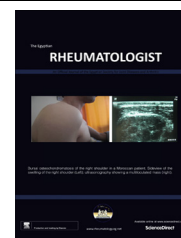




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CASE REPORT

Bursal osteochondromatosis of the shoulder: An exceptional location



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KEYWORDS

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Abstract *Introduction:* The osteochondromatosis is a rare synovial tissue metaplasia. The location at the shoulder is very rare. Bursal affection is exceptional.

Case presentation: We report a case of a male, 41 years old living in north Morocco and presenting to the rheumatology outpatient clinic at Fes Hassan II University Hospital with a swelling of the right shoulder that had been evolving for two years, without any history of trauma, gradually increasing in size, without fever or alteration of the general state. Clinical examination revealed a 5 cm soft, non-tender oval mass on the anterior surface of the right shoulder with no signs of inflammation but limited range of motion. Plain X-ray was normal and diagnostic ultrasonography revealed an avascular multilocular cystic mass. A differential diagnosis was considered and other conditions with comparable pictures were ruled out. The diagnosis was confirmed by a magnetic resonance imaging showing synovitis and distension of the sub acromiodeltoid bursa with many rounded bodies, few millimeters in size of intermediate signal in T1 and T2 without enhancement after intravenous gadolinium. A diagnosis of osteochondromatosis of the subacromiodeltoid bursa (third stage) was made. The patient underwent an open bursectomy and histological examination further verified the diagnosis.

Conclusion: Presenting such rare cases could help in raising the awareness of rheumatologists regarding such conditions when considering a differential diagnosis for patients presenting with monoarticular shoulder swelling. Excision of the tumor has a remarkably better prognosis.

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1. Introduction

Primary synovial chondromatosis is a proliferation of cartilaginous bodies within the synovial membrane, bursa, or tendon sheath. Historically, it has been characterized as a rare, monoarticular, benign arthropathy of uncertain etiology, typically

involving a single large joint in a young adult. Males are affected more commonly than females. 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 [1,2].

Synovial osteochondromatosis is a benign metaplastic proliferative disorder of the synovium characterized by the formation of multiple cartilaginous nodules in the synovium, many of which detach and become loose bodies [3]. The synovial osteochondromatosis is a cartilaginous metaplasia of synovial tissue. This metaplasia may occur at any site where synovial tissue is present, but can also occasionally affect the bursae and tendon sheaths [4–6].

Osteochondromatosis is characteristically monoarticular, most commonly involving the knee. Primary synovial osteochondromatosis is known to be intra-articular and wherever it is observed outside a synovial joint, it is associated with the involvement of the nearby joint [7]. The presenting symptoms are usually diffuse discomfort in the affected joint and decreased range of motion with an accompanying gritty or locking sensation [3].

Primary synovial chondromatosis of the shoulder is a very rare condition [8–10]. It has not been reported to involve a subdeltoid bursa [7]. The frequency of synovial osteochondromatosis of the shoulder ranks 4th after knee, elbow and hip synovial osteochondromatosis.

In this article we present a new case report of bursal osteochondromatosis involving the shoulder subacromiodeltoid bursa.

2. Case presentation

Mr. L.M. 41 years old, merchant, living at Nador, in the north of Morocco, without significant medical history, presented for rheumatology consultation at the University Hospital Hassan II at Fes, with a swelling of the right shoulder that had been evolving for two years, without any history of trauma, gradually increasing in volume, without fever or alteration of the general state.

Clinical examination revealed a soft, non-tender oval mass on the anterior surface of the right shoulder, without signs of inflammation, measuring about 5 cm in diameter (Fig. 1). There was some limitation of the range of motion of the right shoulder. The left shoulder and other joints were normal on examination.



Figure 1 Side view of the swelling of the right shoulder.

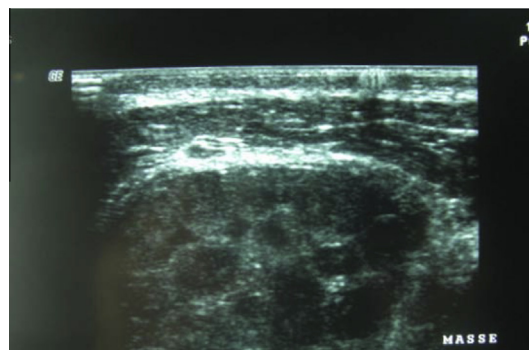


Figure 2 Ultrasonography of the right shoulder showing a multiloculated mass.

Plain radiographs of the shoulder were normal. Ultrasonography of the shoulder revealed a cystic mass containing multiple loculi that were found to be avascular by Doppler (Fig. 2).

Hydatid cyst localized in the shoulder was the most likely diagnosis. A hydatid serology was made, using the ELISA technique, and was positive. Routine laboratory investigations were normal. There was no eosinophilia and the acute phase reactants were normal.

In this stage, several differential diagnoses were discussed, in addition to the hydatid cyst located in the subacromiodeltoid bursa, including:

- Bursal osteochondromatosis of the shoulder.
- A synovial sarcoma which may be accompanied in 32% of cases with calcifications that project extra-articularly since the tumor begins only rarely in the joint (10%) [11]. Malignant bone erosions are seen in 12% of cases.
- Pigmented villonodular synovitis as the MRI highly suggested the diagnosis by showing hemosiderin deposits.
- Multiple rice body formations, which are seen in the septic or rheumatoid affections, whose diagnosis is histological.

To confirm the diagnosis, an MRI was performed; it showed synovitis of the shoulder joint and distension of the subacromiodeltoid bursa with many rounded bodies, few millimeters in size of intermediate signal in T1 and T2, without enhancement after intravenous gadolinium (Fig. 3).

