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الجمعية السعودية لجراحة العظام
Saudi Orthopedic Association

Journal of Musculoskeletal Surgery and Research

Case Report

Multiple synovial osteochondromatosis of the knee in a child: A case report

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Received: 02 December 2022
Accepted: 19 January 2023
EPub Ahead of Print: 03 February 2023
Published: 07 February 2023

DOI
10.25259/JMSR_147_2022

Quick Response Code:

ABSTRACT

We report a case of a child who presented with a clinical history of 11 months of progressive pain and swelling in his right knee. Osteochondromatosis is a rare diseas, whose incidence is 1/100,000. It is a synovial proliferative disease associated with cartilage metaplasia resulting in sporadic multiple intra-articular and extra-articular loose bodies. Our focus is to report a rare case of synovial osteochondromatosis in a child successfully treated with arthroscopic surgery. Due to the rarity of the case, we aimed to remind the practicing surgeons of this diagnosis and share our management.

Keywords: Child, Knee, Osteocartilaginous nodules, Synovectomy, Synovial osteochondromatosis

INTRODUCTION

The synovial membrane may suffer from metaplasia and hyperplasia, which can lead to the formation of cartilaginous nodules within the connective tissue of the synovial membrane of the joints, tendon sheaths, tendons, or bursae. These give rise to what is known as synovial osteochondromatosis. It is an infrequent pathology, which is particularly rare in the pediatric population.[1-3]

The incidence is uncertain but is 1/100,000 inhabitants in some publications.[1] There are few publications with involvement in patients with immature skeletons.[1-4] Monoarticular affections are more frequently reported, where the knee is the most compromised joint (60%), followed by the hip, elbow, shoulder, and rarely smaller joints. The disease is more common in males between the third and fifth decades.[3-5]

The symptoms are non-specific, where joint effusion is accompanied by pain, weakness, or limitation of joint motility, all of which can present slowly over months or years.[6] The potential joint destruction due to the erosion produced by the loose bodies formed, through the synovial joint fluid makes its presentation in children an indication for early treatment.[7-9]

Given the suspected diagnosis of synovial osteochondromatosis, a simple radiograph of the joint can be requested, or more sensitive methods such as magnetic resonance imaging (MRI) or computed tomography (CT) scan can be used, which allow the evaluation of non-calcified

How to cite this article: Navarro Vergara AD. Multiple synovial osteochondromatosis of the knee in a child: A case report. J Musculoskelet Surg Res 2023;7:62-6.

nodules. The diagnostic confirmation is dependent on histological evaluation.[4]

The treatment of this pathology consists of removing the nodules and synovectomy; partial or total, by an arthroscopic or open approach,[8-10] giving satisfactory results with the possibility of occasional recurrences. We are reporting a 9-year-old patient with an 7-month history of joint swelling accompanied by pain without a history of joint locking. The final diagnosis was synovial osteochondromatosis of the right knee.

Our objective was to report the case due to the rarity of its presentation, knowing that it was rarely described in the literature and share our management.

CASE REPORT

A 9-year-old female patient consulted our center with symptoms of intermittent swelling in the right knee related to the daily activity for 11 months [Figure 1]. It was accompanied by occasional moderate-intensity pain but no joint locking. She had a fall from height with and a low impact on the right knee that was managed as a soft-tissue contusion with non-steroidal anti-inflammatory drugs orally and topically for 5 days. The mother reported that the knee was swollen for approximately 2–3 weeks after the traumatic event, which subsided spontaneously with rest and reappeared with sports activities. Later, she began to present with periods of occasional activity-related swelling, which subsided with decreased activities. When the episodes became more frequent, the mother consulted the service. On questioning, they denied other systemic symptoms or local infection signs. The patient has been on carbamazepine since the age of 3 years for two episodes of focal epilepsy that no longer occurred after the treatment had started.

Her physical examination revealed a knee joint effusion of moderate intensity, pain on flexion and extension, and decreased joint range of motion (40° of extension and 110° of flexion). She also had a sensation of free bodies in the infrapatellar area anteriorly, without knee locking, which is noteworthy. A simple radiograph showed fragments with a bony density that could be faintly appreciated in the proximal anterior tibia and the popliteal fossa with the knee in semi-flexion [Figure 2].

