1):108-111.
7. Park Jung Ho. "Arthroscopic treatment for synovial chondromatosis of the subacromial bursa: a case report." *Knee Surgery, Sports Traumatology, Arthroscopy*. 2007; 15(10):1258-1260.
8. Kumar Anil, Arvind Aggarwal, Virender K Sahni. "Primary synovial osteochondromatosis of a subdeltoid bursa." *Indian journal of orthopaedics*. 2010; 44(1):104.
9. Kiritsi O, Tsitas K, Grollios G. "A case of idiopathic bursal synovial chondromatosis resembling rheumatoid arthritis." *Hippokratia*. 2009; 13(1): 61.
10. Peh WCG. "Osteochondroma and secondary synovial osteochondromatosis." *Skeletal radiology*. 1999; 28(3):169-174.
11. Chillemi Claudio, Mario Marinelli, Vincenzo de Cupis. "Primary synovial chondromatosis of the shoulder: clinical, arthroscopic and histopathological aspects." *Knee Surgery, Sports Traumatology, Arthroscopy*. 2005; 13(6):483-488.
12. Ko Jih-Yang. "Synovial chondromatosis of the subacromial bursa with rotator cuff tearing." *Journal of shoulder and elbow surgery*. 1995; 4(4):312-316.
13. Milgram James W. "Synovial osteochondromatosis: a histopathological study of thirty cases." *The Journal of bone and joint surgery. American volume*. 1977; 59(6):792-801.
14. Razek AAK, Abdel M Castillo. "Imaging appearance of primary bony tumors and pseudo-tumors of the spine." *Journal of neuroradiology*. 2010; 37(1):37-50.
15. Davis RI, Hamilton A, Biggart JD. "Primary synovial chondromatosis: a clinicopathologic review and assessment of malignant potential." *Human pathology*. 1998; 29(7):683-688.
16. Ogilvie-Harris DJ, K Saleh. "Generalized synovial chondromatosis of the knee: a comparison of removal of the loose bodies alone with arthroscopic synovectomy." *Arthroscopy*. 1994 ; 10(2):166-170.
17. Jeffreys TE. "Synovial chondromatosis." *The Journal of bone and joint surgery. British volume*. 1967; 49(3):530-534.
18. Witwity T. "Shoulder rheumatoid arthritis associated with chondromatosis, treated by arthroscopy." *Arthroscopy: The Journal of Arthroscopic & Related Surgery*. 1971; 7(2):233-236.
19. Crawford Matthew D, Hubert T Kim. "New-onset synovial chondromatosis after total knee arthroplasty." *The Journal of arthroplasty*. 2013; 28(2): 375-e1.
20. Day JH, JM Rosas De. "Synovial chondromatosis with rheumatoid arthritis (report of a case)." *Medicina*. 1971; 31(3):222-227.
21. Tahmasebi MN. "Bilateral synovial knee chondromatosis in a patient with rheumatoid arthritis: case-report and literature review." *Archives of Bone and Joint Surgery*. 2014; 2(4):260.