

CASE REPORT

Bilateral Synovial Knee Chondromatosis in a Patient with Rheumatoid Arthritis: Case-report and Literature Review

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*Research performed at Tehran University of Medical Sciences, Shariati Hospital, Tehran, Iran**Received: 2 May 2014**Accepted: 14 August 2014***Abstract**

We presented a female patient with RA complaining of progressive pain, swelling, and crepitation of the knee joints that was diagnosed with bilateral synovial chondromatosis (SC) of both knees. Radiographies revealed characteristic findings of SC including multiple calcified multifaceted loose bodies within both knees. Removal of cartilaginous segments as well as total synovectomy was performed arthroscopically on the left side and via open surgery on the right side. Short-term postoperative follow-up of our patient revealed improved knee function and resolution of all symptoms.

Key words: Bilateral, Rheumatoid arthritis, Surgery, Synovial Chondromatosis

Introduction

Synovial chondromatosis (SC) is a very rare disease commonly affecting large joints including knee, hip, and shoulder (1-3). Although the condition has been described as a benign neoplasm of the synovium, its progressive dissemination into the articular structures will result in joint destruction (3-7). The etiology of SC is not clearly known yet; nevertheless metaplasia of synovial lining tissue into chondrocytes has been explained as a probable cause (8). It usually affects a single joint in which the most common site is the knee and is twice as frequent in men as in women (5, 9-14). It is commonly seen during the third to fifth decades of life presenting with aggravating joint pain, swelling, crepitus, and limited range of motion (9-14). Moreover, palpable swelling and effusion may be the only clinical finding (11).

Most chondromatoses originate from the intracapsular structures while extracapsular structures have been rarely reported to be involved (11). Nevertheless, SC in a rheumatoid arthritic knee is even rarer. Despite few reports on bilateral chondromatosis of ankles and disseminated SC of the knee (11, 15), bilateral SC of the knee has not been reported. To the best of our knowledge, this is the first case reporting simultaneous bilateral SC of the knee in the presence of rheumatoid arthritis.

Case Presentation**Clinical History**

A 34-year-old female presented with progressive pain in both knees, swelling, limited range of motion, and frequent joint giving way and locking. The patient had a history of 10-year rheumatoid arthritis (RA) affecting both elbows as well as the knees, and was receiving prednisolone and methotrexate [Figure 1]. She was also reporting morning stiffness in wrists and hands. The primary diagnosis was assumed to be the systemic manifestation of RA involving multiple joint. At the time of admission to the hospital, the patient was somewhat independent in terms of activities of daily living with limited ability to walk without assistive devices.

Physical Examination

On examination, loose bodies were palpable inside the knees with marked swelling of the joints filling the suprapatellar pouch. No skin abnormality was noted over the knees. The range of right knee motion was 10° -90° (10 degrees of flexion contracture) while left knee range of motion was 5° -90° (5 degrees of flexion contracture). There was no ligamentous laxity and patella was stable as well. Patient's elbows showed limited range of motion (40-110) with 40 degrees of flexion contracture on both sides. On the other hand, swelling was notable over the

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Figure 1. Elbow radiograph in a patient with rheumatoid arthritis.

wrists and the proximal interphalangeal joints of both hands with no obvious deformity. Examination of the shoulders, hips, and ankles was unremarkable.

Imaging

Plain X-ray of both knees revealed multiple calcified loose bodies. Moreover, extensive calcification of bursa was evident on lateral views showing extension of calcification into proximal calf and distal thigh [Figure 2]. Since radiographic characteristics were typical for SC, other imaging modalities such as magnetic resonance imaging and computed tomography were not performed.