Given the suspicion of a tumor process, a CT scan with 3D reconstruction was requested, which was reported [Figure 3], with a suspected diagnosis of multiple synovial osteochondromatosis. An MRI scan with gadolinium was performed to exclude other differential diagnoses [Figure 4a-c], where synovitis was observed. Multiple hypointense nodules on T1 and T2 were observed scattered throughout the joint. Chondral and meniscal injuries were ruled out. In addition to synovial osteochondromatosis, fractures tear chronic (mainly from the upper pole of the patella), pathologies that occur with synovitis such as pigmented villonodular synovitis (PVNS), meniscal lesions, tumors such as chondroma and synovial chondrosarcoma, and pathologies that can occur with bodies free as osteochondritis dissecans and osteonecrosis were excluded as possible differential diagnoses.

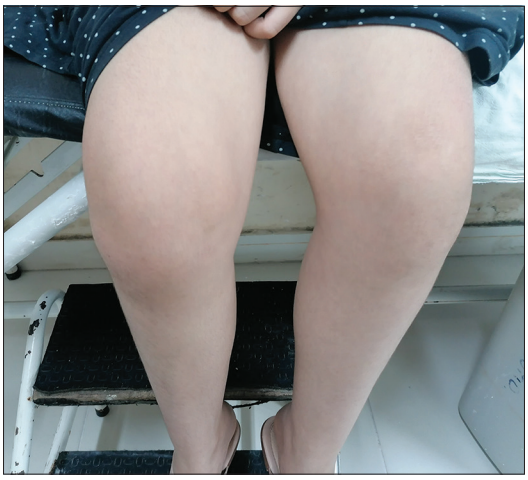

Figure 1: Clinical presentation. Right knee with local swelling compared to the opposite side.

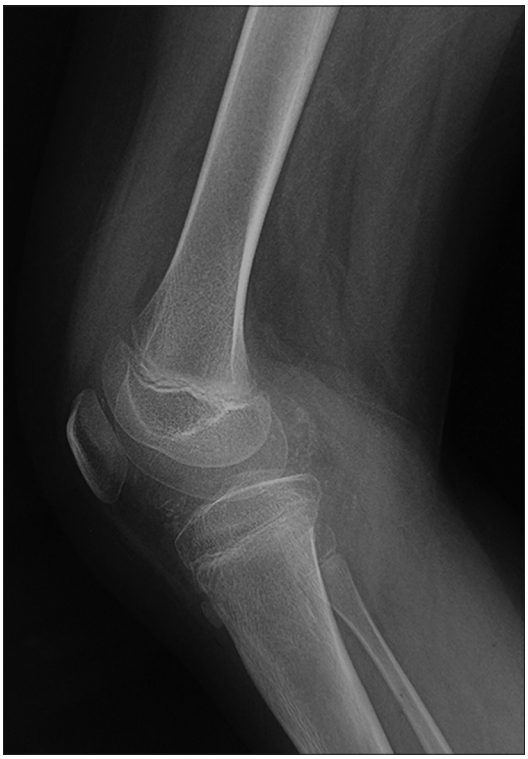

Figure 2: A plain lateral radiograph of the knee shows disseminated bone density images in the anterior and posterior area.

Given the aforementioned findings and the case discussion with specialists with experience in managing similar cases, surgical intervention was decided using arthroscopy[Figure 5a and b], supported by a minimal lateral suprapatellar approach. Excision of multiple fragments was performed [Figure 6]. In addition, synovial samples were taken, which were sent for the pathological study that confirmed the presumptive diagnosis of chondromatous synovitis with the exclusion of a neoplastic process [Figure 7].

In the immediate post-operative period, she was instructed to rest at home, avoid impacts, and use crutches for walking. After 2 weeks, the sutures were removed, and she was allowed to start recreational physical activities. The patient had an excellent recovery, with the restoration of joint range of motion. She resumed her full activities and recreation a month after the procedure.