A diagnosis of osteochondromatosis of the subacromiodeltoid bursa (third stage) was made. The patient underwent an open bursectomy and histopathological examination further verified the diagnosis of bursal osteochondromatosis of the shoulder.

3. Discussion

The osteochondromatosis is a synovial metaplasia in which connective tissue cells acquire the capacity for chondrogenesis [3]. The cartilaginous foci can form pedunculated bodies that may be free (chondromatosis). These cartilaginous bodies can then calcify or ossify and become radiopaque (osteochondromatosis) [11,12].

The osteochondromatosis can be either primary (about 10% of cases) where the etiology and pathogenesis are unknown, occurring in healthy joints, or secondary occurring in joints affected by a degenerative process (osteoarthritis,

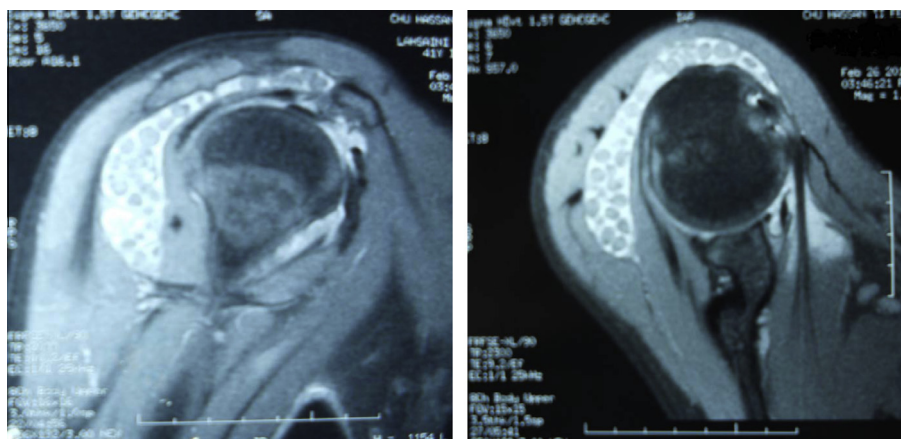


Figure 3 MRI of the right shoulder showing synovitis with several rounded formations of subacromial–subdeltoid bursa.

synovitis, trauma. . .). The initiation of the metaplastic process is secondary to a reaction of the synovial membrane to residual embryonic mesenchymal cells or vasomotor and inflammatory reactions caused by microtrauma. Synovial metaplasia affects the knee in more than half of the cases, then in order of decreasing frequency, elbow, hip, shoulder and ankle. It affects slightly more men than women and is found most often in adulthood between 20 and 50 years (range 13–90 years) [13].

The condition is often associated with few symptoms which explain the delay between the onset of clinical signs and consultation. Moreover, the diagnosis is delayed because chondromas are radio-visible only when they become ossified [14].

The disease progresses into three stages; the *early stage* is characterized by the synovial hypertrophy with the formation of chondroma encased in the synovium. These chondromas can become pedunculated and present ossification in the center. Calcifications can also occur in the necrotic areas, the *intermediate stage* is characterized by the release of chondroma or osteochondroma in the joint, spontaneously or after trauma, while in the third stage, chondromas or osteochondroma remains and synovial inflammation fades [15]. Malignant transformation is extremely rare, but it has been reported in a few instances [16–18].

Radiographs suggest the diagnosis at an advanced stage of disease, when chondromas become radiopaque. Ultrasound can allow reliable diagnosis and has the advantage of being inexpensive and noninvasive [19], but CT scan is the examination of choice followed by the MRI [20] with many advantages: demonstration of foreign cartilaginous radiolucent bodies, determining their number and exact location, which are essential before surgical or arthroscopic resection.

A dozen cases of osteochondromatosis of the shoulder have been reported in the literature [21]. Primary synovial chondromatosis of the shoulder has been reported in a 24-year-old man with a 6-month history of right shoulder pain and decreased range of motion. Computed tomography and MRI findings also led to the diagnosis. Furthermore as with our case, histologic examination confirmed the diagnosis [10]. In another case report, primary synovial osteochondromatosis has been presented in a 52-year-old woman having a large number of loose bodies in a huge tumor in the subdeltoid bursa. Plain X-ray revealed soft tissue swelling only with no areas of calcification. On MRI, multiple nonosseous loose bodies were visualized in the bursa deep into the deltoid muscle [7].

This case report is notable for the unusual bursal location and by the diagnostic problem, which was wrongly oriented toward a hydatid disease of the shoulder but the MRI and later histological examination confirmed the diagnosis of osteochondromatosis.

On the therapeutic level, there is no medical treatment known for osteochondromatosis. Its treatment is exclusively surgical excision of the entire bursa. The gross appearance of multiple foreign bodies in the size of chickpeas is characteristic of the condition. The recurrence rate is almost zero after surgical excision of the bursa (total excision of the bursa) [22,23]. The treatment of choice is excision of the synovium and removal of the loose bodies [3].

In the absence of treatment; the evolution is toward osteoarthritis. Arthroscopic partial synovectomy was performed in a similar case and is recommended in the treatment for synovial chondromatosis of the shoulder because of low morbidity and early functional return [10]. In another case, on surgical excision of subdeltoid bursa, a biopsy confirmed it to be cartilaginous loose bodies in synovial lining suggestive of metaplastic transformation of the synovial tissue [7]. Intervention is necessary before significant joint damage occurs, either by arthroscopy in early stages or by arthrotomy with or without synovectomy in advanced stages.

In conclusion, presenting such rare cases could help in raising the awareness of rheumatologists regarding such conditions when considering a differential diagnosis for patients presenting with monoarticular shoulder swelling. Excision of the tumor has a remarkably better prognosis.

Conflict of interest

None.

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