Treatment

As previously reported in the literature (5-6), the choice of treatment for synovial chondromatosis is surgery. We performed an open medial arthrotomy on the right knee to remove the loose bodies followed by total synovectomy [Figure 3]. Furthermore after three

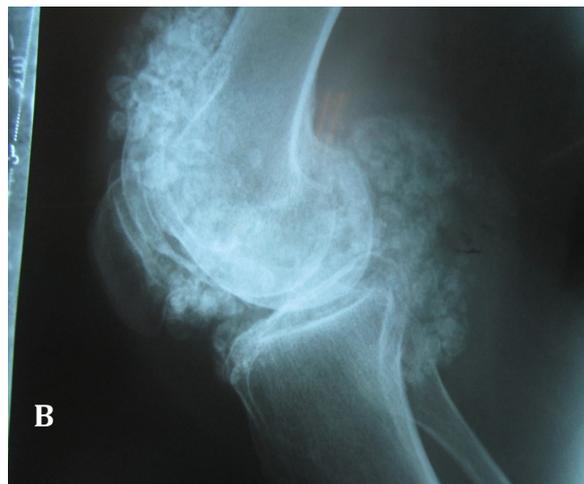
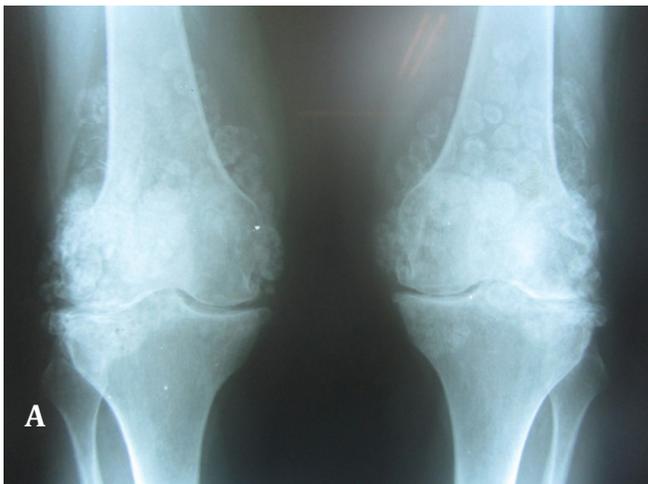


Figure 2. A. AP and B. lateral view of the knees showing multiple typical calcification and ossification due to synovial chondromatosis.

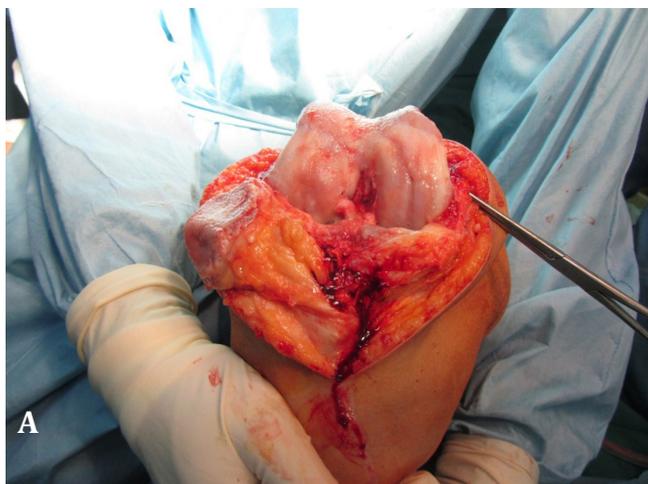


Figure 3. Open surgery of the right knee. A. Extensive cartilage damage due to chondromatosis. B. Right knee involvement by synovial chondromatosis.



Figure 4. Arthroscopic surgery of the left knee. A. Total synovectomy. B. Loose body removal. C. Multiple loose bodies removed from the joints.

month, the left knee was operated arthroscopically for the removal of loose bodies and total synovectomy [Figure 4].

Postoperative Rehabilitation and Follow-up

Early active assistive range of motion (ROM) was started following both surgeries. After six months, she gained a range of 5-120 degrees on both sides. Symptoms resolved significantly and patient was able to walk without any difficulty. Quadriceps muscle strength test revealed a muscle force of 4/5 on both sides [Figure 5].