DISCUSSION

Knee pain in children with painful local swelling opens up a wide range of diagnostic possibilities; some of the causes being devastating pathologies and some others are more innocent.[2-7] Worst scenarios or diseases that require early intervention, such as neoplastic processes or chronic infections, should always be ruled out.[7-10]

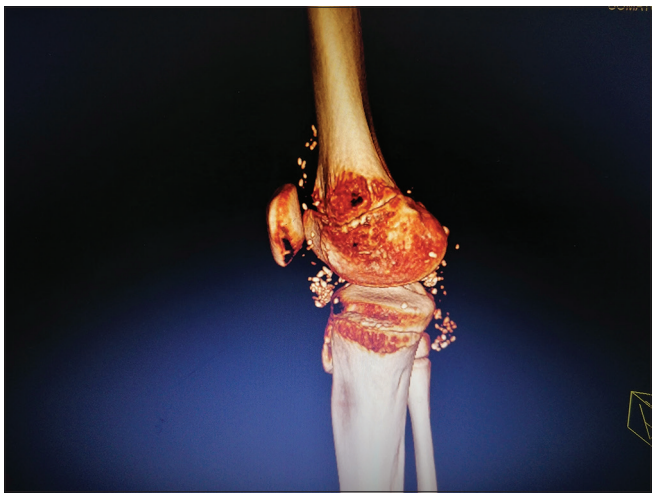

Figure 3: CT with 3D reconstruction shows disseminated knee nodules. The gold standard for diagnosis of synovial osteochondromatosis. CT: Computed tomography.

Two recent publications reporting similar cases made our diagnostic work-up easier and early and helped us define appropriate management.[1,2] Although synovial osteochondromatosis is classified as a benign pathology, it requires early intervention to avoid structural damage due to local irritation from the loose bodies.

There are different reports where synovial osteochondromatosis was classified as secondary since it was related to previous events in the affected area or due to previous systemic pathologies.[5-8] This was excluded in our case. When the etiology is primary, it is characterized by relapsing or recurrent, requiring more than one surgical procedure to resolve the problem.[8,9]

In our case, tomographic images with 3D reconstruction led us to an accurate diagnosis, whereas MRI images were used to rule out malignant pathologies or the existence of other associated lesions.[1-6]

Backed by the opinion of colleagues with experience in managing similar cases and in the literature, we opted for mixed treatment for the excision of the nodules and synovectomy. There are recommendations that support adjuvant radiotherapy and chemotherapy to avoid recurrence in cases classified as refractory or recurrent synovial osteochondromatosis.[8,9] However, the strongest evidence continues to be surgical intervention, either arthroscopically or openly.[8-10]

The extent of the lesion will define the type of excision of the nodules and synovectomy, with the arthroscopic approach being the least aggressive form and that was shown to be less prone to recurrences in the study published almost 20 years ago by Ogilvie-Harris.[7-11] The possibility of recurrence seems to be almost nil when the synovectomy is performed open surgery by Biazzo *et al.*, where some surgeons leave the possibility of a second surgery if the symptoms reoccur.[6-9]

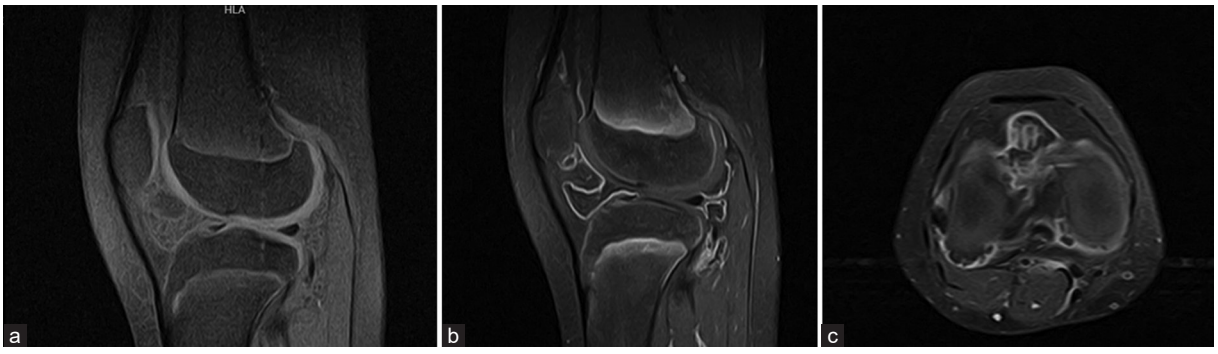

Figure 4: (a-c) Magnetic resonance images with contrast enhancement show that there is no evidence of associated injuries.

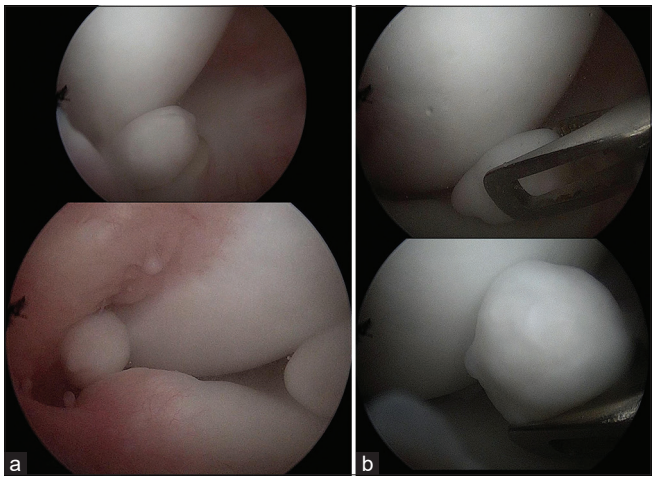

Figure 5: (a and b) Images of arthroscopic portals show free nodules are identified in the joint, with diffuse synovial erythema.

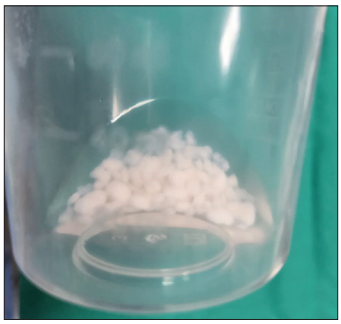

Figure 6: Collection of the multiple resected nodules.

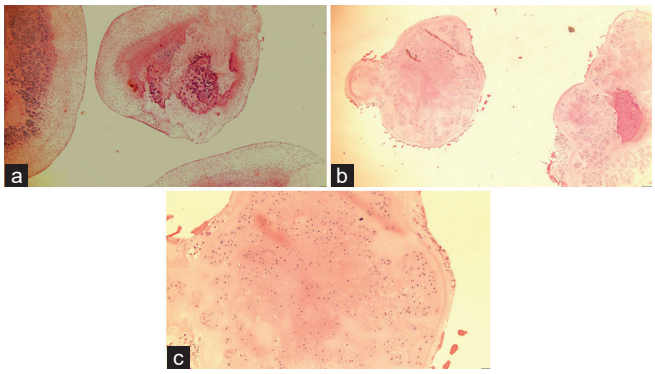

Figure 7: (a-c) Histological section. Hyaline cartilage nodule. Multiple grouped chondrocytes are observed. Minimal nuclear atypia and a slight increase in cellularity. Foci of endochondral ossification are observed.

As the presentation is at an early age, the risk of local chondral destruction or the possibility of malignancy has a greater potential than in adult cases since recurrence can frequently occur throughout the patient's life. Temponi *et al.* reported a rate of up to 5%, given the difficulty in the distinction between recurrence and low-grade chondrosarcoma.[5] For these reasons, patients must be closely monitored by the team on a regular basis, with suspicion of malignancy in the event of sudden onset symptoms, early recurrences, or images suggestive of bone marrow infiltration.

CONCLUSION

Synovial osteochondromatosis in the knee region is a rare pathology. Nevertheless, it should not be left out of the differential diagnosis in patients with swelling, pain, and other symptoms. We recommend surgical management with complete excision of the lesions, including every single free body when the lesion causes symptoms.

AUTHOR'S CONTRIBUTION

The author has critically reviewed and approved the final draft and is responsible for the manuscript's content and similarity index.

ETHICAL APPROVAL

Approved by the Institutional Committee 11.29.2022. The consent of the parents was obtained for the publication of the case. No. HT-CE 01-XI.

DECLARATION OF PATIENT CONSENT

The author certifies that he has obtained all appropriate patient consent forms. In the form, the patient's parent has given his consent for the patient's images and other clinical information to be reported in the journal. The parent understands that the patient's name and initials will not be published, and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed

FINANCIAL SUPPORT AND SPONSORSHIP

This study did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CONFLICTS OF INTEREST

There are no conflicting relationships or activities.

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