Discussion

The importance of our case presentation was bilateral synovial chondromatosis concurrent with rheumatoid arthritis (RA). To date, we could not find any other reports on bilateral SC of the knees but only one report on shoulder RA that was along with chondromatosis (16). Metaplastic transformation of synovial lining cells into chondrocytes has been explained as a potential underlying mechanism (8). However, the stimulus of

this transformation is uncertain. Few authors have reported occurrence of SC concurrently with RA while some others have documented SC following arthroplasty surgery (6,16-18). Overall, it is not clear whether an inflammatory response to the surgical trauma or the rheumatoid disease may provoke such a transformative process.

In most cases as in our patient, SC originates from intraarticular and then extends to the extra-articular space (9, 11, 19). The initial X-ray of the knees was suggestive of SC. Given the picture of multiple round and cubic loose bodies with calcification was characteristic of SC, because of bilateral involvement, female gender, and concurrence with RA, preoperative diagnosis of SC was difficult.

Although SC is usually reported to affect joints unilaterally, there are few reports on bilateral involvement (6, 15). Men are affected twice more than women most of whom are diagnosed in the third to fifth decades of life (1, 5-6, 8, 19). Importantly, the initial imaging of the knees with extensive intra-articular calcification may raise the suspicion to tumoral calcinosis although generalized calcification usually affects African and Caribbean races and often presents in multiple sites (11). Nonetheless, histological studies on removed bodies should ultimately exclude the malignant potential of the suspected condition.

Diagnosis of SC relies on identification of cartilaginous nodules in synovium of the joints, tendon sheaths and bursae (8, 10, 14). This may not be easily possible unless in advanced stages in which loose bodies are calcified and are detectable within plain X-ray (20). Moreover, advanced imaging modalities including magnetic resonance imaging or tomography scanning may show nonspecific findings due to similar intensity of loose bodies with the synovial fluid. Similarly, clinical presentation of the disease including pain, locking, limited ROM, and swelling are not exclusive for the diagnosis and may overlap with other conditions including meniscus damage, osteoarthritis, osteochondritis dissecans, osteochondral fractures,

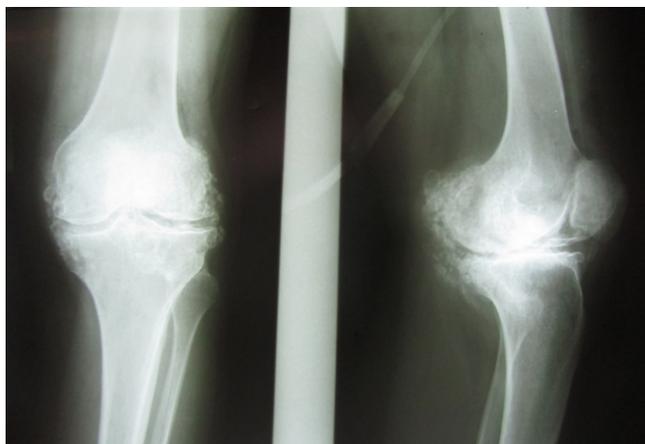


Figure 5. A. Postoperative X-ray of the right and B. left knee.

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.

Because the SC starts and disseminates from within the joint, with respect to presence of other conditions such as destructive joint diseases, arthroscopic removal of the free bodies may be sufficient. However, when SC is largely diffused, removal of free cartilaginous segments along with the synovium seems inevitable. For this reason, we performed arthrotomy and total synovectomy along with removal of the loose bodies to provide a curative treatment and prevent future recurrence of the disease.

After 6 month of follow-up, our patient showed improved function with resolution of symptoms including pain, swelling, giving way and locking, and regained knee extension. However, a longer follow-up may be needed to observe restoration of other functions most importantly the ROM after total synovectomy, and to monitor the recurrence of the disease because of

potential malignant transformation. It is not clear to us whether the patient would have benefited from a more extensive diagnostic approach for the earlier diagnosis in order to receive less invasive surgery given that synovial tissue is preserved and arthrotomy is reserved for advanced stages. Hence, we suggest reviewing the reported cases with consideration of timely diagnosis and long-term treatment outcomes, which may enable drawing a consensus in regards to the clinical diagnosis and surgical management of SC.

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